Health Information and Quality Authority

An tÚdarás Um Fhaisnéis agus Cáilíocht Sláinte

Health Technology Assessment of Domiciliary Invasive Ventilation for Adults with Spinal Cord Injuries

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Safer Better Care

About the Health Information and Quality Authority

The Health Information and Quality Authority (HIQA) is an independent statutory authority established to promote safety and quality in the provision of health and social care services for the benefit of the health and welfare of the public.

HIQA's mandate to date extends across a wide range of public, private and voluntary sector services. Reporting to the Minister for Health and engaging with the Minister for Children, Equality, Disability, Integration and Youth, HIQA has responsibility for the following:

- **Setting standards for health and social care services** — Developing person-centred standards and guidance, based on evidence and international best practice, for health and social care services in Ireland.

- **Regulating social care services** — The Chief Inspector within HIQA is responsible for registering and inspecting residential services for older people and people with a disability, and children's special care units.

- **Regulating health services —** Regulating medical exposure to ionising radiation.

- **Monitoring services** — Monitoring the safety and quality of health services and children's social services, and investigating as necessary serious concerns about the health and welfare of people who use these services.

- **Health technology assessment** — Evaluating the clinical and cost-effectiveness of health programmes, policies, medicines, medical equipment, diagnostic and surgical techniques, health promotion and protection activities, and providing advice to enable the best use of resources and the best outcomes for people who use our health service.

- **Health information** — Advising on the efficient and secure collection and sharing of health information, setting standards, evaluating information resources and publishing information on the delivery and performance of Ireland's health and social care services.

- **National Care Experience Programme** — Carrying out national service-user experience surveys across a range of health services, in conjunction with the Department of Health and the HSE.

Foreword

Damage to the spinal cord can result in catastrophic and life-threatening injuries, which can lead to the affected person suffering from significant long-term morbidity and requiring life-long care. The higher up the spinal cord that the injury occurs, the more extensive the range of impairments will generally be. Patients with complete C1-C3 level cervical lesions will experience the most severe level of impairment and tend to require a ventilator and tracheostomy to breathe. While the majority of these individuals will eventually be weaned off the ventilator over time, a minority will have lifelong dependency on invasive mechanical ventilation - defined by the requirement for the insertion of an artificial airway, usually tracheostomy, in the larynx region.

As part of the Irish integrated care pathway for patients with spinal cord injuries which has been developed by the National Clinical Programme for Rehabilitation Medicine, it is currently standard practice to send ventilator-dependent patients with spinal cord injuries home with an appropriately funded homecare package when safe to do so. However, discharge planning for these patients remains a huge challenge. As a result, these patients often remain hospitalised for a prolonged period of time, sometimes several years, before being discharged home. These delayed discharges can have a detrimental impact on the individual, their family and carers, management of intensive care unit bed capacity, and the wider healthcare system due to the significant cost of and resources associated with treating these patients in hospital

Work on the health technology assessment was undertaken by an evaluation team in HIQA. A multidisciplinary Expert Advisory Group was convened to advise the evaluation team during the course of the health technology assessment. HIQA would like to thank its evaluation team, the members of the Expert Advisory Group and all who contributed to the preparation of this report.

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Health Information and Quality Authority

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Conflicts of Interest

None reported.

Key Findings and Advice to the Minister for Health and the Health Service Executive

At the request of the Clinical Lead for the National Clinical Programme for Rehabilitation Medicine in the Health Service Executive (HSE), and the Programme Manager for the Spinal Cord System of Care Programme in the National Rehabilitation Hospital (NRH), the Health Information and Quality Authority (HIQA) agreed to undertake a health technology assessment (HTA) on domiciliary invasive ventilation for adults with spinal cord injuries (SCI). Work commenced on this HTA in August 2022. This HTA aimed to assess the organisational, budget impact and resource implications associated with, and the social and ethical issues arising from, the provision of care to ventilator-dependent adults with SCI within their own homes.

The key findings of this HTA, which informed HIQA's advice to the Minister for Health and the HSE, were:

- The range of impairments resulting from SCI are largely determined by the location on the spinal cord in which the lesion occurs, and the completeness of the injury.
- Overall, C1-C4 traumatic SCI are rare in Ireland and numbers fluctuate from year to year. The number of C1-C4 discharges (in patients aged 15 years and older) from the National Spinal Injuries Unit (NSIU) in the Mater Misericordiae University Hospital (MMUH) between 2008 and 2021 ranged from 9 to 27 cases per annum over the 14 year period. Males accounted for 82% (n=185) of C1-C4 SCI discharges from the NSIU during this period. The most common age groups at time of admission were those aged 65 to 74 (22%, n=49) and those aged 15 to 24 year olds (17%, n=39); however, cases in those aged 65 and older represent an increasing proportion of incidence.
- Patients with complete C1-C3 level cervical lesions will experience the most severe level of impairment and tend to require a ventilator and tracheostomy to breathe. Those requiring permanent invasive mechanical ventilation, and whose condition is considered clinically appropriate to be discharged home, will account for a very small proportion of the total number of patients admitted with SCI (estimated from the literature to be approximately 3 to 5% of all individuals with tetraplegia). Currently there are an estimated five to ten adults with SCI in Ireland receiving invasive mechanical ventilation at home.
- Besides respiratory failure, other clinical conditions that impact significantly on patients with high cervical SCI include autonomic dysreflexia, pain,

anxiety/depression, neurogenic bladder and bowel dysfunction and pressure injuries. These individuals require lifelong complex care from a range of specialties.

- The National Clinical Programme for Rehabilitation Medicine has developed an integrated care pathway (extending from pre-hospital care to discharge and lifelong care) for patients with SCI in Ireland, in line with its model of care. However, there are challenges (such as funding and staffing) with the implementation of this care pathway, particularly at the discharge stage for patients requiring domiciliary ventilation, leading to significant delays in discharge to home.
- While domiciliary ventilation has been used and recommended for a range of conditions under specific circumstances (for example, where the patient is medically stable, correct supports are in place, the patient's family is supportive, and other relevant criteria are satisfied), it is important to note that there are some safety concerns associated with the provision of such complex care in the home setting. There are reports in the literature of ventilation failure, hypoxic brain injuries and deaths associated with the use of domiciliary ventilation, though these are rare. In the absence of important risk mitigation factors, such as, a rigorous clinical governance system, proper training, sufficient staffing, resources and supports, there is the potential for serious consequences where care is provided in the home setting. Regardless of the setting in which care is provided for these patients, it is important to note that an element of risk will always be involved given the reliance on a ventilator to stay alive.
- A scoping review of international practice and standards relevant to the provision of permanent domiciliary invasive ventilation in adults with spinal cord injury was undertaken. Seventeen guidance documents were included.
    - In terms of governance, guidance documents discuss the importance of having clear roles and responsibilities for the domiciliary ventilation service. Individuals receiving domiciliary ventilation and their families should be clear about who to contact for different reasons (for example, acute illness, equipment malfunction or patient transfers). There should be a clear understanding among the various clinicians involved as to who is ultimately responsible for which aspects of the individual's care in the community.
    - There is consistency across international guidance documents that 24/7 care, involving at least one trained homecare staff who has demonstrated competency for the tasks required, is essential for these individuals due to their complex healthcare needs. However, there is a lack of consensus on the requirement for all homecare staff to have nursing qualifications.

  - In general, the guidance documents all discuss the importance of appropriate preparation and timing of the transition to the home setting; anticipation of present and future requirements in terms of environment and training of those providing care; and anticipation of disease progression.
  - A comprehensive, holistic and person-centred approach to care, delivered from a well-coordinated interdisciplinary team, which respects the preferences of the individual, is advocated in these guidance documents.
- A budget impact analysis (BIA) was undertaken from the perspective of Irish publicly-funded health and social care system, namely the HSE, to estimate the costs of providing a comprehensive homecare package to a patient with SCI who requires invasive mechanical ventilation, as well as the costs of developing a bespoke training and support role on a national footing.
  - Assuming 196 hours of paid formal care per week (that is, one staff member providing 24/7 care over the course of a week with 28 hours of overlap with a second staff member), the total budget impact for domiciliary ventilation care provision is estimated at €3.4 million over five years per patient. The majority of expenditure is associated with the cost of labour (€2.8 million, 83%).
  - There is substantial uncertainty regarding the incremental budget impact of providing a comprehensive homecare package compared with hospital care, with estimates ranging from €0.58 million less to €1.41 million more per patient over five years. It is therefore unclear whether homecare costs more or less than hospital care for these patients.
  - In the event that a patient is provided with 336 hours of paid formal care per week (that is, one nurse and one healthcare assistant (HCA) each providing 24/7 care), the total budget impact of this level of care provision is estimated at €4.8 million over five years per patient, which is 42% higher than the base case.
  - In scenario analysis, altering the ratio of nursing to HCA care was found to be influential. Relative to the base case, in a scenario where it was assumed that there is 28 hours of nursing care and 168 hours of HCA care per week, the total budget impact is reduced by €0.7 million (21%) over five years.
  - Of note, some patients are currently receiving in excess of 196 hours of paid formal care per week. While a 'typical' package might be well defined in terms of elements, it might not be in terms of intensity, because individual need will vary from patient to patient.

  - The total cost of establishing and implementing a new bespoke role in the community that would provide ongoing training and support to patients, families and homecare workers, is estimated at €0.45 million over a five-year period.
- A conservative estimate of the incidence of SCI patients suitable for domiciliary ventilation is one person every second year. If there is an expectation that one new patient will require this service every second year, and accounting for the bespoke training and support role, then the resulting five-year total budget impact for the HSE overall is estimated to be between €6.7 million and €9.2 million, with the higher estimate reflecting the cost for three consecutive patients who require 336 hours of paid care per week. In contrast, assuming a worst case scenario of two new patients every year, the total cost to the HSE overall is estimated to be between €21.2 million and €29.8 million over five years.
- Substantial organisational barriers to the provision of homecare services for these patients were identified.
  - Currently the funding of homecare packages for these patients with substantial care needs is a challenge as these are funded from various different sources including local budgets; the funding of high dependency patients may overwhelm local budgets in particular.
  - Other barriers include the limited availability of highly trained staff in the community, the lack of a statutory homecare scheme, the lack of an explicit care pathway in the community and the significant out-of-pocket costs for patients and families.
  - The role of the homecare worker is complex and challenging, due to balancing the needs of managing the patient and respecting the privacy of the family. This environment can lead to high staff turnover, creating risks for achieving sustained high quality care.
- Appropriately adapted housing is an essential prerequisite for the delivery of homecare services for these patients. However, there are limits to the funding available for housing modifications, and this funding is means tested. In addition, some patients do not own their own house and so provision of alternative accommodation by local authorities may be necessary. A review of the full suite of Housing Adaptation Grants for Older People and People with a Disability is currently underway by the Department of Housing, Local Government and Heritage.
- With regards to the national delivery of care for these patients, a range of micro-level (patient and family interaction) organisational factors for consideration include: development of explicit discharge criteria and protocols; a more flexible process for obtaining housing adaptation funding and or provision of housing;

greater access to step down facilities and respite services; and provision of recommended supports and services. For some patients, where discharge home may not be feasible, residential care facilities may offer an alternative setting for their long term care, and this option should be explored.

- Meso-level (healthcare organisation and the community) organisational factors for consideration include: development of a centralised budget with, where necessary, an associated national tender framework; development of a model of complex discharge planning similar to that used for paediatric services; development of an explicit care pathway within the community; care coordination; the model of homecare provision; development of a bespoke training and support role in the community; involving other National Clinical Programmes; and greater involvement of clinical engineers with respect to planning the maintenance of equipment for these patients in the community.
- Creation of a nationwide ring-fenced budget could prevent local budgets being depleted, allowing funding for other important community services to be preserved. It is important to remember that it is already standard practice to send these patients home; however, provision of a systematic nationwide service may enable such a transition to happen in a quicker and more coordinated fashion. Development of a clinical pathway with a national system of governance and funding may also indirectly help address the unmet needs of other patient groups who are seeking to receive care in their home.
- In terms of the macro-level (health policy) factors, there is currently a policy initiative for the safe discharge home of these patients with an appropriately funded package of care in the context of an impending statutory homecare scheme.
- Living at home on a ventilator with high cervical SCI is associated with substantial challenges for both the patient and their family. The complications arising from the injuries are profound and can have a hugely negative impact on patients requiring round-the-clock care from homecare staff. Families have expressed their strong desire to provide the best possible care for their loved ones, but often to their own detriment (physically, emotionally and financially). Carer burden is particularly problematic among family carers, given the high level of dependency.
- However, these patients have also demonstrated resilience in adapting to their ‘new normal’ and a willingness to live a fulfilling life despite the obstacles. Provision of the required supports and services for these patients and their families, on an ongoing basis, may alleviate some of the hardship and make the care provision more sustainable. The importance of a good relationship with family and friends is considered an important factor that motivates patients and adds value to their lived experience.

- In terms of respect for autonomy, provision of a national delivery of care for these patients should be viewed as supportive of patient autonomy where a clear preference to move home is expressed. However, it is crucial that patients and families are fully informed of the benefits and risks of domiciliary ventilation, and that a true choice of options is provided to the patient that respects the preference of the individual.
- From an ethical point of view, consideration needs to be given to the potential for the benefits of shifting such complex care from a hospital to a community setting to be outweighed by its associated harms. Potential benefits include that it would respect patients' preferences to receive care at home, would free up scarce intensive care unit (ICU)/high dependency unit (HDU) beds, may improve equity of access for patients given the current geographical disparities in funding and service provision and may improve patients' quality of life. However, this needs to be weighed against the increased safety concerns associated with providing complex care at home, the substantial burden on families, and the opportunity cost to other patient groups.

Arising from the findings of this HTA, HIQA's advice to the Minister for Health and the Health Service Executive is as follows:

- Some patients with complete C1-C3 level cervical lesions have a lifelong requirement for mechanical invasive ventilation. It is estimated that up to two new patients with SCI may require permanent ventilation in Ireland each year, though in some years there are no new patients. Internationally, domiciliary (at-home) ventilation is considered the standard of care for ventilator-dependent patients with SCI where clinically appropriate.
- While it is currently standard practice in Ireland to provide domiciliary ventilation for these patients, there is no formalised approach for discharge and ongoing care in the community. This combined with the fragmented funding system involving multiple budget holders results in delayed discharges, which has substantial implications for patient and family quality of life and for health service provision.
- An international review found that while standards and practice vary with respect to the qualification of staff providing care, common features of a domiciliary ventilation service include:
    - a system of governance with clear roles and responsibilities
    - clinical guidance in relation to the management of patients' complex healthcare needs
    - defined processes for discharge, communication and ongoing monitoring
    - dedicated funding for the required equipment and staffing

  - a requirement for 24/7 care by trained homecare staff who have demonstrated competency for the tasks required
  - education and training programmes for patients, families and homecare staff
  - processes for managing ethical and safeguarding issues, including advanced care planning.
- With respect to costs:
  - The total budget impact for domiciliary ventilation care provision, per patient, is estimated at €3.4 million over five years. However, this could be as high as €4.8 million per patient depending on the number and grade of homecare staff involved. It is unclear whether care at home would cost more or less than hospital care for these patients; however, domiciliary ventilation may alleviate some of the challenges currently experienced by patients and families, and would free up ICU/HDU bed capacity.
  - The total cost of establishing and implementing a new bespoke role in the community that would provide ongoing training and support to patients, families and homecare workers, is estimated at €0.45 million over a five-year period.
- With respect to any decision to developing a national provision of service, consideration should be given to:
  - an overarching clinical governance framework that takes into account the substantial requirements for complex discharge planning and well-coordinated care that may involve multiple government departments, agencies and clinical programmes
  - development of a national clinical pathway for ventilator-dependent SCI patients that extends into the community, that includes specification of roles and responsibilities for service providers
  - a centralised budget for provision of care
  - a national training and support role to support the delivery of a safe, effective and quality assured service, and to improve the resilience and continuity of care
  - how ethical aspects (for example, balance of benefits and harms for patients and families, best use of available resources) are managed in decision-making
  - exploring the barriers that currently exist to providing domiciliary ventilation in long-term care facilities and how these might be addressed

- a system for clinical monitoring, audit and evaluation to ensure safer, better care.

Executive summary

Health technology assessment (HTA) is a multidisciplinary process that summarises information about the medical, social, economic, and ethical issues related to the use of a health technology. A HTA is performed in a systematic, transparent, unbiased, and robust manner with the intention of supporting evidence-based decision-making regarding the optimal use of resources in health and social care services.

This report summarises the findings of a HTA on the provision of care to ventilator-dependent adults with spinal cord injuries (SCI) within their own home.

Background

The range of impairments resulting from SCI are largely determined by the location on the spinal cord in which the lesion occurs, and the completeness of the injury. Patients with complete C1-C3 level cervical lesions will experience the most severe level of impairment and tend to require a ventilator and tracheostomy to breathe. While the majority of these individuals will eventually be weaned off the ventilator over time, a minority will have lifelong dependency on invasive mechanical ventilation - defined by the requirement for the insertion of an artificial airway, usually tracheostomy in the larynx region.

An integrated care pathway for patients with SCI has been developed in Ireland by the National Clinical Programme for Rehabilitation Medicine, in line with its model of care. Although the National Rehabilitation Hospital (NRH) can admit and manage ventilated patients since 2019 as part of this pathway, discharge planning for these complex cases remains a huge challenge for several reasons, including the current fragmented funding model for community services and the limited availability of suitably trained homecare staff in the community. There may also be regional variation in the availability of the necessary resources to enable the homecare of these individuals. As a result, these individuals often remain hospitalised for a prolonged period of time, often several years, before being discharged home. These delayed discharges can have a detrimental impact on the individual, their family, and management of intensive care unit (ICU) bed capacity in the NRH and the wider healthcare system.

At the request of the Clinical Lead for the National Clinical Programme for Rehabilitation Medicine in the Health Service Executive (HSE), and the Programme Manager for the Spinal Cord System of Care Programme in the NRH, HIQA agreed to undertake a HTA on domiciliary invasive ventilation for adults with SCI. Work commenced on this HTA in August 2022. This HTA aimed to assess the organisational, budget impact and resource implications associated with, and the social and ethical issues arising from, the provision of care to ventilator-dependent adults with SCI within their own home.

Burden of disease and epidemiology

In general, permanent ventilator assistance is required for complete SCI at C1-C3 due to diaphragmatic paralysis. Patients with C3-C4 injuries experience diaphragmatic dysfunction, but may retain partial ventilatory function. Weaning from a ventilator may be achieved in patients with C3 injuries, while injuries of C4 and below are usually associated with successful weaning, in the absence of other complications. Apart from completeness and level of injury, other barriers to weaning include older age, smoking history, active pneumonia, the severity of impaired consciousness, high level injury severity score, underlying cardiorespiratory diseases and other complicating comorbid conditions.

Overall, C1-C4 traumatic SCI are rare in Ireland and numbers fluctuate from year to year. Traumatic SCI are predominantly a male phenomenon, accounting for 82% (n=185) of C1-C4 SCI discharges from the National Spinal Injuries Unit (NSIU) in the Mater Misericordiae University Hospital (MMUH) between 2008 and 2021. Incidence of C1-C4 discharges from the NSIU fluctuated from 2.7 to 3.6 cases per million population over the 14 year period (with the highest recorded in 2013 at 5.9 cases per million and the lowest in 2010 at 2 cases per million). The most common age groups at time of admission to the NSIU were those aged 65-74 (22%, n=49) and those aged 15-24 year olds (17%, n=39). Cases in those aged 65 and older represent an increasing proportion of incidence. However, establishing the causes of apparent trends is challenging as the numbers of cases are small. Falls and road traffic accidents were the leading causes of traumatic SCI in Ireland between 2008 and 2022. Data from Ireland and internationally indicate that the incidence of SCI in older age groups is increasing, largely driven by falls in these older individuals.

The key factors associated with increased risk of mortality and morbidity in patients with traumatic SCI are a higher level of injury, the completeness of the injury, older age, ventilator dependency, and the presence of multi-system trauma. There are many complications associated with cervical SCI. Among these, specific areas of concern for management include venous thromboembolism (VTE), neurogenic and spinal shock, SCI-induced bradycardia, respiratory complications, acute autonomic dysreflexia, pressure injuries, neurogenic bowel and bladder dysfunction, mental health issues, and pain. Pneumonia and respiratory failure remain a significant cause of death in patients with SCI.

Median life expectancy for patients with SCI reportedly ranges from 2.8 to 43 years from time of injury, with survival time related to age at time of injury. In addition to age, factors that influence life expectancy include sex, level and completeness of injury, and ventilator dependency. Ventilator dependency was the factor associated with the lowest life expectancy, reportedly reducing life expectancy by approximately 75% compared with the general population.

Patients with C1-C3 SCI requiring permanent mechanical ventilation, and whose condition is considered clinically appropriate to be discharged home, will account for a very small proportion of the total number of patients admitted with SCI (estimated from the literature to be approximately 3 to 5% of all individuals with tetraplegia). Between 2008 and 2022, at least eight patients who required ventilation assistance were discharged from the NSIU. It is estimated that up to two patients with SCI annually are anticipated to require permanent ventilation in Ireland, though in some years there are no new patients. It is estimated that there are currently between five and ten adults with SCI in Ireland receiving invasive mechanical ventilation at home.

Description of the technology and outcomes

Traumatic injury to the cervical spine is associated with variable degrees of respiratory dysfunction, dependent on the level and completeness of injury. There are two main types of mechanical ventilation – invasive and non-invasive. Invasive ventilation is generally defined by the requirement for the insertion of an artificial airway, usually tracheostomy in the larynx region, while non-invasive ventilation (NIV) generally involves applying tightly fitting face or nasal masks without any artificial airway. Invasive mechanical ventilation rather than NIV is generally necessary for patients with complete high cervical SCI, particularly during the initial management phase, due to the lack of neural control of breathing.

An alternative intervention to mechanical ventilation is 'phrenic nerve stimulation', also called 'diaphragm pacing', which involves the surgical implantation of an electrode around the phrenic nerve(s), connected to a radio-frequency receiver usually implanted in the chest wall. Phrenic nerve stimulation provides electrical stimulation of the phrenic nerve which results in diaphragmatic contraction.

Secretion management is crucial in patients with SCI who are invasively ventilated; this is due to the inability of these individuals to clear secretions on their own. Airway clearance is important in order to prevent atelectasis (that is, a complete or partial collapse of the lung) and respiratory infections. Effective secretion management can be achieved through the use of various techniques (for example, air stacking and glossopharyngeal breathing) and equipment (for example, mechanical insufflation / exsufflation (MIE) device).

Besides respiratory failure, other clinical conditions that impact significantly on patients with high cervical SCI include autonomic dysreflexia, pain, anxiety/depression, neurogenic bladder and bowel dysfunction and pressure injuries. Providing appropriate bladder, bowel and skin care to these individuals is complex, lifelong and requires a range of health professionals.

There are a range of different medical conditions that may lead to the requirement for long-term ventilation, such as amyotrophic lateral sclerosis (ALS) which is a sub-set of motor neurone disease, chronic obstructive pulmonary disease (COPD), myasthenia gravis and SCI. Provision of mechanical ventilation services at home (or domiciliary ventilation) is the preferred option for many individuals who require ventilation for extended periods of time. The prevalence of domiciliary ventilation varies across and even within jurisdictions but overall the prevalence is generally low (population-based estimates in the literature range from approximately 0 to 30 per 100,000 population). International literature estimates suggest that patients with SCI account for a small proportion of all domiciliary ventilation service users, ranging from 2% to 19.9%, with higher proportions among invasively ventilated cohorts. Prevalence of domiciliary ventilation is increasing internationally, driven largely by the increased use of NIV. The use of invasive ventilation in the domiciliary setting is less common, but is more prevalent in patients with SCI.

Although there are some data to support the safe and effective use of domiciliary invasive ventilation in other populations, data specific to the SCI population in particular are very limited. While domiciliary ventilation has been used and recommended for a range of conditions under specific circumstances, it is important to note that there are some safety concerns associated with the provision of such complex care in the home setting (for example, ventilation failure). However, from the patient’s perspective, there is often a preference to return home once the right supports and equipment are in place, even if this does entail some added risk.

Scoping review of international practice and standards

A scoping review of international practice and standards relevant to the provision of permanent domiciliary invasive ventilation in adults with SCI was undertaken. The search included guidelines, position papers, protocols and standards (collectively referred to as ‘guidance documents’) that related to, or were applicable to a population with SCI.

Seventeen guidance documents from Australia, Canada, Germany, Italy, New Zealand, Poland, Switzerland, the Netherlands, the UK, and the US, were identified for inclusion. Ten contain SCI-specific guidance regarding domiciliary invasive ventilation while seven contain more general, non-condition specific guidance.

In relation to domiciliary invasive ventilation guidance, detailed recommendations are provided for the following topics:

- discharge process
- communication and monitoring post-discharge
- equipment
- staffing.

Patients with high cervical cord injuries generally have multiple, complex healthcare needs, often as a direct or indirect consequence of the SCI. Recommendations are provided in relation to the management of the following issues:

- respiratory complications
- bladder complications
- bowel complications
- skin integrity
- sexual health
- venous thromboembolism
- pain
- autonomic dysreflexia
- emotional wellbeing, mental health and substance abuse
- cardiometabolic disease and nutrition
- bone health
- palliative care.

In relation to broader issues of SCI care pathways and supports, guidance is issued in relation to:

- education and training
- governance structures and care pathways
- ethics and safeguarding.

In terms of governance, guidance documents discuss the importance of having clear roles and responsibilities for the domiciliary ventilation service. Individuals receiving domiciliary ventilation and their families should be clear about who to contact for different reasons (for example, acute illness, equipment malfunction or patient transfers). There should be a clear understanding among the various clinicians involved as to who is responsible for which aspects of the individual's care in the community.

There is consistency across guidance documents that 24/7 care, involving at least one trained homecare staff who has demonstrated competency for the tasks required, is essential for these individuals due to their complex healthcare needs. However, there is a lack of consensus on the requirement for nursing qualifications in the provision of care to these individuals.

In general, the guidance documents all discuss the importance of appropriate preparation and timing of the transition to the home setting; anticipation of present and future requirements in terms of environment and training of those providing care; and anticipation of disease progression. A comprehensive, holistic and person-centred approach to care, delivered from a well-coordinated interdisciplinary team, which respects the preferences of the individual, is advocated in these guidance documents. While international standards and practice in this area may vary, there are many common recommendations that may be useful to inform a national provision of service in Ireland.

Budget impact analysis

A budget impact analysis (BIA) was undertaken to estimate the costs of providing a comprehensive homecare package to a patient with SCI who requires invasive mechanical ventilation, as well as the costs of developing a bespoke trainer and support role on a national footing. The analysis adopted the perspective of the Irish publicly-funded health and social care system, namely the HSE. Only direct medical costs to the HSE were estimated. The BIA projected costs over a five-year time horizon consistent with national guidelines. A 'typical' HSE funded homecare package was costed based on international guidance and current practice with input from Expert Advisory Group (EAG) members. For the purpose of this BIA, the comparator was the patient remaining in hospital for five years instead of being discharged home when clinically suitable to do so. Four different costing methods were used to estimate the hospital care costs in the comparator group.

Costs included inpatient care, equipment and consumables, medication, labour, healthcare service use, and trainer and training costs. Equipment and consumable requirements and costs were obtained through discussions with clinical experts, HSE managers and HSE procurement staff who had oversight of national contracts, invoices and quotes, and were able to provide estimates. Where cost estimates were unavailable, best guess estimates were provided by clinical experts.

Assuming 196 hours of paid formal care per week (that is, one staff member providing 24/7 care over the course of a week with 28 hours of overlap with a second staff member), the total budget impact for domiciliary ventilation care provision was estimated at €3.4 million over five years per patient. The majority of expenditure over a five-year time horizon was associated with the cost of labour (€2.8 million, 83%). Assuming that a patient is provided with 336 hours of paid formal care per week (that is, one nurse and one HCA each providing 24/7 care), over a five-year time horizon, the total budget impact of this level of care provision was estimated at €4.8 million per patient, which is 42% higher than the base case. It is important to note that while the 'typical' homecare package may be tightly defined in terms of elements, it might not be in terms of intensity, because individual need will vary from patient to patient. The total cost of establishing and implementing a new bespoke role in the community that would provide ongoing training and support to patients, families and homecare workers, was estimated at €0.45 million over a five-year period.

There was substantial uncertainty regarding the five-year incremental budget impact of providing a comprehensive homecare package compared with hospital care for patients, with estimates ranging from €0.58 million less to €1.41 million more in the base case analysis. It is therefore unclear whether homecare would cost more or less than hospital care for these patients. Uncertainty relating to nursing costs was found to contribute most to uncertainty in the total budget impact analysis (lower bound: €2.9 million, upper bound: €3.9 million). Uncertainty in this parameter was associated with 30% variation in the total budget impact over a five-year time horizon. Given the substantial contribution of labour to the total costs of a homecare package, scenario analysis was undertaken to examine the impact on the five-year total budget impact of altering the ratio of nursing to HCA care. As the ratio of nursing care to HCA decreases the total budget impact also decreases. Relative to the base case, in a scenario where it was assumed that there was 28 hours of nursing care and 168 hours of HCA care per week, the total budget impact was reduced by €0.7 million (21%) over five years.

If there is an expectation that one new patient will require this service every second year, and accounting for the bespoke training and support role, then the resulting five-year total budget impact for the HSE overall is estimated to be between €6.7 million and €9.2 million, with the higher estimate reflecting the cost for three consecutive patients who each require 336 hours of paid care per week. In contrast, assuming a worst case scenario of two new patients every year, the total cost to the HSE overall is estimated to be between €21.2 million and €29.8 million over five years.

There are other substantial costs that fall on patients and families, as well as other government departments that were not considered as part of this BIA. For example, there are large upfront costs such as housing and car adaptations that, while heavily subsidised by the State, still incur substantial out-of-pocket expenses for patients and families. The uncertainty surrounding medical card approval is another source of worry for many patients and families.

A limitation of this BIA was the high degree of uncertainty in relation to the estimated hospital care costs. In the protocol for this HTA, it was planned that the cost of domiciliary invasive ventilation would be compared with the cost of long-term invasive ventilation in both hospital and residential care settings. However, given that no residential units in Ireland currently accept ventilated SCI patients and hence there is a lack of available data, the evaluation team was unable to provide cost estimates for the provision of care in residential care settings. It was the expert opinion of the EAG that it is more challenging to find a suitable residential care placement than it is to send the patient home, given that the care requirements far exceed what is usual in the vast majority of residential care settings, and that this is an important gap in service.

Organisational considerations

An assessment was undertaken of the necessary organisational changes associated with the provision of domiciliary ventilation to patients with SCI on a national footing. While it is already standard practice to discharge these patients home as soon as it is safe to do so, there are substantial organisational barriers to the provision of homecare services for these patients:

- Currently the funding of homecare packages for these patients with substantial care needs is a challenge as these are funded from various different sources including local budgets; the funding of high dependency patients may overwhelm local budgets in particular.
- Other barriers include the limited availability of highly trained staff in the community, the lack of a statutory homecare scheme, the lack of an explicit care pathway in the community and the significant out-of-pocket costs for patients and families.

Appropriately adapted housing is an essential prerequisite for the delivery of homecare services for these patients. However, there are limits to the funding available for housing modifications, and this funding is means tested. In addition, some patients do not own their own house and so provision of alternative accommodation by local authorities may be necessary. A review of the full suite of Housing Adaptation Grants for Older People and People with a Disability is currently underway by the Department of Housing, Local Government and Heritage.

The organisational considerations for this HTA are discussed at three different levels of the healthcare system (micro-, meso- and macro-level). The micro-level factors relate primarily to patient and family interactions; the meso-level relate to healthcare organisation and the community; and macro-level relate to health policy.

Micro-level organisational factors for consideration include:

- development of explicit discharge criteria and protocols to assist with the complex discharge process and so that patients and families can have realistic expectations for the future

- a more flexible process for obtaining housing adaptation funding that can enable families to carry out necessary renovations, provision of housing for those without their own home, and greater access to step down units to facilitate the transition home and respite services to provide relief for carers and families
- provision of recommended supports and services to enable the patient to engage in their communities to the fullest possible extent
- greater access to ongoing face-to-face supports for patients and families in the community.

Meso-level organisational factors for consideration include:

- development of a centralised national systematic, dedicated and integrated budget with an associated national tender framework to cover personal care, nursing care and equipment; this would likely alleviate the administrative burden and inequity of the system, and expedite the approval of a fully funded homecare package for these patients
- development of a model of complex discharge planning similar to that used for paediatric services (as recommended by the HSE Integrated Care Pathway for the Management of SCI)
- development of an explicit care pathway within the community for these patients, with defined involvement and clear roles and responsibilities for all involved parties, including processes for the transfer of ventilator-dependent children with SCI into adult services.
- care coordination, in the context of healthcare staffing issues, to ensure that informal and formal supports, from all required disciplines, work effectively together to ensure the best outcomes for the patient with SCI
- the model of homecare provision, and, specifically, whether healthcare assistants providing care with nursing oversight is feasible and appropriate, and how competency can be assured in those providing care; substituting 24/7 nursing care with 24/7 trained HCA care with appropriate nursing oversight would reduce the overall cost of a care package for these patients and would likely increase the availability of staff. However, it is critical that any potential savings and increased access to homecare services do not compromise patient safety.
- development of a bespoke training and support role in the community to provide quality assurance of domiciliary invasive ventilation services
- involving other National Clinical Programmes such as Respiratory Medicine in further developing the care pathway into the community

- involving clinical engineers with respect to the maintenance of equipment for these patients in the community, and having a clear understanding of the governance of these equipment.

In terms of the macro-level factors, there is currently a policy initiative for the safe discharge home of these patients with an appropriately funded package of care in the context of an impending statutory homecare scheme. It is important to consider that development of a clinical pathway with a national system of governance and funding may also indirectly help address the unmet needs of other patient groups (for example, younger people with disabilities residing in nursing homes who wish to transition home).

A bespoke national training and support nurse was costed as part of the BIA. It is possible that such a role could be developed to provide ongoing support and information to patients and families, and provide training to homecare staff, ensuring that they are competent to provide the level of care required for these patients.

Should a decision be made by the HSE to implement a national, systematic, dedicated and integrated budget with an associated national tender framework for the provision of homecare to invasively ventilated patients with SCI, this will require substantial organisational cooperation across multiple agencies and programmes.

Patient, social and ethical considerations

An examination was undertaken of the potential patient, social and ethical considerations relating to the provision of domiciliary invasive ventilation services for adults with SCI. The considerations outlined are framed both in the context of the experiences of the patient and their family, and also through an ethical lens.

Living at home on a ventilator with high cervical SCI is associated with substantial challenges for both the patient and their family. Patients have expressed how these life-changing injuries are catastrophic and can often require a prolonged period of adjustment. The complications arising from the injuries are profound and can have a hugely negative impact on patients requiring around-the-clock care from homecare staff. Before the patient can be discharged with a home care package, the home environment must be adapted to meet the medical and accessibility needs of the patient, which can be a considerable financial burden to patients and their families. Fundraising and the charitable support of the wider community has been noted to be an important source of financing for affected families.

Families have expressed their strong desire to provide the best possible care for their loved ones, but often to their own detriment (physically, emotionally and financially). Carer burden is particularly problematic among these family carers, given the high level of dependency. Greater provision of paid care, professional support, and respite care have been recommended in the literature to alleviate carer burden and make care provision more sustainable.

The role of the homecare worker in the patient's home is complex and challenging. Homecare workers have a difficult balancing act of managing the intricate care needs of the patient while respecting the privacy of the family, which can sometimes lead to tensions when there are conflicting goals. Good communication between the patient, family and homecare worker is important for establishing trust. Issues such as high staff turnover and training insufficiencies represent serious risks to achieving sustained high quality care for these patients. Patients need ongoing support from competent homecare workers with an understanding and knowledge of the issues related to living with a spinal cord injury and requiring ventilator care.

However, these patients have also demonstrated resilience in adapting to their 'new normal' and a willingness to live a fulfilling life despite the obstacles. Provision of the required supports and services for these patients and their families, on an ongoing basis, may alleviate some of the hardship and make the care provision more sustainable. The importance of a good relationship with family and friends is considered an important factor that motivates patients and adds value to their lived experience.

The four principles approach to biomedical ethics (otherwise known as principlism) was used as a framework for the ethical analysis: respect for autonomy (that is, respecting the decision-making capacities of an autonomous person); non-maleficence (that is, avoidance of harm), beneficence (that is, providing benefits and balancing benefits against risks and costs); and justice (that is, fairness in the distribution of benefits, risks and costs).

In terms of respect for autonomy, provision of a national delivery of care for these patients should be viewed as supportive of patient autonomy where a clear preference to move home is expressed. However, it is crucial that patients and families are fully informed of the benefits and risks of domiciliary ventilation, and that a true choice of options is provided to the patient that respects the preference of the individual.

In terms of the benefit-harm balance, consideration needs to be given to the potential for the benefits of this national provision of care to be outweighed by its associated harms. Some of the key benefits are that it would respect patients' preferences to receive care at home, would likely free up scarce ICU/HDU beds and may improve patients' quality of life. However, this needs to be weighed against the increased safety concerns associated with providing complex care at home, the substantial burden on families, and the opportunity cost to other patient groups.

From the perspective of justice and equity, provision of a systematic nationwide service may improve equity of access for these patients given the current geographical disparities in funding and service provision. Creation of a nationwide ring-fenced budget could prevent local budgets being depleted, allowing funding for other important community services to be preserved. However, it is associated with a significant opportunity cost given the substantial nursing requirements. It could also be argued that provision of a dedicated nationwide service exclusively to invasively ventilated patients with C1-C3 SCI may not be equitable to other patient groups, who may have the potential to benefit from access to this service.

Conclusion

High cervical SCI requiring permanent mechanical invasive ventilation are rare but have substantial consequences for patients, families and health services. The results of this assessment support the development of a national clinical pathway for this patient cohort that extends into the community. This pathway should have an overarching clinical governance framework, be funded through a centralised budget, and would benefit from a national training and support role. This new clinical pathway, would potentially alleviate some of the challenges experienced by patients and families, streamline the process for securing homecare packages, and increase efficiencies in the health system through freeing up scarce ICU/HDU beds. Development of a bespoke training and support role could aid quality assurance of domiciliary invasive ventilation services nationally through the ongoing provision of competency-based training. It is important that the clinical pathway considers the importance of appropriate preparation and timing of the transition to the home setting; anticipation of present and future requirements in terms of environment and training of those providing care; and anticipation of disease progression. A comprehensive, holistic and person-centred approach to care, delivered from a well-coordinated interdisciplinary team, which respects the preferences of the individual, is critical for its success. It is crucial that a clinical monitoring, audit and evaluation plan for continuous improvement of the clinical pathway is developed. Patient, social and ethical aspects should also be carefully considered during decision-making and accounted for during subsequent implementation.

Plain language summary

Injuries to the spinal cord can be very different depending on the part of the spine that is damaged. Severe injuries such as those at or above the lower part of the neck may mean the person needs a machine (ventilator) to allow them to breathe. Sometimes the patient can return home on ventilator support, where they need 24/7 care. This is called domiciliary ventilation. At the moment the organisation of domiciliary ventilation is difficult. The Health Information and Quality Authority (HIQA) was asked to look at how the HSE can improve the delivery of care to these patients.

There are two main types of ventilation, invasive and non-invasive. Invasive ventilation usually involves a tube being put into someone's throat, which carries air to and from their lungs; this tube is connected to the breathing machine (ventilator). Non-invasive ventilation usually uses a face mask to supply air. For patients with spinal cord injuries at or above the level of the lower neck, invasive ventilation is usually needed. For some of these patients, invasive ventilation is needed round-the-clock and for the rest of their lives. While a ventilator will help the patient to breathe, they will also need a lot of medical care for other reasons, such as managing pain, toileting, preventing blood clots and taking care of the skin. As these patients cannot move their arms or legs by themselves, they will also need help with many other activities in their daily lives.

Spinal cord injuries that occur at or above the lower part of the neck, while rare, can affect anyone, but in Ireland men make up most of these cases. As care needs can differ, it is not always possible for patients that require ventilation on an ongoing basis to be cared for at home. There are fewer than 10 adults in Ireland with spinal cord injuries receiving domiciliary ventilation at the moment.

As part of our assessment, we looked to see if other countries had any guidelines for domiciliary ventilation. We found 17 guidelines altogether, from Australia, Canada, Germany, Italy, New Zealand, Poland, Switzerland, the Netherlands, the UK, and the US. These guidelines gave advice about aspects like sending a patient home from the hospital, looking after the patient when they got home, and the equipment and staff that would be needed. The main thing that all of the guidelines agreed on was that there should be at least one trained person caring for the patient 24/7.

We then looked at the cost of providing domiciliary ventilation in Ireland. We found that this type of care costs about €3.4 million over five years for each patient; however costs may differ depending on how much support the individual patient needs. It is therefore unclear whether managing patients at home is more or less costly than care in a hospital.

We looked at ways in which domiciliary ventilation in Ireland for spinal cord injury patients could be better coordinated. Suggestions for the HSE to consider include development of a care pathway for sending patients home, so that there is a better understanding of everyone's roles and responsibilities. A clear governance system would help coordination between the many different agencies and care providers working across hospital and community settings. A change to the system of funding may reduce delays arising from having multiple budget holders. A new national support role may also be needed to help provide and coordinate training to homecare staff and to support patients and families. Alternative accommodation such as nursing homes may be needed for some patients.

Lastly, we looked at the ethical and social issues that might come up. We found that there are a lot of challenges for the patient and their family when the patient is living at home on a ventilator. It can take a long time to adjust to life after this type of injury and complications can be very harmful and distressing. It is important to respect the wishes of the patient, but it is also important to make sure that the patient is safe and that domiciliary ventilation is the best way to care for them. We found that there can be a big cost for the family if they need to make changes to their house, buy an adapted vehicle, or if they need to give up their job to enable the patient to live at home. If the patient cannot get a medical card then they might also have to buy some expensive equipment themselves. Some family members who want to give the best possible care might be harming their own physical and emotional health to do this. So, while domiciliary ventilation might help to free up beds in hospital intensive care unit settings, it could pass the burden to the family instead.

Currently, the limits of local budgets mean that some people with spinal cord injuries cannot be cared for at home, or it can take a long time to organise the funding. Access may be improved if there is a central budget for this service, as it may speed up the process and make things fairer for patients across the country. However, some other patient groups with a lot of care needs who might also like to be cared for at home might find this unfair if their needs are not also provided for. As domiciliary ventilation may be very costly, the HSE needs to find the right balance between allowing patients the choice as to where they receive their care and ensuring that this does not impact access to services for other patients.

List of abbreviations used in this report

AARC	American Association for Respiratory Care
ABF	activity based funding
AC	alternating current
ACP	advanced care planning
AHCD	advanced health care directives
AHRQ	Agency for Healthcare Research and Quality
AIS	ASIA Impairment Scale
ALS	amyotrophic lateral sclerosis
ASIA	The American Spinal Injury Association
ATS	American Thoracic Society
ATP	adenosine triphosphate
BASCIS	British Association of SCI Specialists
BIA	Budget Impact Analysis
BiPAP	Bi-level positive airway pressure
BL	bilateral
BMJ	British Medical Journal
BSFC	burden scale for family caregivers
Can SCIP	Canadian Spinal Cord Injury Practice
CEO	Chief Executive Officer
CHNs	community healthcare networks
CHOs	community healthcare organisations
CI	confidence interval
CIT	Community Intervention Team
COPD	chronic obstructive pulmonary disease
CPAP	continuous positive airway pressure
DC	direct current
DNA	Deoxyribonucleic Acid
DRG	diagnosis related group
DSD	detrusor sphincter dyssynergia
EAG	Expert Advisory Group
ECC	enhanced community care
ENT	ear, nose and throat
ERJ	European Respiratory Journal
EUnetHTA	European Network of Health Technology Assessment
GDP	gross domestic product
GI	gastrointestinal
GMS	general medical card scheme
GP	general practitioner
HCA	Health Care Assistant
HDI	human development index score
HDU	high dependency unit
HIQA	Health Information and Quality Authority

HMV	home mechanical ventilation
HPO	Healthcare Pricing Office
HPRA	Health Products Regulatory Authority
HRQL	health related quality of life
HSE	Health Service Executive
HTA	Health Technology Assessment
Icare	insurance and care
ICU	intensive care unit
IPC	infection prevention and control
IQR	interquartile range
ISCoS	International Spinal Cord Society
ISNCSCI	The International Standards for Neurological Classification of Spinal Cord Injury
IV	Intravenous
MACE	Malone antegrade continence enema
MAID	medical assistance in dying
MASCIP	Multidisciplinary Association for Spinal Cord Injury Professions
MIE	mechanical insufflation exsufflation
MMUH	Mater Misericordiae University Hospital
MSCIS	Model Spinal cord Injury Systems
NCPE	National Centre for Pharmacoeconomics
NICE	National Institute for Health and Care Excellence
NIV	non-invasive ventilation
NRH	National Rehabilitation Hospital
NSCISC	National Spinal Cord Injury Statistical Center
OHS	obesity hypoventilation syndrome
OT	Occupational Therapist
OWSA	one way sensitivity analysis
PCRS	Primary Care reimbursement Service
PEDro	physiotherapy evidence database
PICo	Population, Area of Interest and Context
PRESS	peer review of electronic search strategies
PRISMA-ScR	Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews
PVA	Paralysed Veterans of America
RAD	respiratory assist devices
RCT	randomised control trial
RISCI	Respiratory information for Spinal Cord Injury
RTA	road traffic accident
SCI	spinal cord injuries
SD	standard deviation
SII	Spinal Injuries Ireland
SPC	summary of product characteristics
SRI	severe respiratory insufficiency
TAS	treatment abroad scheme
TRIP	Turning Research Into Practice
TSANZ	Thoracic Society of Australia and New Zealand

TSCI	traumatic spinal cord injuries
UNCRPD	United Nations Convention on the Rights of Persons with Disabilities
UTI	Urinary Tract Infection
VAC	voluntary anal contraction
VAT	value added tax
VTE	venous thromboembolism
WHO	World Health Organization

1. Introduction

1.1. Background to the request

A request was received by HIQA in April 2021 from the Clinical Lead for the National Clinical Programme for Rehabilitation Medicine in the Health Service Executive (HSE), and the Programme Manager for the Spinal Cord System of Care Programme in the National Rehabilitation Hospital (NRH), to carry out a health technology assessment (HTA) of domiciliary ventilation for adults with spinal cord injuries (SCI). This request was prioritised for inclusion in the HIQA Board approved HTA work plan. Work commenced on this HTA in August 2022.

Spinal cord injury is damage to the spinal cord that causes changes in its function, either temporary or permanent. These changes translate into loss of muscle function, sensation, or autonomic function in parts of the body served by the spinal cord below the level of the lesion.(1, 2) The consequences of SCI depend on the severity and location of the injury on the spinal cord; injuries that occur higher up the spinal cord (that is, the cervical spine) and that cause complete loss of function below that level of injury, result in the most serious symptoms (for example complete paralysis of all four limbs and torso, respiratory failure).(3) Individuals with complete high cervical SCI (C3 or higher) will generally require invasive mechanical ventilation to survive as neural control of breathing is inhibited.(4) While the majority of these individuals will eventually be weaned off the ventilator over time,(5) a minority will have lifelong dependency on invasive mechanical ventilation.(6) Some individuals with SCI retain neural control of breathing (for example, those with lower or incomplete SCI), but because of diminished respiratory reserve, cannot be weaned completely and require non-invasive ventilation (NIV). For these particular individuals, the requirement for NIV may be continuous or, more frequently, for a proportion of the day or just nocturnally. While acknowledging that some individuals with SCI may require NIV, the focus for this particular HTA is on those requiring invasive ventilation due to the more complex and pressing healthcare needs associated with this patient cohort.

An integrated care pathway for patients with SCI has been developed in Ireland by the National Clinical Programme for Rehabilitation Medicine, in line with its model of care.(2, 7) Although the NRH can now admit and manage ventilated patients successfully as part of this pathway, discharge planning for these complex cases remains a huge challenge for several reasons, including the current fragmented funding model for community services and the limited availability of suitably trained homecare staff in the community. There may also be regional variation in the availability of the necessary resources to enable the homecare of these individuals. As a result, these individuals often remain hospitalised for a prolonged period of time, often several years, before being discharged home.(2) These delayed discharges can have a detrimental impact on the individual, their family and carers, management of bed capacity in the NRH, and the wider healthcare system due to the significant cost of treating these patients in hospital.

In line with Sláintecare reforms, there is an ambition to achieve a universal single-tier health and social care system, which provides equitable access to services based on need, and not ability to pay. One of the key objectives of Sláintecare reforms is about delivering the right care, in the right place, at the right time, by the right team.(8) Specifically, a key aspect of the reform is the shift of care out of acute hospitals into the community and closer to a person's home, where safe to do so. In line with international best practice and guidance, domiciliary ventilation is considered the standard of care for ventilator-dependent individuals with SCI where clinically appropriate.(9-12) Despite this, and despite its provision being consistent with the vision of Sláintecare,(8) this service has only been provided in a very limited number of circumstances in Ireland to date.

As the provision of domiciliary ventilation for SCI patients is already included in the HSE's integrated care pathway, a comprehensive assessment of its clinical effectiveness, safety and cost effectiveness was not considered necessary. To date, the provision of this care has been organised at a local level on an individual patient basis. The purpose of the HTA is therefore to assess the organisational, budget impact and resource implications associated with the systematic provision of this care; it will also consider the social and ethical issues arising from the provision of care to individuals with substantial needs in a home setting. The focus of this HTA is on an individual's own (domestic) home as opposed to a nursing home or other residential care setting. The HTA will inform a decision by the HSE regarding the national delivery of care for adults (including young adults ≥ 16 years old) with spinal cord injuries who require permanent invasive mechanical ventilation and in whom discharge home is deemed clinically appropriate.

1.2. Terms of reference

Following initial scoping of the available evidence, the terms of reference of this assessment were agreed between HIQA and the HSE's National Clinical Programme for Rehabilitation Medicine and the NRH's Spinal Cord System of Care Programme.

For adults with spinal cord injuries who require permanent mechanical invasive ventilation and in whom discharge home is deemed clinically appropriate, the terms of reference for the HTA are to:

- describe the epidemiology and burden of disease for this population
- briefly describe the technology and outcomes associated with domiciliary invasive ventilation
- review international practice and standards relating to the provision of domiciliary invasive ventilation for this population
- examine the potential organisational and resource implications associated with providing domiciliary invasive ventilation for this population
- assess the budget impact of providing mechanical invasive ventilation for this population at home compared with in hospital or long-term care settings
- consider the social and ethical aspects associated with providing domiciliary invasive ventilation for this population
- based on the findings of this assessment, provide advice to the Minister for Health and the HSE.

1.3. Overall approach

Following agreement on the terms of reference between HIQA, the HSE and the NRH, HIQA appointed an evaluation team comprising staff from the HTA Directorate to carry out the assessment.

HIQA convened an Expert Advisory Group (EAG) comprising representation from key stakeholders including the HSE, clinicians with specialist expertise in the management of patients with spinal cord injuries requiring mechanical ventilation, those involved in funding community services, and patient and family carer representation. The role of the EAG is to inform and guide the process, provide expert advice and information, and to provide access to data where appropriate. A full list of the membership of the Expert Advisory Group is available in the acknowledgements section of this report.

The terms of reference for the Expert Advisory Group are to:

- contribute to the provision of high quality and considered advice by HIQA to the Minister for Health and the HSE
- contribute fully to the work, debate and decision making processes of the group by providing expert guidance, as appropriate

- be prepared to provide expert advice on relevant issues outside of group meetings, as requested
- provide advice to HIQA regarding the scope of the analysis
- support the Evaluation Team led by HIQA during the assessment process by providing expert opinion and access to pertinent data, as appropriate
- review the project plan outline and advise on priorities, as required
- review the draft report from the Evaluation Team and recommend amendments, as appropriate
- contribute to HIQA's development of its approach to HTA by participating in an evaluation of the process on the conclusion of the assessment.

The Terms of Reference of the HTA were reviewed by the EAG at its first meeting. Draft Chapters and the HTA protocol were circulated to the Expert Advisory Group for review and discussed at two formal meetings of the group, with amendments made where appropriate. The protocol for the HTA was published on HIQA's website in February 2023.(13) The final version of the assessment was approved by the Board of HIQA in May 2023, and was then submitted to the Minister for Health and the HSE as advice and published on HIQA's website in June 2023.

1.3.1. Terminology

In the literature, the term *'patient'* is often used interchangeably with other terms such as *'service user'* or *'client',* particularly if the person is not receiving acute care in a hospital setting. However, for consistency, the term *'patient'* is used throughout this HTA to refer to the individual with SCI, where appropriate. Similarly, the terms *'carer', 'caregiver'* and *'supporter'* are often used synonymously with *'family'* and *'family carers'*, to refer to the unpaid support and assistance provided by a person who is generally a family member, partner or friend. For consistency, the term *'carer'* is used throughout this HTA to describe this unpaid caregiving role, unless there is a need to specify the family or other individuals. Many different professionals provide formal care and support to these patients in their homes. While some of these may be classified as *'healthcare professionals'* under the Professional Qualifications Directive (for example, nurses), others may not (for example, healthcare assistants) and for this reason this term is not used to describe these professionals providing homecare in this HTA.(14) Instead the term *'homecare workers'* is used when describing staff who are funded for the delivery of supports and services to the patient in their home, unless there is a need to specify a particular profession.

2. Burden of disease and epidemiology

Key points

- The range of impairments resulting from spinal cord injuries (SCI) are largely determined by the location on the spinal cord in which the lesion occurs, and the completeness of the injury. Patients with complete C1-C3 level cervical lesions will experience the most severe level of impairment and tend to require a ventilator and tracheostomy to breathe.
- In general, permanent ventilator assistance is required for complete SCI at C1-C3 due to diaphragmatic paralysis. Patients with C3-C4 injuries experience diaphragmatic dysfunction, but may retain partial ventilatory function. Weaning from a ventilator may be achieved in patients with C3 injuries, while injuries of C4 and below are usually associated with successful weaning, in the absence of other complications.
    - Apart from completeness and level of injury, other barriers to weaning include older age, smoking history, active pneumonia, the severity of impaired consciousness, high level injury severity score, underlying cardiorespiratory diseases and other complicating comorbid conditions.
- In Ireland and internationally, tetraplegia was reported to be the most common spinal cord injury type, with upper cervical (C1-C4) injuries accounting for a larger number of cases than lower cervical (C5-C8) injuries. Between 2008 and 2021, the most frequent upper cervical injury type in Ireland was C4.
- Traumatic SCI are predominantly a male phenomenon, accounting for 82% (n=185) of C1-C4 SCI discharges from the National Spinal Injuries Unit (NSIU) in the Mater Misericordiae University Hospital (MMUH) between 2008 and 2021.
- Overall, C1-C4 traumatic SCI are rare in Ireland and numbers fluctuate from year to year. Incidence of C1-C4 discharges (in patients aged 15 years and older) from the NSIU fluctuated from 2.7 to 3.6 cases per million population over the 14 year period (with the highest recorded in 2013 at 5.9 and the lowest in 2010 at 2). The absolute number of discharged cases ranged from 9 to 27 cases per annum over the 14 year period.
- The most common age groups at time of admission to the NSIU were those aged 65-74 (22%, n=49) and those aged 15-24 year olds (17%, n=39). Cases in those aged 65 and older represent an increasing proportion of incidence.

However, establishing the causes of apparent trends is challenging as the numbers of cases are small.

- Falls and road traffic accidents were the leading causes of traumatic SCI in Ireland between 2008 and 2022. Data from Ireland and internationally indicate that the incidence of SCI in older age groups is increasing, largely driven by falls in these older individuals.
- The key factors associated with increased risk of mortality and morbidity in patients with traumatic SCI are a higher level of injury, the completeness of the injury, older age, ventilator dependency, and the presence of multi-system trauma.
    - There are many complications associated with cervical SCI. Among these, specific areas of concern for management include venous thromboembolism (VTE), neurogenic and spinal shock, SCI-induced bradycardia, respiratory complications, acute autonomic dysreflexia, pressure injuries, neurogenic bowel and bladder dysfunction, mental health issues, and pain.
    - Pneumonia and respiratory failure remain a significant cause of death in patients with SCI.
- Median life expectancy for patients with SCI reportedly ranges from 2.8 to 43 years from time of injury, with survival time related to age at time of injury. In addition to age, factors that influence life expectancy include sex, level and completeness of injury, and ventilator dependency. Ventilator dependency was the factor associated with the lowest life expectancy, reportedly reducing life expectancy by approximately 75% compared with the general population.
- Patients with C1-C3 SCI requiring permanent mechanical ventilation, and whose condition is considered clinically appropriate to be discharged home, will account for a very small proportion of the total number of patients admitted with SCI (estimated from the literature to be approximately 3 to 5% of all individuals with tetraplegia).
- Between 2008 and 2022, at least eight patients who required ventilation assistance were discharged from the NSIU. It is estimated that between zero and two patients with SCI annually are anticipated to require permanent ventilation in Ireland.
- It is estimated that there are currently between five and ten adults with SCI in Ireland receiving invasive mechanical ventilation at home.

2.1. Introduction

The aim of this Chapter is to describe the epidemiology of high cervical spinal cord injuries (SCI) and the associated disease burden experienced by patients as a result of complications cause by the SCI, such as respiratory failure and ventilatory support requirements. The primary focus of the HTA is related to adults with SCI who require permanent mechanical invasive ventilation and for whom discharge is deemed clinically appropriate. This Chapter first describes the pathophysiology and classification of SCIs, then outlines the international literature on the burden of disease in terms of morbidity and mortality, and lastly presents epidemiological data on SCI in Ireland.

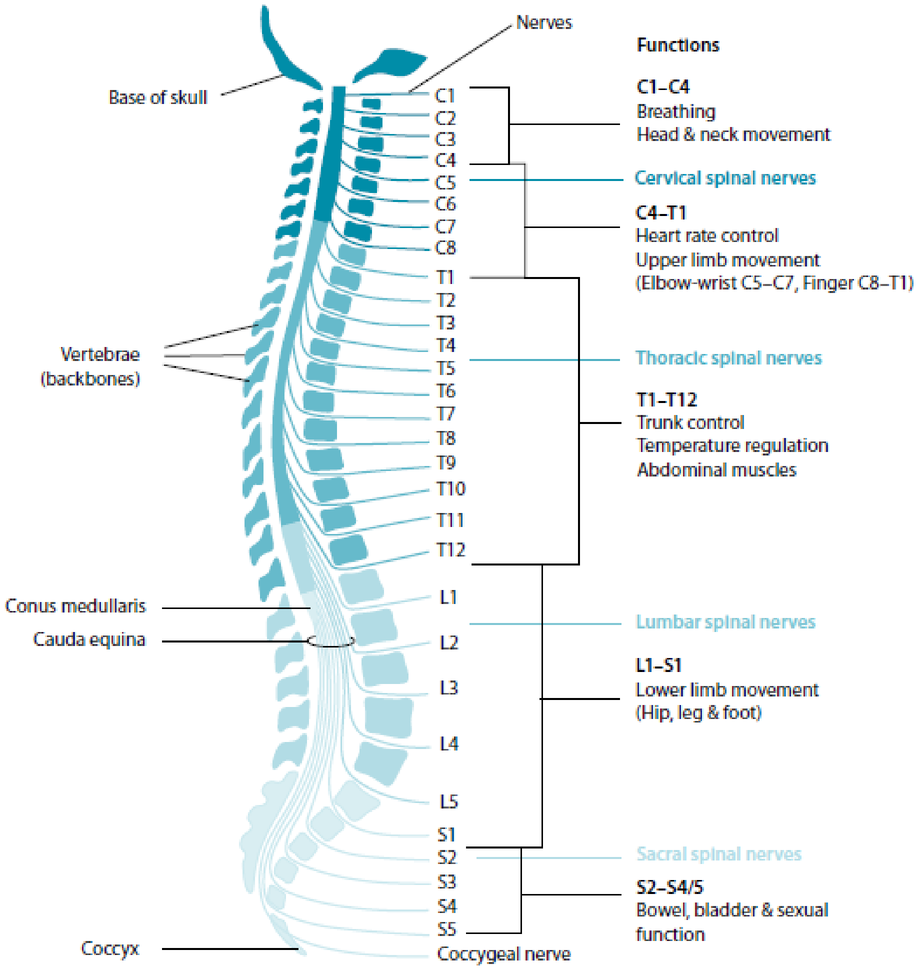
2.2. Spinal cord injury

2.2.1. Pathophysiology

Damage to the spinal cord can be life-threatening or result in life-changing injuries, with the potential for long-term disability in survivors.(15) The management of adults with SCI is complex given that damage to the spinal cord can cause a range of functional losses (motor, sensory and autonomic), depending on the extent of the damage to the spinal cord.(4) Such damage can be the result of traumatic injury, including traffic accidents and falls, or a non-traumatic cause, such as spinal tumours or degenerative disease.(4, 16) According to the published literature, the most common causes of traumatic SCI are transport-related incidents, particularly road traffic accidents, and unintentional falls (see section 2.4.6).(4) In the case of non-traumatic SCIs, the leading causes have been reported to be spinal tumours and degenerative disorders, followed by vascular and autoimmune conditions, although it has been reported that there is a lack of reliable national level data to inform understanding on causation.(4, 16) As detailed in section 2.4.5, the incidence of SCI in older age groups is increasing, largely driven by falls in these older individuals. The published literature from Ireland and internationally, have noted an increased mean age of onset for cases of non-traumatic SCI compared with traumatic SCI.(17-19) In 2019, a review of non-traumatic spinal cord injuries notes that, given such trends, increasing numbers of people are likely to experience non-traumatic spinal cord injuries in the future due to ageing populations.(16)

The higher up the spinal cord the lesion occurs, the more extensive the range of impairments will generally be (Figure 2.1).(4) After a spinal cord injury, all the nerves above the level of injury continue to function normally. At the level of injury, messages are blocked from being transmitted from above that level to below the level of injury. In brief, cervical SCI commonly causes sensory and motor loss in the arms, torso and legs (that is, tetraplegia). In addition, these patients with cervical SCI generally have no control over their bowel and bladder movements, and tend to require a ventilator and tracheostomy to breathe (if the lesion is C4 or higher) (Figure 2.1), because the lesion directly impacts on autonomic control. Thoracic SCI typically causes sensory and or motor loss in the legs and trunk (that is, paraplegia). Lumbar SCI commonly causes sensory and motor loss in the legs and hips.(4) Of note, all forms of SCI may also result in chronic pain, and mental health issues are common among these individuals.(20) In addition, patients may retain some residual function beneath the level of the injury, in which case the injury is referred to as incomplete. A complete injury, in contrast, results in permanent lack of function below the level of the injury.(15)

Figure 2.1: Longitudinal organisation of the spinal cord (with cervical, thoracic, lumbar and sacral segments shaded); spinal vertebrae, spinal nerves, and a rough representation of major spinal cord functions



Reproduced with permission from the WHO.(4) Reprinted from International Perspectives on Spinal Cord Injury, WHO, Chapter 1: Understanding spinal cord injury, Page 5., Copyright WHO (2013). URL: https://www.who.int/publications/i/item/international-perspectives-on-spinal-cord-injury; Date accessed 21 Oct 2022

Following a traumatic SCI, the mechanisms of primary and secondary injuries can be divided into separate phases:(21)

- acute phase (less than 48 hours from the time of injury)
- sub-acute phase (between 48 hours and 14 days)
- intermediate phase (between 14 days and six months)
- chronic phase (more than six months).

Briefly, the acute phase refers to primary and secondary physiological processes immediately resulting from the initial traumatic event. The primary injury refers to the initial dislocation and damage of the vertebral column, causing compression of the spinal cord. This results in additional damage to the cells of the central nervous system around the affected region, such as neurons and oligodendrocytes (that is, a specialised type of glia cell of the central nervous system), along with the surrounding vasculature. Significant cell death and blood vessel damage trigger a sustained secondary injury cascade, causing an influx of pro-inflammatory cells and signals that further damage the spinal cord and impair neurological function.(21)

The subacute phase is characterised by a sustained cycle of cell death and pro-inflammatory microenvironment around the damaged spinal cord. Ischaemia, excitotoxicity and metabolic dysregulation contribute to neuron and glia (that is, cells of the central nervous system) cell death. Ongoing cell death and release of cellular products (adenosine triphosphate (ATP), deoxyribonucleic acid (DNA), potassium) leads to activation of microglial cells (that is, immune cells of the central nervous system), the infiltration of other pro-inflammatory immune cells, and the resulting release of pro-inflammatory cell signals. This additionally contributes to ongoing cell death of neurons and oligodendrocytes. The release of high levels of neurotransmitters such as glutamate from these dying neurons can in turn cause excitotoxic cell death due to overactivation of receptors on surrounding cells. These processes can cyclically sustain the secondary injury cascade, resulting in further damage to the spinal cord.(21)

The subsequent intermediate-to-chronic phases are marked by a shift towards regulation and regeneration as the inflammatory response subsides.(21) The combination of neural precursor cell pools and exhibited neuronal and synaptic plasticity of neurons in the central nervous system may contribute to partial regeneration and functional recovery of the injured spinal cord over time. However, there are inherent properties of the central nervous system that can inhibit neuronal cell regeneration, as well as physical barriers to vascular remodelling, such as cystic cavities and glial scars formed as a result of the considerable cell death and damage occurring in the acute and sub-acute phases. The cellular mechanisms occurring during these latter phases are the subject of ongoing research and investigation.(21)

2.2.2. Classification of spinal cord injuries

Classification of a patient's spinal cord injury involves complex steps requiring extensive clinical examination of the patient. The International Standards for Neurological Classification of Spinal Cord Injury (ISNCSCI) is a clinical documentation tool to facilitate the standardised clinical examination of sensory and motor function to quickly determine the level and severity of a spinal cord injury.(22) The ISNCSCI guidelines were first developed in 1982 to provide consistent classification of the neurological level and extent of SCI.(23) The ISNCSCI is published by the American Spinal Injury Association (ASIA) and maintained by the International Standards Committee of ASIA and the International Spinal Cord Society (ISCoS), with the publication of the eighth edition in 2019.(15) The ISNCSCI tool has been noted to have been adopted internationally by clinical communities and used in clinical trials.(21)

Broadly speaking, the ISNCSCI outline the detailed elements required to carry out sensory and motor examinations to generate scores to characterise sensory and motor functioning (ASIA impairment scale), and the completeness of the injury.(22) The sensory level of injury is examined by systematically performing light touch and pin prick assessment for each spinal segment to determine the lowest spinal segment with normal sensory function.(15) The motor level of injury is determined using a 6-point graded score (from zero to five) for the five key muscle groups of the upper extremities and five in the lower extremities. The neurological level of injury can then be determined as the level where motor and sensory function are both intact bilaterally. The neurological level of injury is reported as incomplete if some degree of sensation remains intact at the sacral S4/S5 level, and both voluntary anal contract and deep anal pressure are present. Otherwise, the neurological level of injury is considered complete (that is, there is a total loss of function and feeling below the injury level). Together, the sensory, motor and neurological level of injury indicate which ASIA Impairment Scale classification is assigned, as described in Table 2.1. It should be noted that the ISNCSCI tool does not include additional elements required to provide a full and comprehensive neurological examination, such as deep tendon reflexes or evaluation of position sense.(15) From a prognostic perspective, the distinction between complete and incomplete is important as those with incomplete injuries often have capacity for recovery whereas those with complete injuries often do not.

Table 2.1: ASIA Impairment Scale (AIS) and description of grades.(24)

ASIA Impairment Scale	Description of grades
A – Complete	No sensory or motor function is preserved in the sacral segments S4-5.
B – Sensory incomplete	Sensory but not motor function is preserved below the neurological level and includes the sacral segments S4-5 (light touch or pin prick at S4-5 or deep anal pressure) AND no motor function is preserved more than three levels below the motor level on either side of the body.
C – Motor incomplete	Motor function is preserved at the most caudal sacral segments for voluntary anal contraction (VAC) OR the patient meets the criteria for sensory incomplete status (sensory function preserved at the most caudal sacral segments S4-5 by LT, PP or DAP), and has some sparing of motor function more than three levels below the ipsilateral motor level one either side of the body. (This includes key or non-key muscle functions to determine motor incomplete status). For AIS C – less than half of key muscle functions below the single NLI have a muscle grade ≥ 3.
D – Motor incomplete	Motor incomplete status as defined above, with at least half (half or more) of key muscle functions below the single NLI having a muscle grade ≥ 3.
E – Normal	If sensation and motor function as tested with the ISNCSCI are graded as normal in all segments, and the patient had prior deficits, then the AIS grade is E. Someone without an initial SCI does not receive an AIS grade.
ND	To document the sensory, motor and NLI levels, the ASIA Impairment Scale grade, and/or the zone of partial preservation (ZPP) when they are unable to be determined based on the examination results.

Key: DAP – deep anal pressure; ISNCSCI – International Standards for Neurological Classification of Spinal Cord injury; LT – light touch; NLI – neurological level of injury; PP – pin prick; S4-5 – sacral level 4-5; SCI – spinal cord injury.

Source: American Spinal Injury Association.(24) Used with permission. International Standards for Neurological Classification of Spinal Cord Injuries (ISNCSCI) Worksheet, 2019. https://asia-spinalinjury.org/international-standards-neurological-classification-sci-isncsci-worksheet/

2.3. Burden of disease

2.3.1. Spinal cord injury complications

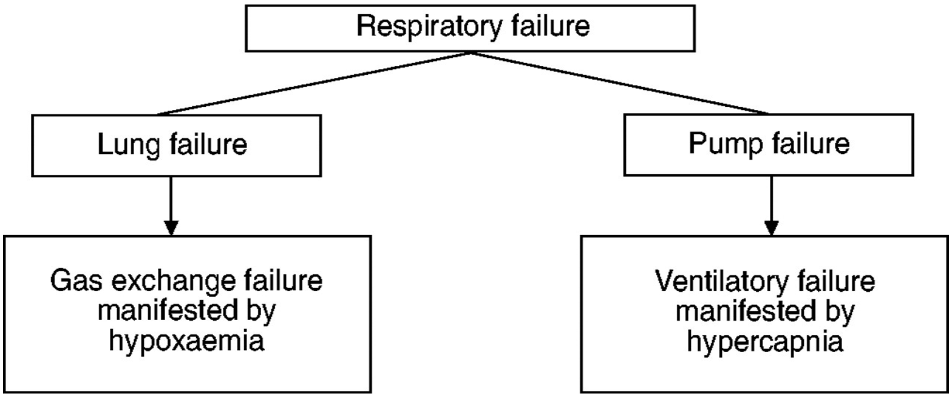
Spinal cord injuries can lead to many long-term complications depending on the location and severity of the injury. Patients with high cervical SCI are particularly affected by many of these secondary complications. Specific areas of management that need to be considered for these individuals include, but are not limited to, venous thromboembolism (VTE) prophylaxis, neurogenic and spinal shock, SCI-induced bradycardia, respiratory complications, prevention and management of acute autonomic dysreflexia, spasticity, pressure injuries, neurogenic bowel and bladder dysfunction, mental health issues, and pain.(15) Patients with SCI may also sustain brain trauma, which may result in cognitive impairment.(25)

While there are many complications associated with cervical SCI (see Section 4.3.5), the following sections address three clinical issues that require specific consideration due to the considerable long-term morbidity and increased risk of mortality associated with their presentation in patients with high cervical SCI, and the requirements for their management. These include respiratory failure, neurogenic bowel and bladder dysfunction, and pressure injuries; the pathophysiology of these will be outlined in the following sections with the associated management discussed in Chapter 3.

2.3.1.1. Respiratory failure

Respiratory failure is a condition in which the respiratory system fails in one or both of its gas exchange functions, that is, oxygenation of and or elimination of carbon dioxide from the blood. It is diagnosed by an arterial oxygen tension ($P_a\,O_2$) of <8.0 kPa (60 mmHg), an arterial carbon dioxide tension ($P_a\,CO_2$) of >6.0 kPa (45 mmHg) or both.(26) The respiratory system broadly consists of two parts: the lung (that is, the gas-exchanging organ) and the pump that ventilates the lungs (which includes the chest wall and associated muscles, respiratory controllers in the central nervous system, and the innervating spinal cord and peripheral nerves). In general, failure of the lung leads to hypoxaemia (low oxygen levels) with normocapnia (normal carbon dioxide levels) or hypocapnia (low carbon dioxide levels), which is called Type I respiratory failure. Failure of the pump results in alveolar hypoventilation and hypercapnia (high carbon dioxide levels), which is called Type II respiratory failure (Figure 2.2).(26) In the context of patients with high cervical SCI, it is type II (hypercapnic) respiratory failure that is experienced, which is sometimes chronic in nature.

Figure 2.2: The two main types of respiratory failure



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Traumatic injury to the cervical (and upper thoracic) spinal cord is associated with variable degrees of respiratory dysfunction, dependent on the level and completeness of injury; ventilator support is often required, particularly for cervical SCI, at least in the short term.(27) Early tracheostomy is recommended to assist in mechanical ventilation during the acute and more chronic phases of SCI management.(15) The desired outcome for patients with SCI requiring ventilator assistance is to ultimately wean them from the mechanical ventilator once they are able to breathe spontaneously; however, this may not be possible in a minority of patients with SCI (estimated to be around 3% to 5% of those with tetraplegia),(28, 29) and so long-term ventilation may be required in these individuals.(30) In its 2021 annual report, the National Spinal Cord Injury Statistical Center (NSISC) in the US reported that 327 of 8,940 (3.4%) patients with tetraplegia who were alive 12 months post-injury were dependent on mechanical ventilator use. This is in contrast with patients with paraplegia, where only 19 of 8,562 (0.2%) were dependent on mechanical ventilator use one year post-injury.(29)

The diaphragm is the primary muscle of inspiration and hence the degree of diaphragmatic involvement in SCI influences the ability to wean from ventilator support.(31) While reports describe slightly different implications by injury level,(15, 31) permanent ventilator assistance is usually, but not always, required for complete SCI at C1-C3 due to diaphragmatic paralysis.(31) Patients with C3-C4 injuries experience diaphragmatic dysfunction, but they may have partial ventilatory function.(15) Initial temporary ventilation with subsequent weaning may be achieved in those with complete SCI at C3. In the absence of other factors that would prevent ventilatory weaning, complete injuries at C4 and below and incomplete SCI at any level are usually associated with successful weaning.(31) Other barriers to weaning include older age, smoking history, active pneumonia, the severity of impaired consciousness, high level injury severity score, underlying cardiorespiratory diseases and other complicating comorbid conditions (such as those involving impaired central drive).(30, 32)

A 2021 systematic review and meta-analysis investigated weaning success and duration of mechanical ventilation in patients with SCI requiring mechanical ventilation.(33) The authors found the following to be clinically relevant predictors of increased odds of weaning failure: a high number of comorbidities ($p = 0.01$), high injury severity score ($p < 0.0001$), high-level cervical lesions (that is, C1-C3 compared to C4-C7) ($p < 0.0001$), elevated heart rate ($p = 0.0085$), and the presence of tracheostomy ($p < 0.0001$). The factors associated with a longer duration of mechanical ventilation were later admission to a specialised SCI centre ($p < 0.001$), high-level cervical lesions (that is, C1-C4 compared with C5-C8) ($p = 0.01$), complete lesions ($p = 0.048$), low tidal volume and high positive end expiratory pressure in the first 24 hours from admission ($p = 0.0094$; $p = 0.0027$, respectively), and presence of tracheostomy ($p < 0.0001$).(33) The association between completeness of injury and requirement of mechanical ventilation was also reported in a 2020 systematic review and meta-analysis investigating extubation failure in patients with cervical SCI.(34) The authors reported that the odds of extubation failure were 2.76 (95% CI, 1.14 to 6.70) times greater for patients with complete cervical SCI compared to those with incomplete lesions.(34)

2.3.1.2. Neurogenic bladder dysfunction

Neurogenic bladder dysfunction is caused specifically by damage to the brain, nerves or spinal cord and is distinct from bladder dysfunction caused by other issues (for example, pelvic floor problems or ageing).(35) The majority of patients with SCI experience neurogenic bladder dysfunction, however the type and symptoms are dependent on the extent and location of the injury. Control of urination involves both bladder reflexes (in which emptying is triggered when the bladder is full) and voluntary control (in which urine can be held until a socially appropriate time to empty). After SCI, nerve signals that control urination are blocked and this leads to changes in bladder control and sensation.(35)

In relation to patients with injuries above T12, spastic bladder (also called reflex or overactive bladder) occurs, which involves the unpredictable emptying of the bladder as a result of overactive bladder muscles.(35) This is largely due to the blockage of the sympathetic efferent nerve supply to the bladder and urethra (located between T11 and L2) which provides inhibitory input to the bladder detrusor muscle.(36) For individuals with spastic bladder, the bladder sphincter muscles are also usually overactive and this is called detrusor dyssynergia or detrusor sphincter dyssynergia (DSD). When both the bladder sphincter and the bladder wall muscles contract, this causes an increased pressure which can ultimately damage the bladder and kidneys.(35) Patients with SCI are also at risk of complications such as autonomic dysreflexia (if injury above T6), urinary tract infections (UTIs), kidney and bladder stones, and kidney damage.

2.3.1.3. Neurogenic bowel dysfunction

It is estimated that over 80% of all patients with SCI experience some degree of neurogenic bowel dysfunction.(37-39) The most commonly reported symptoms include faecal incontinence, constipation, need for digital/manual evacuation of the rectum, abdominal distension or discomfort, and haemorrhoids.(37) The symptoms are considered moderate to severe in 39-50% of patients with SCI, with the severity of dysfunction associated with depression and reduced quality of life.(37) The evidence suggests that bowel symptoms are more severe in patients with complete SCI, however it is unclear as to whether symptoms are more or less common in those with cervical and upper thoracic lesions than in those with lower spinal cord lesions.(37)

SCI affect bowel function mainly in the colorectum and anal canal. In general, parasympathetic activity enhances secretion and peristalsis (that is, the contraction and relaxation of intestinal muscles that pushes the contents along) while relaxing the gastrointestinal (GI) sphincters. Parasympathetic innervation stems from the vagus nerve and the S2-S4 regions of the spinal cord. In contrast, sympathetic innervation (stemming from T9-L2 regions) reduces secretion and peristalsis while contracting GI sphincters.(37) The internal anal sphincter is under reflex control of the enteric nervous system and the sacral spinal cord, whereas the external anal sphincter muscle is partly under voluntary control of the pudendal nerve (S3-S5), so that defecation can be interrupted until socially appropriate.(37)

Those with lesions above S2 tend to have increased tone of the external anal sphincter muscle and additionally increased tone and contractility of the rectum.(37) Spastic bowel (also called reflexic bowel and less commonly upper motor neuron bowel) usually occurs with injuries at T12 and above.(40) For these individuals, the bowel's natural reflexes are retained, however they no longer have voluntary control over their bowels which may result in reflex defecation when something is present in the rectum. Due to the tightened anal sphincter muscles, people with spastic bowel are generally unable to pass a stool without assistance.

2.3.1.4. Pressure injuries

Spinal cord injuries can result in a number of skin complications, in particular pressure injuries. A pressure injury is defined as "localised injury to the skin and/or underlying tissue usually over a bony prominence, as a result of direct pressure or shear and the resulting deformation of the underlying soft tissues."(41) Pressure injuries complicate the rehabilitation process for patients with SCI and can result in prolonged hospitalisations, delayed community reintegration, reduced quality of life and loss of self-esteem. All patients with SCI are at risk for the development of pressure injuries and almost all will develop at least one serious pressure injury during their lifetime.(41) In particular, the prevalence of pressure injuries increases among those with longer SCI durations, especially among those 10 years or more post injury.(42)

Immobility due to SCI is a major risk factor for the development of pressure injuries.(43) Other reasons for the increased risk of pressure injuries in patients with SCI include impaired autonomic regulation of subcutaneous blood flow and skin moisture levels, reduced immune response, reduced skin temperature reactivity, and changes in connective tissue composition. Long-term impacts of SCI may also include changes in muscle mass, metabolic function, and body weight, all of which can directly or indirectly affect skin integrity.(44)

2.3.2. Mortality, cause of death and life expectancy

2.3.2.1. Mortality

Premature mortality is increased in patients with SCI.(4, 45) In a 2013 report by the WHO, it was estimated that the mortality risk is highest in the first year after SCI and remains high compared with the general population; it was found that those with SCI are two- to five- times more likely to die prematurely than those without SCI.(4) Similarly, in 2015, a systematic review and meta-analysis investigated mortality in patients with spinal cord injuries and estimated an overall pooled standardised mortality ratio of 2.45 (95% CI, 1.86 to 3.22) relative to the general population.(45) The first year after injury has been reported to have the highest risk of mortality for patients with SCI.(4, 21) A 2017 overview of traumatic SCI noted that mortality estimates during the acute in-hospital period ranged from 4% to 17%, while after hospital discharge, 3.8% of patients died within the first year of injury, dropping to 1.6% in the second year and 1.2% thereafter.(21) A 2015 systematic review and meta-analysis investigating mortality after SCI reported that in-hospital mortality for patients with a cervical traumatic SCI varied between 4.2% and 26.2%.(45)

There are several factors associated with increased risk of mortality in patients with traumatic SCI, including the severity and completeness of injury, higher level of the injury (that is, cervical SCI are associated with higher mortality than thoracic or lumbar SCI), older age, ventilator dependency, and the presence of multi-system trauma.(4, 21, 45, 46)

Multiple studies have reported that higher level lesions and complete SCI are associated with increased risk of mortality.(4, 15, 45-47) A 2015 systematic review and meta-analysis investigating mortality after SCI reported a pooled hazard ratio estimate of 1.79 (95% CI, 1.51 to 2.12) when comparing all-cause mortality risk for patients with tetraplegia versus paraplegia, and reported that studies found decreasing survival with increased severity of SCI and lesion level.(45) In 2017, Savic et al. reported on long-term survival after SCI data from two spinal centres in Britain between 1943 and 2010. The authors reported that mortality odds ratios increased with level of injury and the odds of mortality for patients with upper cervical (C1-C4) SCI was almost four times higher than for patients with incomplete AIS D injuries (odds ratio 3.85, 95% CI, 3.22 to 4.58; $p < 0.0001$).(46)

A retrospective analysis of in-hospital mortality among patients with complete acute traumatic SCI at a tertiary care centre in India was published in 2020. The authors found an increased risk of mortality for patients with high tetraplegia (hazard ratio

4.14, 95% CI 1.84 to 9.34) and low tetraplegia (hazard ratio 3.44, 95% CI 1.87 to 6.32) compared with patients with paraplegia.(47) In this study, the mortality rate in patients with paraplegia and tetraplegia was 3% (n = 15/464) and 22% (n = 64/294), respectively. Mortality as a proportion was similar among patients with high tetraplegia (n = 10/46, 22%) and low tetraplegia (n = 54/194, 22%). The overall mortality rate among patients admitted with traumatic SCI was 10% (n = 79/758). Upper cervical tetraplegia (grade C1-C4) was the smallest cohort of patients (n = 46/758, 6%), followed by lower cervical tetraplegia (grade C5-C8) (n = 248/758, 33%), while the majority of patients had thoracic or lumbar paraplegia (n = 464/758, 61%).(47)

Older age at the time of SCI has been found to be significantly associated with increased risk of mortality.(21, 45, 47, 48) A 2011 publication reported survival outcomes of SCI patients receiving assisted ventilation on or after admission between 1981 and 2005 in England and found increasing age was associated with increased risk of death in these patients.(48) The hazard ratio for patients aged 46 and older was 3.1 (95% CI, 1.26 to 7.85; $p < 0.017$) compared with the reference group aged 0 to 30 years.(48) A 2022 publication observed a similar association between older age and increased mortality in India. The authors reported a statistically significant increased odds of mortality associated with increasing age (odds ratio (95% CI); 1.03 (1.01 to 1.06); $p = 0.017$) among their cohort of patients with complete acute traumatic SCI.(47)

In addition, both the need for, and duration of, ventilator support has been reported to be associated with increased risks of mortality in patients with SCI.(46-49) In 2017, Savic et al. reported that ventilator-dependent SCI patients have a statistically significant increased odds of mortality (odds ratio (95% CI), 7.47 (5.37 to 10.40); $p < 0.0001$) compared to patients with SCI at level T1 or below.(46) Previously, a retrospective review of patients with acute SCI requiring ventilation on or after admission between 1981 and 2005 reported that mechanical ventilation at the time of discharge was associated with increased probability of death.(48) The hazard ratio for patients on mechanical ventilation at discharge was 2.26 (95% CI, 1.04 to 4.90; $p = 0.039$) compared with patients weaned from mechanical ventilation at discharge.(48) Similarly, data from India indicated an increased risk of mortality among patients with acute traumatic SCI requiring use of a ventilator (hazard ratio (95% CI); 14.44 (8.31 to 25.07); $p < 0.001$) compared with no ventilator use.(47) In 2018, a study reported on a cohort of 106 patients with cervical SCI admitted to a neurotrauma ICU in India between 2017 and 2018.(49) The authors found that increased duration of ventilator dependence was associated with an increased odds of mortality (odds ratio (95% CI), 3.30 (1.73 to 6.27); $p < 0.001$).(49)

A 2020 publication from the USA reported that long term respiratory management outcomes for patients with high level SCI have not improved considerably over the past 40 years.(50) Several studies have reported on risk factors associated with increased ventilator requirement and duration among patients with SCI.(33, 34, 51) As described in Section 2.3.1, factors associated with increased duration of mechanical ventilation and extubation failure include higher level lesions and the completeness of injury.(33, 34) In addition, in a 2022 systematic review investigating the timing of tracheostomy in patients with acute traumatic SCI, it was reported that early tracheostomy was not associated with decreased short-term mortality in a group of 2,072 patients (relative risk (95% CI); 0.84 (0.39 to 1.79); p = 0.65).(51)

2.3.2.2. Cause of death

Respiratory complications have been reported to be the most significant cause of morbidity and mortality in the acute cervical SCI period.(4, 32, 34) A 2011 retrospective review of acute SCI patients in England reported on the cause of death among a cohort of weaned and ventilated patients (n = 189). There were 134 patients weaned from ventilator and 55 patients requiring ventilator support at the time of discharge. Respiratory complications were the leading cause of death among both weaned (n = 20/35 (57.1%)) and ventilated patients (12/21 (57.1%)).(48) A 2013 review of respiratory management in patients with SCI in Spain reported that approximately 66% of patients with acute SCI experience respiratory complications, such as atelectasis, pneumonia, and respiratory failure requiring mechanical ventilation.(32) In 2022, a systematic review into the incidence of pneumonia in patients with SCI reported that the overall incidence of pneumonia ranged from 0.03 to 7.21 patients per 100 days, with a higher incidence in the acute phase and ICU setting than later in the course of the injury.(52) A retrospective analysis of in-hospital mortality at the Indian Spinal Injuries Centre, New Delhi, was published in 2021.(47) This study provides information on cause of death in patients with complete acute traumatic spinal cord injury, including specifically for those with an upper cervical (C1-C4) SCI. Of 46 patients with C1-C4 SCI (6% of patients), 10 died while in hospital (22%). The causes of death in these patients were respiratory complications (n = 3), septicaemia (n = 2) and death from cardiovascular causes (n = 5). For patients with lower cervical SCI, 22% died (n = 54 / 248), with the most frequent causes of death being respiratory complications (n = 20, 38%), septicaemia (n = 18, 34%) and cardiovascular causes (n = 9, 17%).(47)

Data from 31 SCI centres in 13 Italian regions collected between 2013 and 2014 were reported in 2020. This cohort was restricted to 510 eligible patients with traumatic SCI who had survived the immediate acute period post injury.(53) Of these 510 eligible patients, there were 187 patients with tetraplegia (37%) and 201 with paraplegia (39%) with A, B, or C completeness of injury according to the AIS (Table

2.1). Of these patients, 13 died during the rehabilitation period, of which 11 had tetraplegia and two had paraplegia with A, B, or C completeness of injury. The cause of death was not reported, however, the most frequently reported complications during the hospitalisation period were respiratory (n = 11/13), pressure injuries (n = 10/13), spine instability (n = 5/13) and multi-resistant infections (n = 5/13). Similar complications were reported in the surviving 497 patients; the three most frequent complications were respiratory complications (n = 171, 34.4%), pressure injuries (n = 151, 30.4%) and infections (n = 66, 13.3%).(53)

2.3.2.3. Life expectancy

Patients with SCI are more likely to die prematurely compared with the general population.(4, 45) A 2015 systematic review and meta-analysis of mortality and longevity after SCI reported that the median survival after traumatic SCI ranged from 2.8 to 43 years; survival time will relate to age at time of injury.(45) For overall global one-year survival, the authors reported an overall pooled estimate of 92.8% (95% CI, 89.4 to 95.2) and for Europe of 95.6% (95% CI, 81.0 to 99.1), each with very high heterogeneity (I^2 values of 96.9% and 97.7%, respectively).(45) Using long term data from two spinal centres in Britain from 1943 to 2010, a 2017 publication noted that estimated life expectancies improved significantly between the 1950s and 1980s, with a smaller improvement again in the early 2000s.(46) The authors reported life expectancy for patients with SCI to range from 18.1% to 88.4% of that of the general population, depending on factors such as age, sex, level and completeness of injury, and ventilator dependency. In their report, ventilator-dependent persons had the lowest estimated life expectancy of all injury groups compared with that of the general population; using the example of a 20-year-old male surviving through to first-year post injury, the life expectancy was estimated as 21.9% of the general population between 1943 to 1949, improving to 46.3% of that of the general population between 2010 and 2014.(46)

The National Spinal Cord Injury Statistical Center (NSCISC) in the US published a life expectancy calculator, with estimates based on long-term survival experiences of persons treated at spinal cord injury centres contributing to the Model Spinal Cord Injury Systems (MSCIS) programme.(54, 55) In 2006, Straus et al. published sample life expectancies for a 25-year-old white male with non-violent SCI without ventilator dependence using data from the NSCISC and MSCIS, a reduced version of which is displayed in Table 2.2 (restricted to SCI levels C1-C4).(55)

Table 2.2: Sample life expectancies for 25-year-old white male, by injury level and ASIA grade from Strauss et al. 2006(55)

Group	Life expectancy (years)
General population	50.9
C1-3, grade A	25.4
C1-3, grades B and C	32.2

C4, grade A	26.4
C4, grades B and C	34.9

Note: C1-3 refers to upper cervical spinal cord injuries of levels one to three; C4 refers to upper cervical spinal cord injuries of level four.

Using the NSCISC life expectancy calculator, life expectancies were calculated for a 25-year old white male and female with complete C1-4 level SCI and are displayed in Table 2.3.(54) Compared with the general population, ventilator dependency decreased the life expectancy by 38.6 years and 43.4 years for the example male and female with C1-C4 complete SCI, respectively.

Table 2.3: Sample life expectancies for 25-year-old white male and female with complete C1-4 level SCI generated using the NSCISC life expectancy calculator.(54)

Group	Life expectancy (years)
General population estimate male	51.1
C1-C4 complete SCI male, no ventilator	24.73
C1-C4 complete SCI male, with ventilator	12.51
General population estimate female	55.9
C1-C4 complete SCI female, no ventilator	27.44
C1-C4 complete SCI female, with ventilator	12.51

Note: Injury date selected was 1 January 2022; Current level of education chosen was High School graduate; Type of insurance chosen was Medicare/Medicaid/Other; Cause of SCI for non-ventilator option was motor-vehicle accident. C1-4 refers to upper cervical spinal cord injuries of levels one to four.

There was little difference in life expectancy estimates between the samples published by Strauss et al. in 2006 and the updated life expectancy calculator of the NSCISC. The updated calculator estimates include patient data with follow-up through to January 2022 and age-specific standardised mortality ratios used by the NSCISC since 2016. Life expectancy was reduced by approximately 50% for both males and females with C1-C4 complete SCI who did not require ventilator support, compared with the general population. The requirement for ventilator support considerably reduced the life expectancy estimates for both males and females, reducing it by approximately 75% compared with the general population (Table 2.3).

2.4. Epidemiology of SCI in Ireland

2.4.1. Methods

The National Spinal Injuries Unit (NSIU) in the Mater Misericordiae University Hospital (MMUH), Dublin is responsible for all patients who have a traumatic injury to the spinal cord in Ireland. In an average year approximately 700 patients are referred to the service from hospitals around the country. There are other units in Ireland (Beaumont Hospital, University Hospital Galway and Cork University Hospital) that treat patients with traumatic SCI, though the numbers treated here are much smaller. The NSIU provides specialised, coordinated, interdisciplinary, medical and surgical care to patients with spinal cord injuries.(56) A descriptive analysis of demographic, clinical and administrative data on discharges from the NSIU was undertaken using Microsoft Excel 2013 and R-4.2.1. These data pertain specifically to patients aged 15 years and older with C1-C4 traumatic spinal cord injuries (TSCI) discharged between January 2008 and December 2021, inclusive (n=226). Additional data were available up to October 2022 and results for this 10 month period are presented for some variables (n=14). These data concern a subset of all traumatic SCI in Ireland as they are limited to one tertiary treatment unit. Also, details of inpatient deaths were not available in the dataset received so it is possible that some discharges may represent deaths (the numbers of which are expected to be small). Whilst this HTA is primarily concerned with spinal cord injuries of C1-C3 due to the higher likelihood of the need for permanent invasive ventilation in this cohort, data for C4 injuries are presented as invasive ventilation can sometimes be required in this cohort of patients also. Data presented in this Chapter relate to: number of cases, sex, age, aetiology, geographical residence, length of hospital stay and level of injury. For each of these variables, data from the NSIU are presented followed by a brief overview of related academic literature findings.

2.4.2. Incidence

A total of 226 patients with traumatic SCI C1-C4 were recorded as discharged from the NSIU between 2008 and 2021, with a further 14 discharges recorded between January and October 2022.

Table 2.4 displays the incidence rates and the number of cases of traumatic SCI C1-C4 discharged from the NSIU from 2008 to 2021. The population denominator is derived from Census data. The census takes place every five years with results for intervening years based on population and migration estimates.(57) The table shows the population numbers largely increasing year on year. Incidence of C1-C4 discharges from the NSIU fluctuated from 2.7 to 3.6 cases per million population over the 14 year period (with the highest recorded in 2013 at 5.9 and the lowest in

2010 at 2). The absolute number of discharged cases varied from 12 in 2008, 27 in 2013 and 18 in 2021. Overall, C1-C4 traumatic SCI is rare in Ireland and numbers fluctuate from year to year.

Table 2.4: Incidence of traumatic spinal cord injuries C1-C4 (2008-2021)

Year	Population	Number of TSCI C1-C4	Incidence (Per million Population)
2008	4,422,100	12	2.7
2009	4,459,300	13	2.9
2010	4,470,700	9	2.0
2011	4,588,252	16	3.5
2012	4,585,400	17	3.7
2013	4,593,100	27	5.9
2014	4,609,600	13	2.8
2015	4,635,400	15	3.2
2016	4,761,865	10	2.1
2017	4,792,500	18	3.8
2018	4,857,000	15	3.1
2019	4,921,500	25	5.1
2020	4,977,400	17	3.4
2021	5,011,500	18	3.6

Figure 2.3 shows the number and dispersion of patients with levels of injury C1-C4, C1-C3, and C4 over the 14 year period under review. In 2008 the number of C1, C2 and C3 cases (blue line) combined was six, as was the total number of C4 cases (red line) that year. Across the 14 year period the numbers remain relatively consistent, albeit with fluctuations. For example, cases of C1-C3 rise from six in 2008 to 17 in 2013 and 11 in 2021. Cases of C4 rise from six in 2008 to 12 in 2018, decreasing to seven in 2021.

Figure 2.3 Number of cases of patients with C1-C4 spinal cord injuries discharged from the National Spinal Injuries Unit (2008-2021)

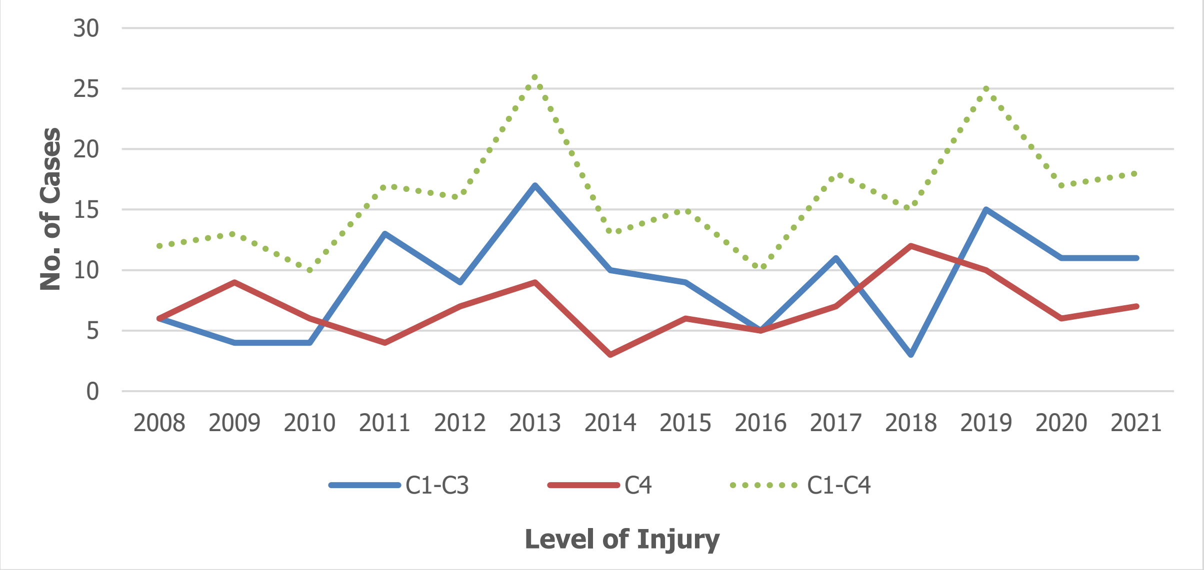
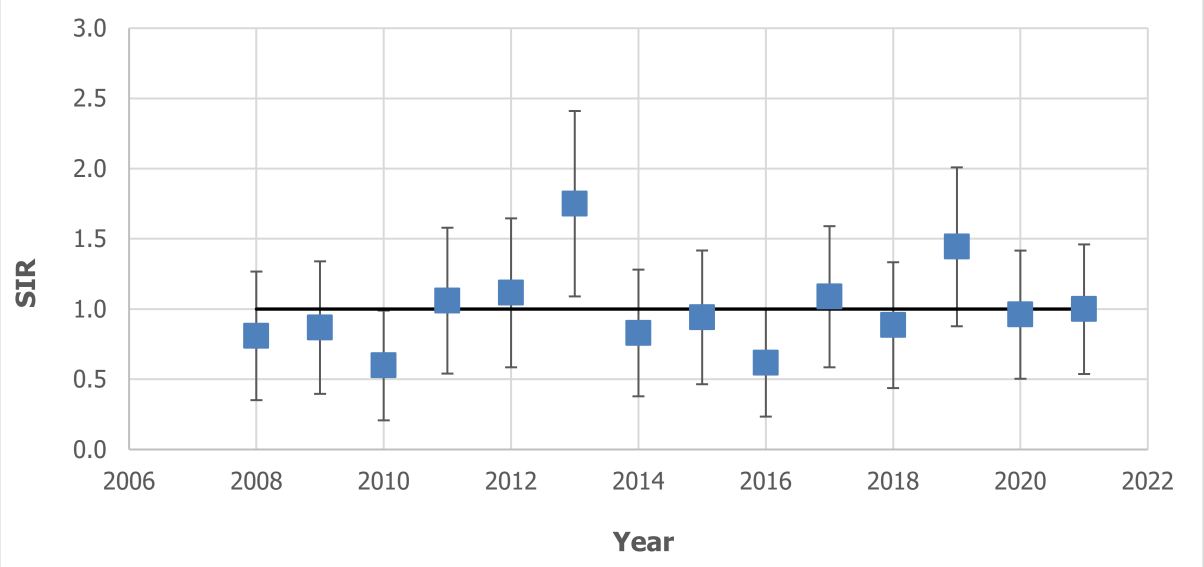


Figure 2.4 presents the age standardised incidence ratio (SIR) for each year of data; this figure shows that variation in incidence over time may be influenced by changing demographics. The SIRs were calculated using the age-specific averages across the time period (as reported for eight age bands) to estimate the annual expected incidence. The age standardised incidence ratio has fluctuated between 2008 and 2021, but with few exceptions the confidence intervals for the yearly SIR have included 1, suggesting no difference from the average over that time.

Figure 2.4 Age standardised incidence ratio of C1-C4 spinal cord injuries discharged from the National Spinal Injuries Unit (2008-2021)



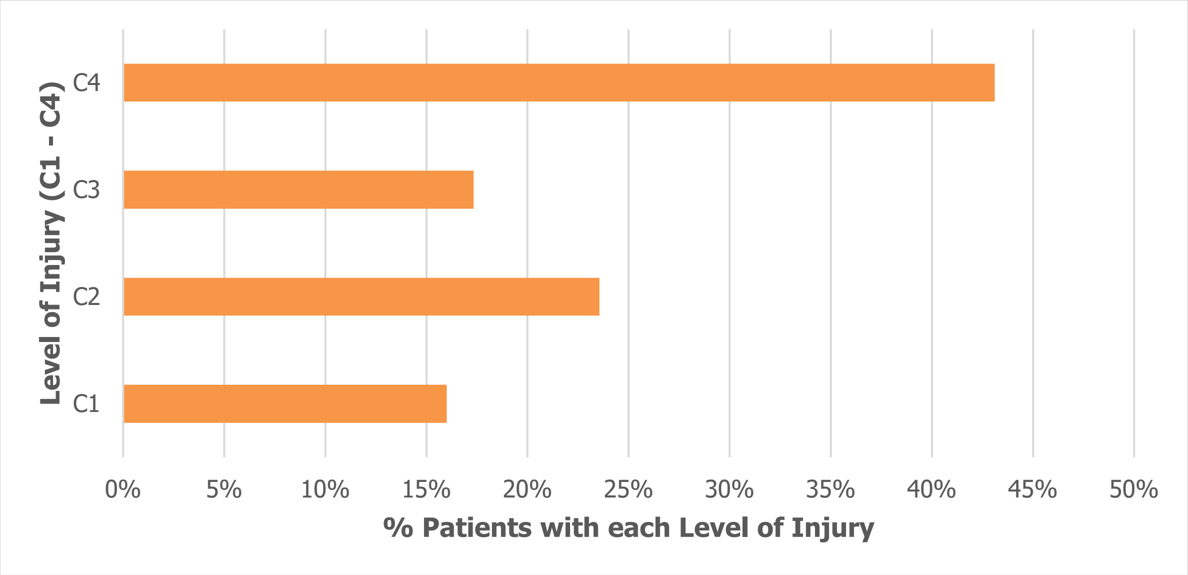
Notes: the expected incidence was calculated using average age-specific incidence over the full time period applied to annual population estimates.

In 2019, Smith et al. published results of their prospective epidemiological study on traumatic SCI in Ireland throughout 2016. They reported 61 cases of traumatic SCI in Ireland, corresponding to an overall incidence of 12.8 cases per million persons. Of these 61 cases, 16 (26.2%) were recorded as having upper cervical (C1-C4) lesions.(58) In 2018, the authors reported similar findings for the incidence of traumatic SCI in Ireland between 2010 and 2015 (n = 347 cases; overall mean incidence 12.6 (range 11.5 – 13.3) per million persons per year).(59) These findings from Ireland indicate a lower incidence to that of the global incidence of traumatic SCI. A systematic review of the global incidence of traumatic SCI reported the range in incidence was from 8.3 to 150.5 cases per million based on national studies since 2000.(60) An overall estimate for the global incidence of traumatic SCI was previously reported as 23 cases per million persons (95% CI, 13 – 30) as part of an initiative by the International Spinal Cord Society (ISCoS) Prevention Committee.(61) A similar incidence was reported in another report on the epidemiology of traumatic SCI in a population of 4.9 million inhabitants in Northern Italy between 2011 and 2020. The authors estimated an incidence rate of 26.5 cases per million persons (95% CI, 25.0 – 27.9).(62) Of the 1,303 cases of traumatic SCI, approximately a third of cases had high cervical (C1-C4) lesions (n = 389/1,303, 29.9%), while all cervical lesions represented a majority of cases (n = 679/1,303, 52.1%).

2.4.3. Level of injury

Figure 2.5 shows that the most common traumatic SCI level for all years combined, for both males and females, was C4 (43%, n=97), followed by C2 (24%, n=53) and C3 (17%, n=39), with C1 (16%, n=36) being the least common of the four injury levels under review.

Figure 2.5: Percentage of patients with C1-C4 spinal cord injuries according to level of injury (2008-2021)



According to a 2018 report describing cases of traumatic SCI in Ireland between 2010 and 2015, 59% (n = 204/347) of cases had tetraplegia.(59) The majority of these cases had incomplete tetraplegia (n = 149/204, 73%), with approximately a quarter having a complete lesion (n = 55/204, 27%). Data from Northern Italy reported the number of cases by level of injury.(62) The most frequent level of injury in their cohort spanning 10 years was a high cervical (C1-C4) lesion (n = 389/1,303, 29.9%).(62) A low cervical (C5-C7) lesion was recorded for 22.3% (n = 290/1,303) of cases, 21.7% (n = 283/1,303) had a thoracic lesion, while 26.2% (n = 341/1,303) had a lumbar or sacral lesion. In addition, a high cervical lesion was the most frequent injury level among patients that died in the first year after injury (n = 96/224, 42.9%), followed by a low cervical lesion (n = 60/224, 26.8%).(62) A 2016 retrospective cohort study reported that 60.6% (n = 993/1,638) of cases of traumatic SCI in Scotland over a 20-year period had cervical lesions. The authors reported an increase in upper cervical injuries from 21.7% between 1994 and 1998 to 31.2% between 2009 and 2013.(63)

2.4.4. Sex

Males accounted for 82% (n=185) of NSIU discharges across the four injury levels under review from 2008 to 2021. Figure 2.6 displays the annual proportion of males and females discharged from the NSIU. The chart illustrates that traumatic SCI are predominantly a male phenomenon, with males accounting for all discharges from the NSIU in the years 2012 and 2016 (n=16 and n=10 respectively).

Figure 2.6: Percentage of males and females with C1-C4 spinal cord injuries (2008-2021)

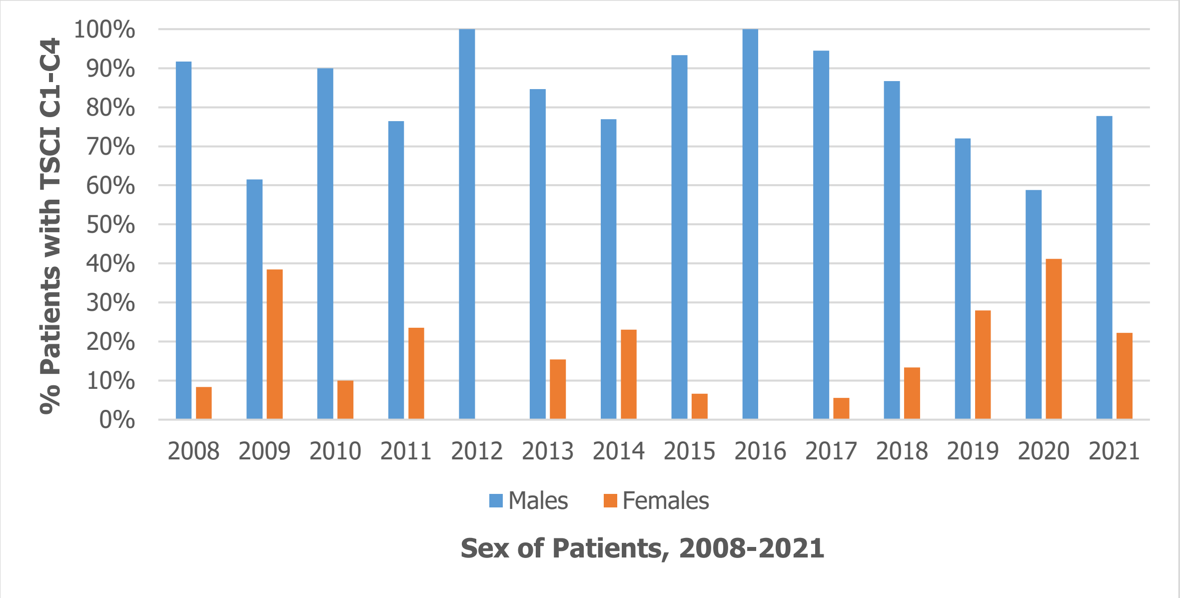
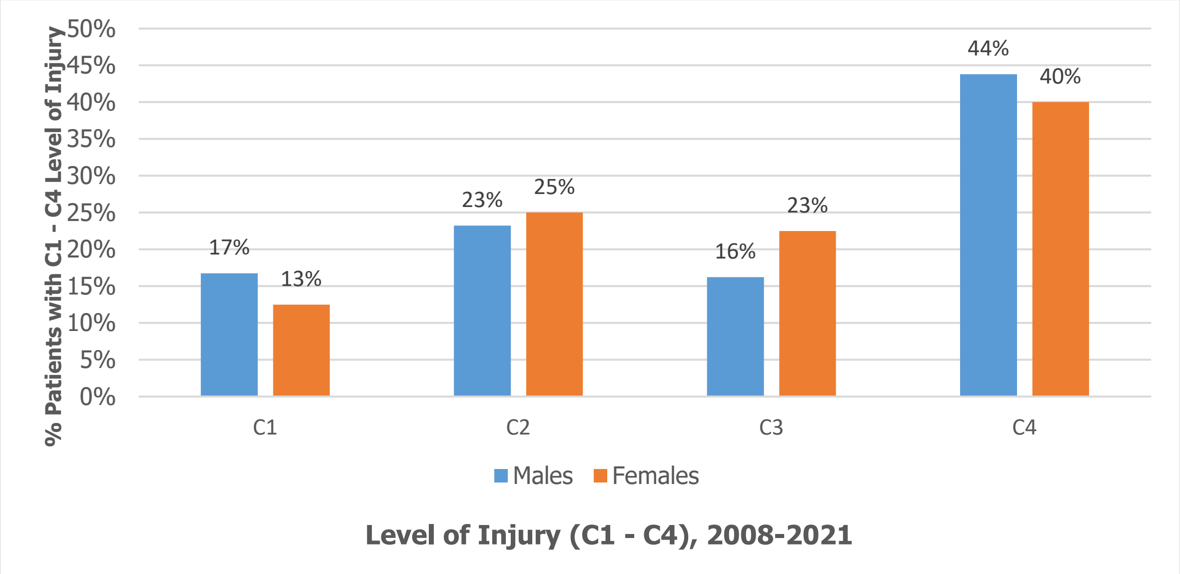


Figure 2.7 displays the breakdown of injury level for males and females for 2008-2021. For both males and females, C4 was the most common level of injury. 44% of males (n=81) and 40% of females (n=16) had a C4 injury. The least common injury level for males was C3 (16%, n=30) and for females it was C1 (13%, n=5). Findings show that both sexes experience similar levels of injury.

Figure 2.7: Percentage of males and females by level of C1-C4 spinal cord injuries (2008-2021)

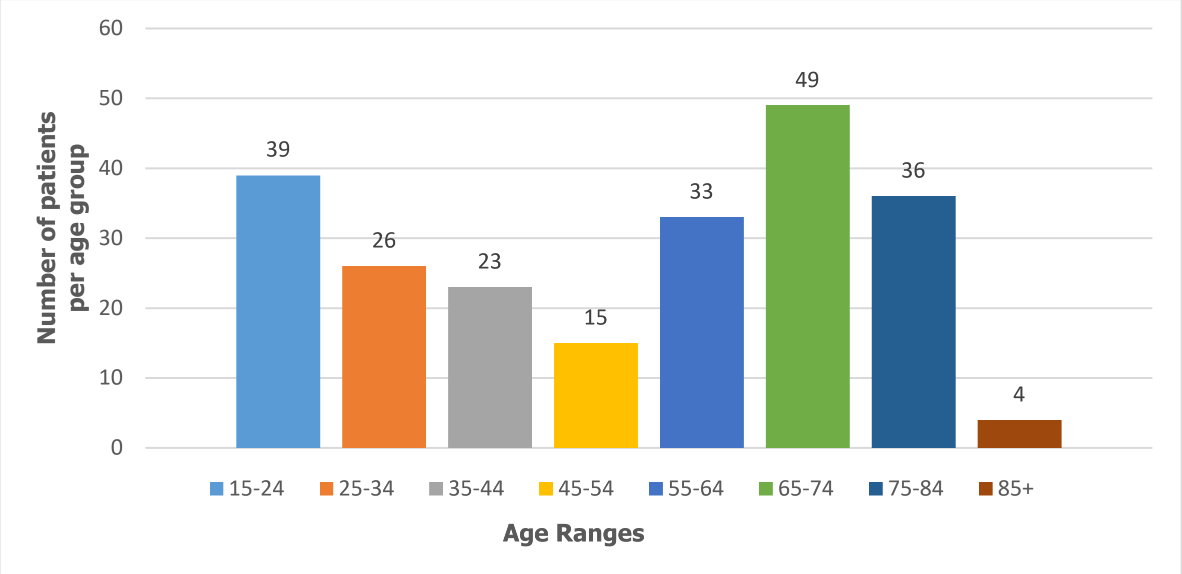


A 2018 report from Ireland noted that 71.5% (n = 248/347) of SCI patients discharged from hospital in Ireland between 2010 and 2015 were male.(59) A similar but slightly lower trend of male case predominance was reported in Northern Italy in 2022. Over their 10-year study period, the majority of patients were male (n = 890/1,303, 68.3%), with an overall male to female ratio of 2.3:1. It was noted that this ratio decreased slightly with age, albeit remaining sizeable at 2:1.(62)

2.4.5. Age

Figure 2.8 displays that over the study period of 14 years (2008-2021), the most common age group at time of admission to the NSIU was those aged 65-74 years (22%, n=49) (Figure 2.8). The 2nd most common age group at admission was 15-24 year olds (17%, n=39). Those with an admission age of 85 and over accounted for 2% (n=4) of cases discharged from the NSIU during this time period. While cases in those aged 65 years and older are an increasing proportion of incidence cases (Chi-squared test for trend, p=0.002), it is unclear whether this is driven by increases in incidence in over 65s, reducing incidence in under 65s, or demographic changes. Due to the small numbers of cases involved, it is challenging to determine the causes of apparent trends.

Figure 2.8: Age groups of patients with C1-C4 spinal cord injuries at time of admission (2008-2021)



In Ireland between 2010 and 2015, it was reported that the mean age of traumatic SCI cases ranged from 44.1 years (SD 19.3) in 2010 to 52.6 years (SD 20.1) in 2014.(59) In a prospective study of all new cases of traumatic SCI in Ireland during 2016, the authors reported the mean age of cases as 52.8 years (SD 19.9).(58) The authors provided the number of cases of traumatic SCI by 15-year age groupings;

the most numerous cases occurred in the 46-60 year old group (n = 15/61, 24.6%) and 61-75 year old group (n = 16/61, 26.2%). There was a considerably higher incidence of traumatic SCI in older age groups compared with younger age groups. This was especially apparent among females, with an incidence rate of 4.5 cases per million among 46-60 year olds and rates of 20.2 and 28.9 cases per million persons among the 61-75 year olds and 76 years and older age groups, respectively.

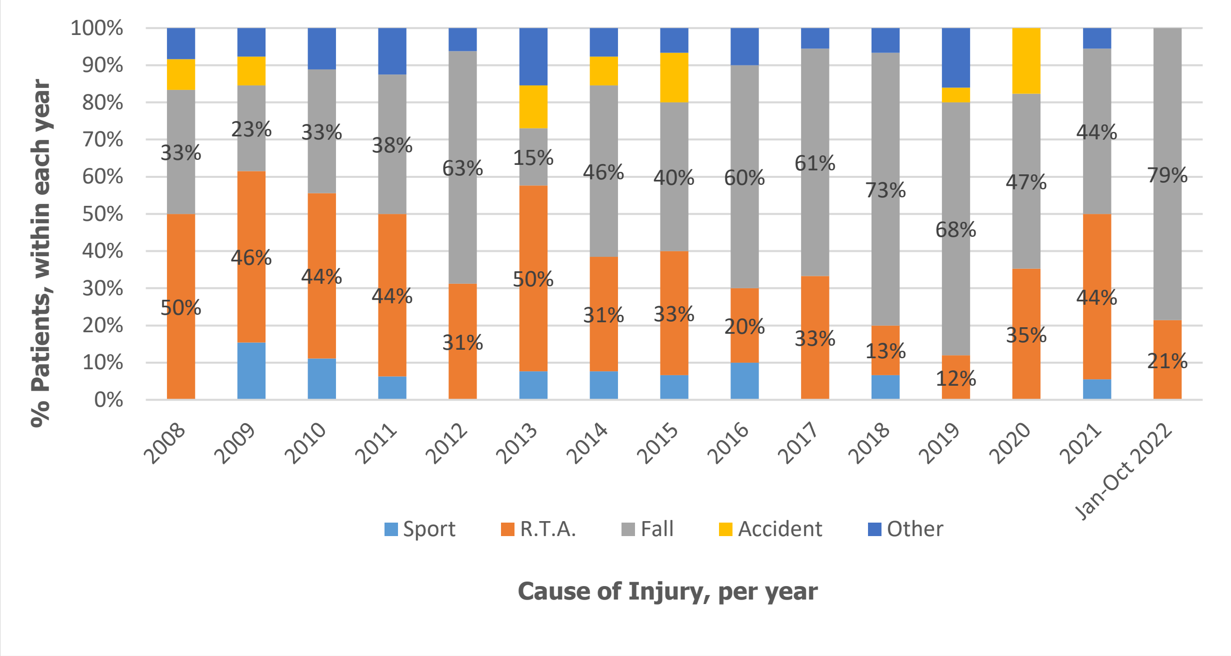
In Northern Italy, the overall mean age of patients with traumatic SCI was 59.2 years (SD 21.4).(62) In addition, the mean age was significantly higher among those who died in the first year after injury compared with those who survived that period (76.6 years (SD 15.0) and 55.5 years (SD 20.7), respectively; $p < 0.0001$). The authors observed a significant trend of increasing incidence of traumatic SCI with each incremental 10-year age group ($p < 0.0001$) from the 25-34 year group (16.5 cases per million persons, 95% CI 13.4 – 20.4) through to the 85 year and older group (87.0 cases per million persons, 95% CI 73.6 – 102.8).(62) Similarly, data from Scotland demonstrated a trend of the age profile of patients with SCI increasing over time; the mean age at injury increased from 44.1 years for the period 1994 to 1998 to 52.6 years for the period 2009 to 2013 ($p < 0.001$).(63) Additionally, a significant increase in the age profile of SCI was noted for patients in the age ranges from 51 years to 85 years ($p = 0.03$). Relatedly, the mean age of patients with a traumatic SCI due to falls (54.2 years) was significantly higher than the mean age of patients who experience SCI due to causes other than falls (39.5 years) ($p < 0.0001$). A similar association was reported in Spain between 2001 and 2015.(64)

2.4.6. Cause

NSIU data categorised causes of injury under five major headings: Sport, Road Traffic Accident (RTA), Fall, Accident, and Other. Figure 2.9 provides a breakdown of the proportions of injuries attributed to each cause for each year of study (including the partial year of data for January to October 2022). The chart illustrates that the main causes of traumatic SCI in each year were RTAs and falls; these accounted for 80 (34%), and 114 (48%) of cases, respectively, among 237 patients admitted between 2008 and 2022 (n=223 for 2008-2021 and n=14 for January to October 2022).

In 2008, 50% (n=6) of C1-C4 traumatic SCI were due to RTAs and 33% (n=4) were due to falls. RTAs also caused the highest proportion of TSCI in 2009, 2010 and 2011. From 2014 this trend changed, with falls accounting for the greatest proportion each year to 2022, (apart from in 2021 when both falls and RTAs accounted for 44% (n=8) of injuries each). From January to October 2022, falls accounted for 79% (n=11) of injuries whilst RTAs accounted for 21% (n=3).

Figure 2.9: Cause of injury in patients with spinal cord injury C1-C4 (2008-2022)



Between 2010 and 2015 in Ireland, it was previously reported that falls accounted for the majority of cases of traumatic SCI (n = 185 / 347, 53.3%), with transport related causes being the second of frequent cause (n = 70 / 347, 20.2%).(59) In 2016, falls remained the most common cause of traumatic SCI (n = 37/ 61, 60.7%), followed by sports and leisure activities (n = 12 / 61, 19.7%).(58)

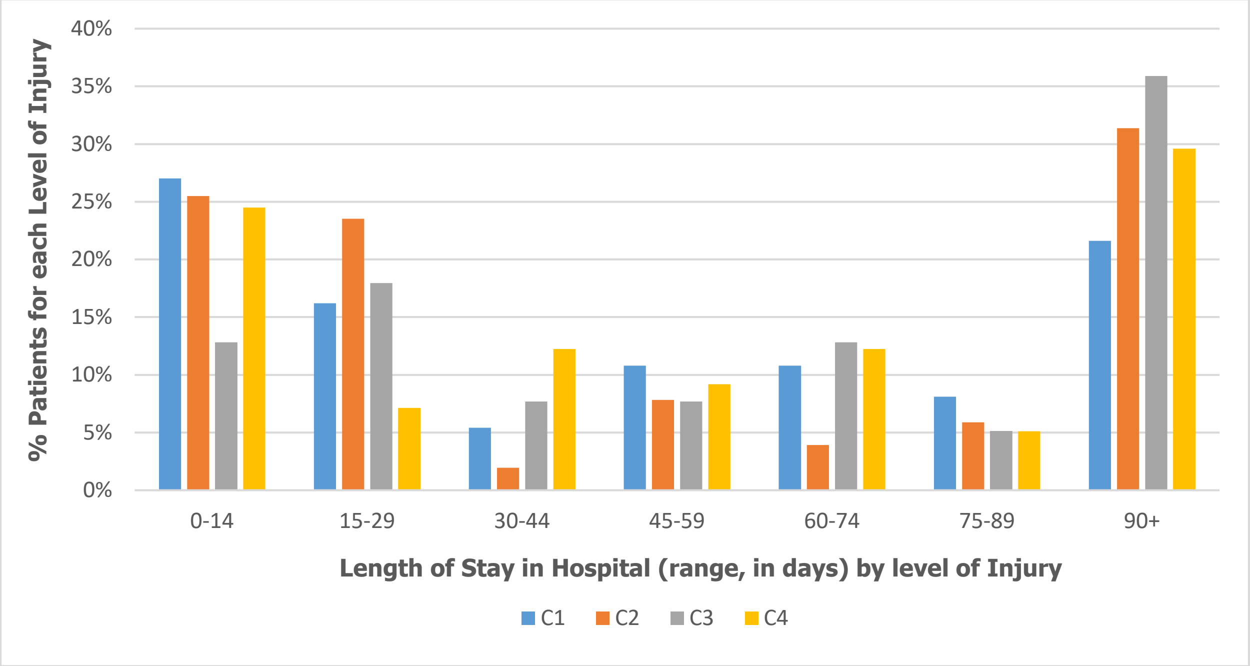
Globally, road traffic accidents and falls have been reported to be the most common cause of traumatic spinal injury.(65) Previously, it was reported that the proportion of traumatic SCI resulting from transport was stable or decreasing in high-income countries but on the increase in low- and middle-income countries.(66) A 2022 systematic review reported that the pooled incidence of traumatic SCI in low- and middle-income countries was 22.6 cases per million persons per year (95% CI, 13.5 – 37.6). Transport accounted for 43.2% (95% CI, 37.8 – 48.6) of cases while 34.2% (95% CI, 44.2 – 56.9) were caused by falls.(67) In Northern Italy between 2011 and 2020, transport was reported as the most frequent cause of traumatic SCI cases (n = 389/1,303, 29.9%).(62) In Scotland, the most common cause of traumatic SCI over a 20-year period was falls (51.7%). The proportion of cases caused by falls increased during the study period, from 41.6% between 1994 and 1998, to 60.0% between 2009 and 2013.(63)

In the United States, traumatic SCI caused by violence, in particular gunshot wounds, account for a considerable proportion of cases, in addition to injuries arising from falls and transport.(29) According to data published in 2021 by the National Spinal Cord Injury Statistical Centre (NSCISC) spanning from 1972 to 2021, transport accounted for 41.8% (n = 14,897/35,675) of SCI cases, and falls resulted in 23.2% (n = 8,263/35,675) cases, while 17.0% (n = 6,068/35,675) cases of SCI were caused by violence. Of the SCI cases resulting from violent causes, the majority were a result of gunshot wounds (n = 5,437/6,068, 89.6%).(29) Between 1972 and 2021, the proportion of SCI due to falls increased from 16.5% (n = 752/4,563) to 31.3% (n = 1,473/4,703), respectively. Transport-related causes remained the most common cause of SCI during this time period, accounting for 37.5% (n = 1,765/4,703) between 2015 and 2021.(29)

2.4.7. Length of stay

Figure 2.10 displays the length of stay in days (range) for these patients in the Mater Misericordiae University Hospital (MMUH), per level of injury. It is important to note that the data do not differentiate between inpatient deaths and discharges. It is possible that some cases with shorter lengths of stay in particular, may represent inpatient deaths. Information regarding where the patient was discharged to is also not available. Between 2008 and 2021, 37 patients with a C1 level injury were discharged from the NSIU. Of these, 27% (n=10) spent 0-14 days and 22% (n=8) spent 90 or more days in hospital. Of the 51 patients with a C2 level injury, 25% spent 0-14 days (n=13) and 31% spent 90 or more (n=16) days in hospital. Of the 39 patients with a C3 level injury, 13% (n=5) spent 0-14 days and 36% (n=14) spent 90 or more days in hospital and of the 98 patients with a C4 level injury 24% (n=24) spend 0-14 days and 30% (n=29) spent 90 or more days in hospital.

Figure 2.10: Length of stay (days) by C1-C4 injury level (2008-2021)



Between 2010 and 2015 in Ireland, the median rehabilitation length of stay for patients with traumatic SCI ranged from 83.5 days (IQR 63.5) in 2011 to 98.5 days (IQR 58.8) in 2014.(59) The authors reported a significant association between the length of stay and level and completeness of injury; the length of stay was significantly longer for those with complete tetraplegia (median 148 days, IQR 91) compared with those with incomplete tetraplegia (median 86 days, IQR 72; $p < 0.001$).(59) In Northern Italy, the mean length of stay for patients, excluding days spent in rehabilitation departments, was reported as 20.4 days (SD 20.4). There was a significant difference ($p < 0.0023$) in length of hospital stay between patients who died in the first 12 months (25.9 days, SD 25.0) and those who survived to 12 months (19.2 days, SD 19.0).(62)

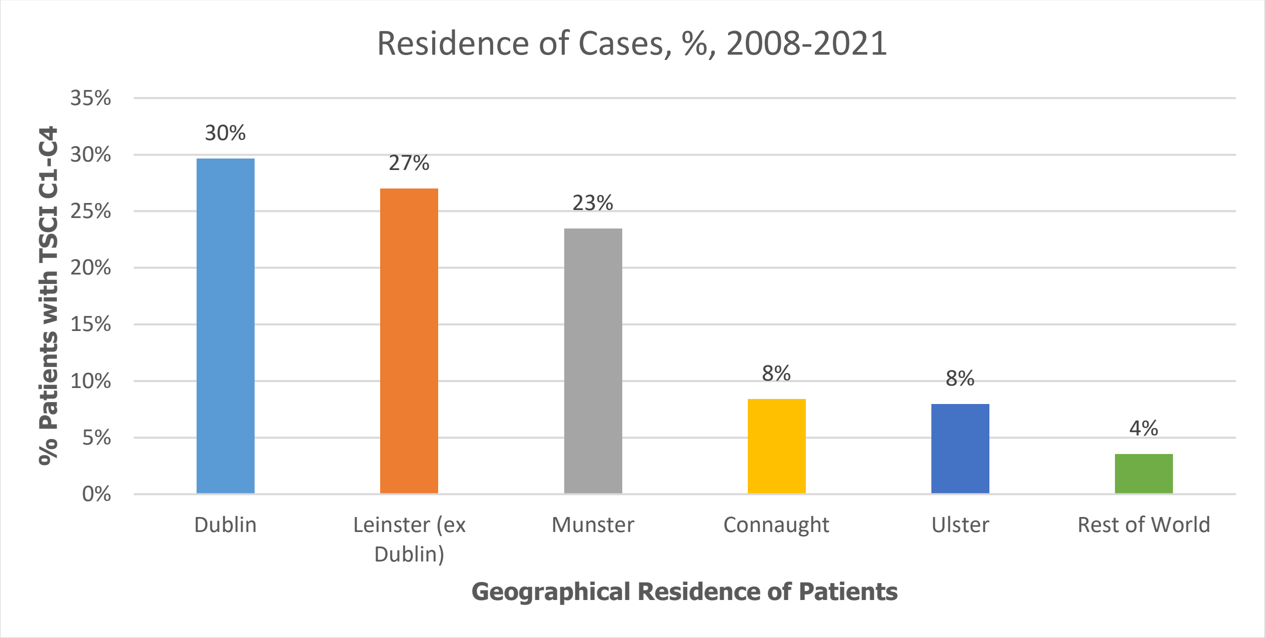
According to data published by the NSCISC for patients in the United States, the median rehabilitation length of stay has decreased for patients with SCI since the 1970s.(29) Overall, the patient group with the largest median number of days hospitalised in the rehabilitation unit was patients with complete tetraplegia, ranging from 122 days between 1972 and 1979, to 68 days between 2010 and 2014 and 58 days between 2015 and 2021.(29)

2.4.8. Residence

Residence refers to the location of the patient's home. Data in Figure 2.11 shows that cases are concentrated in Dublin (30%, n=67) as well as the wider province of Leinster (excluding Dublin) (27%, n=61), followed by Munster (23%, n=53), Connaught (8%, n=19), Ulster (8%, n=18 (three counties)) and Rest of World (4%, n=8). 'Rest of World' refers to patients from Northern Ireland, the United Kingdom, Europe and North America.

These percentages are broadly in line with population proportions in the provinces. Census 2016 results show that Leinster (Dublin included) accounted for 55.3% of the total population, Munster 26.9%, Connaught 11.6% and Ulster 6.2% (three counties).(68)

Figure 2.11: Residence of patients with spinal cord injury C1-C4 (2008-2021)



2.4.9. Ventilator dependence

The NSIU reported on at least eight patients who were dependent on a ventilator at discharge between 2008 and 2022: these included three with a C1 level injury, three with a C2 and two with a C4 injury. This equates to approximately 3% (N=8/240) of all patients with C1-C4 SCI discharged from the NSIU during this period. All of these patients were male, the age ranged from 19 to 75 (median age, 53), and the length of stay ranged from 2 to 749 days (median length of stay, 290 days). It is important to note that information regarding permanent mechanical ventilation requirements had to be extracted manually from patients' medical charts by the Data Management Department in the NSIU, and so it is possible that some ventilator-dependent patients were missed. Expert clinical opinion is that in Ireland there are currently between five and ten adults with SCI receiving invasive mechanical ventilation at home, with none, one or two new patients expected every year.(69)

In 2021, the NSCISC published data on ventilator dependence at admission and discharge for patients with SCI. Information was available at admission to rehabilitation for 18,272 patients with tetraplegia, and for 18,679 such patients at discharge.(29) There were 19.4% of patients with tetraplegia who required the use of a mechanical ventilator at admission, compared with 5.5% at time of discharge. There was considerable intersystem variability observed for the reporting systems; the proportion of patients with tetraplegia who required a ventilator at discharge was noted to vary from 0.0% to 16.9%. Ventilator dependence was lower at both time points for patients with paraplegia, with 5.3% (n = 799/15,203) and 0.4% (66/15,347) requiring ventilator use at admission and discharge, respectively.(29) In addition, the proportion of persons who required mechanical ventilator support at one-year post-injury was reported as 3.4% (n = 327/9,511) and 0.2% (n = 19/8,799) for those with tetraplegia and paraplegia, respectively.(29)

2.5. Discussion

Damage to the spinal cord can result in catastrophic and life-threatening injuries, which can lead to the affected person suffering from significant long-term morbidity and requiring life-long care.(15) The higher up the spinal cord that the injury occurs, the more extensive the range of impairments will generally be. Patients with complete C1-C3 level cervical lesions will experience the most severe level of impairment and tend to require a ventilator and tracheostomy to breathe. While there are many complications associated with cervical SCI, specific areas of management include VTE prophylaxis, neurogenic and spinal shock, SCI-induced bradycardia, respiratory complications, acute autonomic dysreflexia, pressure injuries, neurogenic bowel and bladder dysfunction, mental health issues, and pain.(13, 15) Respiratory complications have been reported to be the most significant cause of morbidity and mortality in the acute period post-injury for patients with acute cervical SCIs.(4, 32, 34)

Although they have severe consequences to the person's life, SCI are rare. Recent reports indicate that the overall incidence of SCI in Ireland are lower than the overall estimate of the global incidence of SCI.(58, 59, 61) These values represent all levels of injury to the spinal cord, from incomplete paraplegia to complete upper cervical tetraplegia. Both in Ireland and internationally, tetraplegia was reported to be the most frequently presenting injury level.(29, 59, 62) Upper cervical tetraplegia was reported to account for a larger number of cases than lower cervical tetraplegia.(59, 62) Within cases of upper cervical tetraplegia, it was noted that in Ireland from 2008 to 2021, C4 level injuries were the most frequent injury type. As such, it is important to note that the number of patients with C1-C3 level injuries represents a small fraction of all cases of SCI. In addition, the number of patients with C1-C3 SCI requiring permanent mechanical ventilation and whose condition is considered clinically appropriate to be discharged home with a domiciliary ventilation package of care, will be a minority of this patient group.

Permanent ventilator assistance is usually, but not always, required for complete SCI at C1-C3 levels due to diaphragmatic paralysis.(13) Patients with C3-C4 injuries experience diaphragmatic dysfunction, but they may retain partial ventilatory function.(15) Initial temporary ventilation with subsequent weaning may be achieved in those with complete SCI at C3. In the absence of other factors that would prevent ventilatory weaning, complete injuries at C4 and below, and incomplete SCI at any level, are usually associated with successful weaning.(31) Apart from completeness and level of injury, other barriers to weaning include older age, smoking history, active pneumonia, the severity of impaired consciousness, high level injury severity score, underlying cardiorespiratory diseases and other complicating comorbid conditions.(30, 32-34)

Overall, the life expectancy and survival of persons with SCI have improved over recent decades in high-income countries, but these individuals are still more likely to die prematurely compared with the general population.(4, 45) These improvements have been reportedly associated with improved management outcomes for secondary conditions such as urologic complications.(4) However, it has been reported that respiratory management outcomes have not significantly improved during this period of time, with pneumonia and respiratory failure remaining a significant cause of death for these patients.(50) The factors that have been reported to impact the life expectancy of patients with SCI include age, sex, level and completeness of injury, and ventilator dependency.(4, 21, 45, 46) Data from the NSCISC in the US indicated that ventilator dependency approximately halved the life expectancy of a person with a upper-cervical SCI compared to that of a similar case without ventilator-dependency.(54)

The international literature reported trends of increasing age among patients with SCI overall.(62, 63) In Europe and the US, the observed tendency towards older mean age among patients with SCI has been suggested to be driven by demographic changes, such as people living longer, and associated changes in the aetiology of SCI.(63, 64, 70) In recent years, the proportion of SCI caused by falls has increased, and the mean age of patients with SCI caused by falls has increased, with falls being the most common cause of SCI in older patients.(63, 64, 70) Older patients may be at greater risk of serious spinal injury from falls due to osteoporosis, a narrow spinal canal, sensory deterioration and risk of comorbidities from medication use.(64) The Irish data from the NSIU from 2008 to 2021 demonstrated that the 65-74 year age group was the most common age group at time of admission. According to Irish census records, between 2011 to 2016, the number of people in Ireland aged 65 and older increased by over two times those aged 15-64, indicating the possibility of a similar trend in Ireland.(71) Older patients with traumatic SCI have been found to have a higher risk of mortality and complications, which may impact a clinical assessment of the appropriateness of domiciliary ventilation. This indicates the need for consideration of short- and long-term management strategies for patients, especially older patients, given these recent trends in the aetiology of SCI.

2.5.1. Strengths and limitations

A strength of the epidemiological analysis presented within this Chapter is that the underpinning data comprise the totality of C1-C4 discharges from the NSIU since 2008. Having 14 years of data may enable trends to be observed and allows for consideration of the wide range of patients with SCI who may require permanent mechanical ventilation. An important limitation of the NSIU epidemiological data is that these represent a subset of all traumatic SCI in Ireland as it is limited to one tertiary treatment unit, though it is the largest unit in the country. There are other centres in Ireland (Beaumont Hospital, University Hospital Galway and Cork University Hospital) that treat patients with traumatic SCI, though the numbers treated here are much smaller.

Another limitation is that there are no data regarding the completeness of injury for these patients (only the descriptor of C1-C4). In addition, the analysis refers to 'discharges' although an unknown, but small number of these patients will have died in hospital. Finally, owing to missing data for some variables, the total number of cases for each outcome ranges from n=226 to n=223 (2008-2021) and from n=240 to n=237 (2008-2022).

2.5.2. Conclusion

Spinal cord injuries are associated with significant mortality and morbidity. The higher up the spinal cord, and the more complete the injuries are, the more severe the symptoms will be. However traumatic SCI leading to C1-C3 injuries are rare, with the requirement for permanent mechanical ventilation even rarer still. Furthermore, those who are clinically suitable for home domiciliary ventilation again represent a subset of these. These injuries predominantly affect males and historically predominated in younger age groups. Data from Ireland and internationally indicate that the incidence of SCI in older age groups is increasing, largely driven by falls in these older individuals. This may have important implications for service provision going forward.

3. Description of the technology and outcomes

Key points

- Traumatic injury to the cervical spine is associated with variable degrees of respiratory dysfunction, dependent on the level and completeness of injury; ventilatory support is often required, at least in the short term, but can be a lifelong requirement in a minority of individuals.
- There are two main types of mechanical ventilation – invasive and non-invasive. Invasive ventilation is generally defined by the requirement for the insertion of an artificial airway, usually tracheostomy in the larynx region, while non-invasive ventilation (NIV) generally involves applying tightly fitting face or nasal masks without any artificial airway. Invasive mechanical ventilation rather than NIV is generally necessary for patients with complete high cervical SCI, particularly during the initial management phase, due to the lack of neural control of breathing.
- An alternative intervention to mechanical ventilation is 'phrenic nerve stimulation', also called 'diaphragm pacing', which involves the surgical implantation of an electrode around the phrenic nerve(s), connected to a radio-frequency receiver usually implanted in the chest wall. Phrenic nerve stimulation provides electrical stimulation of the phrenic nerve which results in diaphragmatic contraction.
- Secretion management is crucial in patients with SCI who are invasively ventilated; this is due to the inability of these individuals to clear secretions on their own. Airway clearance is important in order to prevent atelectasis (that is, a complete or partial collapse of the lung) and respiratory infections. Effective secretion management can be achieved through the use of various techniques (for example, air stacking and glossopharyngeal breathing) and equipment (for example, mechanical insufflation / exsufflation (MIE) device).
- Besides respiratory failure, other clinical conditions that impact significantly on patients with high cervical SCI include autonomic dysreflexia, pain, anxiety/depression, neurogenic bladder and bowel dysfunction and pressure injuries. Providing appropriate bladder, bowel and skin care to these individuals is complex, lifelong and requires a range of health professionals.
- There are a range of different medical conditions that may lead to the requirement for long-term ventilation, such as amyotrophic lateral sclerosis

(ALS) which is a sub-set of motor neurone disease, chronic obstructive pulmonary disease (COPD), myasthenia gravis and SCI. Provision of mechanical ventilation services at home (henceforth called domiciliary ventilation) is the preferred option for many individuals who require ventilation for extended periods of time. The prevalence of domiciliary ventilation varies across and even within jurisdictions but overall the prevalence is generally low (population-based estimates in the literature range from approximately 0 to 30 per 100,000 population).

- International literature estimates suggest that patients with SCI account for a small proportion of all domiciliary ventilation service users, ranging from 2% to 19.9%, with higher proportions among invasively ventilated cohorts.
- Prevalence of domiciliary ventilation is increasing internationally, driven largely by the increased use of NIV. The use of invasive ventilation in the domiciliary setting is less common, but is more prevalent in patients with SCI.
- Although there are some data to support the safe and effective use of domiciliary invasive ventilation in other populations, data specific to the SCI population in particular are very limited. While domiciliary ventilation has been used and recommended for a range of conditions under specific circumstances, it is important to note that there are some safety concerns associated with the provision of such complex care in the home setting (for example, ventilation failure). However, from the patient’s perspective, there is often a preference to return home once the right supports and equipment are in place, even if this does entail some added risk.
- An integrated care pathway for patients with SCI has been developed in Ireland by the National Clinical Programme for Rehabilitation Medicine, in line with its model of care. This care pathway extends from pre-hospital care to discharge and lifelong care. However, there are challenges (such as funding and staffing) with the implementation of this care pathway, particularly at the discharge stage. As a result of these barriers, patients with SCI who are ventilator-dependent might have to remain hospitalised for a prolonged period of time, often several years, before being discharged home.

3.1. Introduction

The purpose of this Chapter is to describe the use of domiciliary invasive ventilation in adults with spinal cord injuries (SCI). A brief description of the management of adults with SCI is provided, followed by an outline of the use of domiciliary ventilation in various populations, and the outcomes associated with domiciliary ventilation. Finally, the current integrated care pathway for SCI in Ireland is described. For the purpose of this HTA, the health technology under consideration is the package of care provided to patients with high cervical SCI availing of domiciliary invasive ventilation.

3.2. Management of adults with spinal cord injuries

In the case of acute traumatic SCI, the initial recognition and treatment of the injuries is crucial to limiting secondary injury to the spinal cord and to providing patients with the best chance of functional recovery.(72) These individuals should ideally be treated in a Level 1 (Major) Trauma Centre.(73) Treatment goals for acute traumatic SCI include decompression of the neural elements, stabilisation of the spine, and maintenance of tissue perfusion in order to attain the best possible outcomes.(73) There is a risk of irreversible neural tissue injury, which, in the presence of multiple traumatic injuries, may include life-threatening abdominal and thoracic injuries; therefore, there is a requirement for complex decision-making in a timely manner. Timing of surgical care for spinal injuries depends not only upon early decompression to improve or prevent further neurological injury, but also on the need to first stabilise the individual's haemodynamics and or treat other life-threatening injuries.(15)

Non-traumatic SCI (that is, resulting from degenerative, neoplastic, vascular or other non-traumatic causes) is more insidious in nature and is increasingly being seen in rehabilitation centres.(73) The medical management of non-traumatic SCI depends on the underlying cause.(16) For example, if the cause is immunological, patients may be initiated on steroids followed by plasma exchange or intravenous immunoglobulin therapy. Long-term immunosuppression depends on the condition and risk of relapse. Tumours, haematomas, and degenerative conditions usually require surgery, although tumours may occasionally be managed with radiotherapy or chemotherapy, depending on their nature.(16)

Spinal cord injuries can lead to many long-term complications depending on the location and severity of the injury. Patients with high cervical SCI are particularly affected by many of these secondary complications, as previously discussed in Chapter 2. Specific areas of management that need to be considered for these individuals include, but are not limited to, venous thromboembolism (VTE) prophylaxis, neurogenic and spinal shock, SCI-induced bradycardia, respiratory complications, prevention and management of acute autonomic dysreflexia, spasticity, pressure injuries, neurogenic bowel and bladder, mental health issues, and pain.(15)

Rehabilitation post SCI forms a critical component of the individual's recovery.(74) The rehabilitation process for patients with SCI is prolonged and resource intensive.(74) Evidence supports the provision of early and intensive rehabilitation programmes to individuals affected by traumatic injury, as these have been shown to improve function, pain, quality of life and mental health outcomes, and to improve outcomes for carers.(75) For example, the National Institute for Health and Care Excellence (NICE) recommend that rehabilitation should be started as soon as possible after a traumatic injury, when the person is ready and able to engage and participate.(75) An interdisciplinary approach is essential in rehabilitation in SCI.(74) NICE recommend that the rehabilitation should be delivered by a multidisciplinary team with the appropriate expertise in rehabilitation after traumatic injury.(75) Rehabilitation for patients with SCI is therefore usually coordinated and delivered by a specialist team in a designated centre (for example, an SCI centre).(76) NICE further recommend that all people who have SCI should have *"a lifetime of personalised care that is guided by a SCI centre"*.(77)

While there are many complications associated with cervical SCI (see Section 4.3.5), there are several clinical issues that require specific consideration. These include respiratory failure, neurogenic bowel and bladder dysfunction and pressure injuries; the pathophysiology of which were outlined in Chapter 2 with the associated management discussed within the following sections.

3.2.1. Respiratory failure

There are three important aspects to the management of respiratory failure associated with SCI. These are ventilator management, phrenic nerve stimulation, and secretion management.

3.2.2. Ventilator management

A mechanical ventilator is a device that is used to provide positive pressure ventilation in order to help normalise an individual's arterial blood gas levels so as to maintain an adequate acid-base balance.(78) This is achieved though gas exchange by enabling the individual to inhale oxygen and exhale carbon dioxide, often through an artificial airway. Generally, individuals are on mechanical ventilation until they are able to achieve spontaneous breathing on their own. Mechanical ventilation (with or without oxygen administration) is indicated when spontaneous breathing is insufficient to sustain life, as is the case in high cervical SCI.(78, 79)

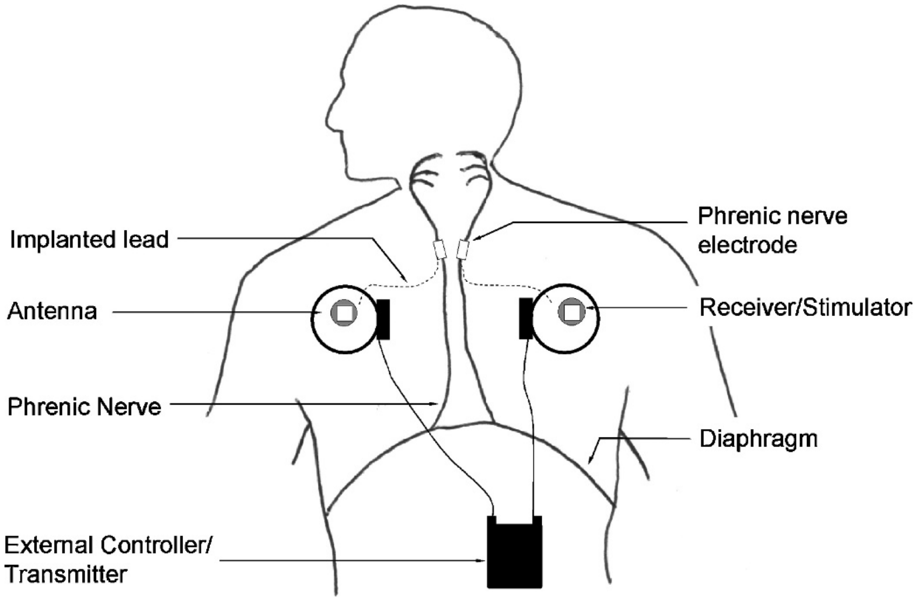
There are two main types of mechanical ventilation – invasive and non-invasive. Invasive ventilation is generally defined by the requirement for the insertion of an artificial airway, usually tracheostomy in the larynx region, while non-invasive ventilation (NIV) generally involves use of a tightly fitting face or nasal mask without any artificial airway.(79) NIV can be carried out using negative pressures (for example, iron lung); however, positive-pressure application is much more common.(79) Bi-level Positive Airway Pressure (BiPAP) is a form of NIV that distributes two levels of pressure in order to provide ventilatory support for the individual. The primary goal of BiPAP is to reverse acute respiratory failure without the insertion of an artificial airway. Continuous positive airway pressure (CPAP) is an alternative type of NIV in which continuous pressure that is greater than atmospheric pressure is maintained throughout the breathing cycle. Importantly with CPAP, the individual must be able to breathe spontaneously and so cannot be used if there is no neural control of breathing as would be the case with high cervical complete SCI. CPAP is most commonly provided to individuals with sleep apnoea.(78) In terms of invasive ventilation, there are many different types of ventilators that can be used. In the intensive care unit (ICU) setting, the ventilators used tend to be larger machines that can provide greater control of a wide range of parameters. However, use of these intensive care ventilators requires specialist expertise and the machines themselves are not easily moved. Outside of the ICU setting, certain smaller BiPAP or "respiratory assist devices" (RAD) can be used invasively (that is, via tracheostomy). While these smaller RAD may lack the functionality seen in intensive care ventilators, their simplicity and portability allows individuals to be cared for in a broader range of settings.(80) Invasive mechanical ventilation rather than NIV is generally necessary for patients with complete high cervical SCI, particularly during the initial management phase due to the lack of neural control of breathing.(5)

While mechanical ventilation, through its effect on alveolar ventilation, can be life sustaining, can improve other symptoms such as breathlessness, and can improve health-related quality of life, it is associated with side effects. For NIV, these side effects include dry throat, facial pains and impaired sleep. In addition, individuals may feel discomfort due to the prolonged use of masks.(81) For invasive ventilation these side effects include barotrauma (this is when the alveoli of the lungs rupture due to overinflation from increased pressure levels), volume trauma / volutrauma (this is when the alveoli become filled with fluid due to high tidal volumes) and ventilator-associated pneumonia (this is a healthcare-associated pneumonia that develops >48 hours after initiation of mechanical ventilation).(78, 79)

3.2.3. Phrenic nerve stimulation

An alternative intervention to mechanical ventilation is 'phrenic nerve stimulation' also called 'diaphragm pacing', which involves the surgical implantation of an electrode around the phrenic nerve(s), connected to a radio-frequency receiver usually implanted in the chest wall. An external transmitter then sends radio frequency signals to the device by an antenna which is worn over the receiver (Figure 3.1).(82) Phrenic nerve stimulation works by providing electrical stimulation of the phrenic nerve resulting in diaphragmatic contraction.(83) In certain ventilator-dependent patients with SCI who have intact phrenic nerve function, phrenic nerve stimulation either by conventional thoracotomy and electrode placement directly upon the phrenic nerves, or via laparoscopic approach involving insertion of electrodes directly into the diaphragm in proximity to the phrenic nerves, holds promise for achieving ventilator independence.(27)

Figure 3.1: Bilateral phrenic nerve pacing system



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Although there is limited evidence that phrenic nerve stimulation improves life expectancy or survival, observational studies have reported an association between the use of phrenic nerve stimulation and significant improvements in quality of life, as well as partial or complete freedom from ventilatory requirements.(85) Phrenic nerve stimulation may offer other benefits to individuals compared with traditional mechanical invasive ventilation, such as, facilitating increased mobility, improvements in speech, and reduction in the fear of ventilator disconnection.(86) Phrenic nerve stimulation is considered a viable long term alternative to invasive mechanical ventilation for many patients with high cervical SCI (particularly C1-2) who are ventilator dependent. However, damage to the phrenic nerve (which is quite common in cervical SCI, particularly in those with C3-C5 injuries) precludes its use.(87) It is important to note that while phrenic nerve stimulation does not require a tracheostomy, these individuals usually have a tracheostomy in place and a back-up ventilator or Ambu®Bag (manual resuscitation bag) on hand in case of device failure.(86) Though uncommon, one of the most serious complications of phrenic nerve stimulation is iatrogenic phrenic nerve injury, which can result in diaphragmatic paralysis or dysfunction. This damage may occur during initial electrode placement or later on due to mechanical failure or development of fibrosis around the nerve.(86) Loss of battery power is the most common source of mechanical failure and is avoided through routine maintenance procedures such as regular battery changes and recharging schedules.(86)

3.2.4. Secretion management

Secretion management is crucial in patients with SCI who are invasively ventilated; this is due to the inability of these individuals to clear secretions on their own. Airway clearance is important in order to prevent atelectasis (that is, a complete or partial collapse of the lung) and respiratory infections.(31) Therefore the success of any ventilatory support for patients with SCI depends on efficient coughing.(88) It is important that these individuals reach a maximal inspiratory capacity before coughing, and that peak cough flow is maximised. Maximal inspiratory capacity can be achieved by techniques such as air stacking, continuous training of the remaining inspiratory muscles, glossopharyngeal breathing, insufflation with a resuscitation bag, or use of a mechanical insufflation / exsufflation (MIE) device (such as a cough assist device). Maximal peak cough flow can be achieved by self-assisted cough techniques while supine or sitting in a wheelchair, assisted cough techniques administered by those providing care, or with MIE.(88) Pharmacological agents can also be considered if secretions are too thick or too thin to expectorate.(31) In certain situations, endotracheal suctioning and bronchoscopy may be necessary.(31)

3.2.5. Bladder management

Patients with complete C1-3 SCI require total assistance with bladder care.(89) The underlying principle for bladder management in patients with SCI is to develop a personalised regular bladder routine. This may include a variety of treatments such as medications (such as antimuscarinics or alpha-adrenergic blockers), catheters (such as urethral or suprapubic) and injections (such as intravesical botulinum toxin).(35) Oral antimuscarinics are recommended as the first-line pharmacological treatment for patients with neurogenic lower urinary tract dysfunction in order to improve overactive bladder symptoms and neurogenic detrusor overactivity, decrease urgency urinary incontinence and lower detrusor pressures.(90) Botulinum toxin type A injections to the bladder wall can be considered as a second-line treatment for the treatment of detrusor overactivity when antimuscarinic drugs have not worked or are poorly tolerated.(90) Indwelling catheters (such as Foley catheters) are inserted directly into the bladder and remain in place to continually drain the bladder. Indwelling catheters may be inserted through the urethra or through a surgically created hole through the abdomen (called suprapubic catheters).(35) Suprapubic catheters are generally preferred over urethral catheters for long term use due to the risk of stress incontinence and damage to the urethra.(90) Bladder surgery is usually only considered if other less invasive treatments are not effective.(35)

3.2.6. Bowel management

Similar to the management of bladder dysfunction, patients with complete C1-3 SCI require total assistance with bowel care.(89) These individuals generally all experience spastic bowel and so are unable to voluntarily defecate. However, activation of bowel reflexes will allow for these passage of stools in individuals with reflexic neurogenic bowel dysfunction.(40, 90)

A basic bowel management programme is the first-line treatment for all patients with SCI diagnosed with neurogenic bowel dysfunction.(37) Basic bowel management, which is individualised to the person, is designed to minimise or eliminate the occurrence of unplanned or difficult evacuations, by evacuating stool at a regular, predictable time. Basic bowel management is usually undertaken every day or every second day, but no less than three times per week.(37) These bowel routines are usually done at the same time each day so the body gets used to the programme. The goal of basic bowel management is usually to be able to complete the routine within 60 minutes on a regular basis to empty stool from the bowel and prevent complications.(40) Components of a basic bowel management programme vary depending on the individual’s needs but generally include:(40)

- diet and fluid management
- oral laxatives (such as stimulants and softeners)
- rectal laxatives (such as suppositories and enemas)
- physical activity
- scheduled bowel care (the process of assisted defecation by a trained carer)
- rectal evacuation methods (such as manual evacuation).

Surgical interventions (such as Malone antegrade continence enema (MACE) procedures) are recommended for patients with SCI with severe neurogenic bowel dysfunction for whom other treatment modalities have failed.(90)

Patients with complete C1-3 SCI are fully dependent on others for bowel care, which needs to happen at the same time on a regular basis to avoid complications. Those providing care need training in how to perform bowel care and how to recognise autonomic dysreflexia, which is a life-threatening emergency. Digital stimulation and other bowel techniques may trigger autonomic dysreflexia in people with injuries at T6 and above. Signs such as sweating, headache, change in heart rate, goose bumps and increasing muscle spasms may indicate autonomic dysreflexia.(40) Of particular importance is to know who to call when there are issues and how to access medical intervention for bowel management should it be necessary.(37)

3.2.7. Prevention and treatment of pressure injuries

The prevention and treatment of pressure injuries in patients with SCI is complex, lifelong and involves a range of health professionals. Given that all patients with SCI are at risk of pressure injuries, it is recommended that pressure injury prevention strategies are implemented as soon as emergency medical conditions and spinal stabilisation status allow.(41) Daily comprehensive visual and tactile skin inspections are recommended, with particular attention paid to areas vulnerable to pressure injury development such as the sacrum, coccyx, ankles and knees.(41) During the initial phases of care, patients with SCI should be turned or repositioned every two hours. Over time the frequency of turning/repositioning may be reduced as tissue tolerance increases. Hoists and other lifting devices are required to assist in moving patients with high cervical SCI in bed, however individuals may have the ability to reposition themselves in a suitably adapted power recline and/or tilt wheelchair.(89) Other critical aspects to a pressure injury prevention strategy include:(41)

- Regular cleansing of the skin
- Ensuring comfortable clothes and room temperature
- Use of pressure redistribution support surfaces
- Proper positioning of pillows and cushions
- Provision of an individualised pressure redistribution system
- Ongoing monitoring of the performance of support surfaces for bed and wheelchair

- Exercise
- Nutrition
- Education.

It is recommended that a comprehensive treatment plan should begin with an assessment of risk, health status and status of the pressure injury. The elements of the treatment plan should address the following: cleansing, debridement, dressings, surgery, nutrition, and positioning and support surface use. The treatment plan may need to be reviewed if there is no evidence of healing within 2-4 weeks.(41)

3.3. Prevalence of domiciliary ventilation

Provision of mechanical ventilation services at home (henceforth called domiciliary ventilation) is the preferred option for many individuals who require ventilation for extended periods of time.(80) Long-term mechanical ventilation may be a) elective and arise from conditions that progressively worsen over time (for example, neuromuscular disorders or COPD), or b) emergent due to failure to wean from mechanical ventilation following an episode of acute respiratory failure (for example, traumatic SCI).(91) Medical conditions that may lead to long-term ventilation include the following(92):

- central nervous system disorders (for example, SCI)
- neuromuscular disorders (for example, ALS, myasthenia gravis, spinal muscle atrophy)
- skeletal disorders (for example, thoracic wall deformities)
- cardiovascular disorders (for example, congenital and acquired heart disorders)
- upper airway disorders (for example, tracheomalacia)
- lower airway disorders (for example, COPD, cystic fibrosis).

Table 3.1 provides a summary of selected international studies which examined the prevalence of domiciliary ventilation. These studies indicate that the prevalence of domiciliary ventilation varies across and even within jurisdictions, but overall the prevalence is generally low. Dybwik et al. conducted a focus group study to examine why the geographical provision of domiciliary ventilation services varied so widely within Norway. The authors concluded that the geographical differences in service provision may be explained, at least in part, by "individual attitudes" by decision-making physicians, and differing levels of "enthusiasm" by the hospitals towards domiciliary ventilation.(93) A study by Garner et al., conducted in Australia and New Zealand, reported that domiciliary ventilation prescribing patterns were dependent on each centre's location, size and experience, with the most densely populated states having the highest prevalence of domiciliary ventilation.(94) Toussaint et al. reported a positive correlation between the prevalence of domiciliary ventilation and the gross domestic product (GDP) per capita ($R^2=0.4359$) and the human development index score (HDI) ($R^2=0.3475$) of a country, indicating that a nation's wealth may be linked with the prevalence of domiciliary ventilation. However, the authors advised caution in the interpretation of this link given issues with some of the underpinning data.(95)

Where reported, patients with SCI accounted for a small proportion of all domiciliary ventilation service users, ranging from 2%(96) to 19.9%,(97) with the latter study focused exclusively on a population availing of invasive ventilation. Moreover, these studies highlighted that NIV is generally much more commonly used than invasive ventilation in a home setting. The prevalence of domiciliary ventilation was reported to be increasing over time, driven largely by increased use of home NIV,(11, 98-103) with a general decline in the overall proportion availing of invasive ventilation reported.(11, 97, 99, 101, 103) The increasing use of NIV may be due to the emerging evidence to support its use in conditions such as OHS and COPD.(95) However, among domiciliary ventilation service users with SCI, invasive ventilation was reported (in one survey-based study) to be more commonly used.(91) It is important to note that the studies outlined in Table 3.1 do not necessarily represent the totality of studies examining the prevalence of domiciliary ventilation, as a systematic literature search was not conducted to identify all such studies.

Table 3.1: Summary of selected studies examining the prevalence of domiciliary ventilation in various jurisdictions

Study	Country/region	Year(s) of data collection	Prevalence of domiciliary ventilation per 100,000 population	% Spinal cord injuries	% Invasively ventilated
Swedevox registry(99)	Sweden	1996-2021	29.8 (in 2021)	~10†	~2
van den Biggelaar et al.(11)	The Netherlands	1991-2020	5.6 (in 2001/2002) 22 (in 2020)	NR	~10
Kotanen et al.(97)‡	Finland	2015-2019	2 (in 2019)	19.9	100
Tan et al.(98)	Singapore	2009-2015	NR	13	34
Rose et al.(91)	Canada	2012-2013	12.9	~13 (of invasive ventilation population) ~5 (of NIV population)	18
Gouda et al.(104)*	Galway/West of Ireland	2000-2012	~16	4	0
Povitz et al.(101)	Ontario, Canada	2000-2012	1.8 (in 2000) 5 (in 2012)	5.3	12.2 (in 2000) 6.3 (in 2011)
Escarrabill et al.(102)	Catalonia, Spain	2008-2011	23	NR	NR
Tan et al.(96)	Western Australia, Australia	2005-2010	NR	2	1
Nasiłowski et al.(103)	Poland	2000-2010	~0 (in 2000) ~2.5 (in 2010)	NR	100 (in 2000) ~50 (in 2010)
Lloyd-Owen et al.(105)	16 European countries	2001	6.6 (across all included countries) (range from 0.1 in Poland to 10 in Sweden; 3.4 in Ireland)	NR	13

Key: NIV – non-invasive ventilation; NR – not reported.
†Category combines spinal cord injuries with idiopathic hypoventilation
‡Study specific for domiciliary invasive ventilation
*Study specific for NIV (reported here as conducted in Ireland)

3.4. Evidence regarding benefits, harms and costs of domiciliary ventilation

The evidence regarding the clinical benefits, harms and costs of domiciliary ventilation is relatively scarce and is based on heterogeneous populations, study designs and methodological approaches, as well as small sample sizes. Very limited data pertain specifically to patients with SCI availing of domiciliary invasive ventilation, given the rare occurrence of this condition and the ethical challenges of conducting comparative research in this population. For the purpose of this section, a high level overview of the domiciliary ventilation evidence is provided, with a focus on the adult SCI population and invasive ventilation, where reported. This section focuses on several key reviews and a HTA on this topic. Two important primary research studies published since the search dates of the included evidence syntheses are also discussed.

3.4.1. MacIntyre et al.: systematic review

A systematic review evaluating the association between domiciliary ventilation and clinically relevant patient-centred and health resource utilisation outcomes was conducted by MacIntyre et al.(106) Based on searches up until August 2013, the review included one randomised control trial (RCT) and 25 observational studies of mixed methodological quality involving a total of 4,425 patients. The largest patient cohort were those with neuromuscular disorders (38%), within which the review authors categorised an unknown number of patients with SCI. It was noted that NIV was used in the majority (85%) of patients across the included studies.

Health related quality of life (HRQL) was generally described as 'good' in individuals across the included studies.(106) Five of the included studies examined HRQL before and after domiciliary ventilation using the SF-36 instrument. A more consistent improvement in HRQL was observed across the mental capacity domains compared with the physical ones, though, on the whole, domiciliary ventilation was generally associated with either no change or a statistically significant improvement in HRQL. Importantly, there was no evidence of a clinically important deterioration in any group within the mental capacity domains. Physical component scores were more heterogeneous, however, with one study involving individuals with neuromuscular disorders reporting a statistically significant deterioration in physical functioning after introduction of domiciliary ventilation.(107) Importantly this was a small study (n=27) which did not involve anyone with SCI. No other deterioration in HRQL was reported across the physical domains in any included studies.

Nine of the included studies in the review examined hospitalisation outcomes.(106) In general, hospitalisation rates and days in hospital overall were reported to be low and decreased following the introduction of domiciliary ventilation services. Where specified, reasons for hospital admission were related predominately to cardiac or respiratory related problems. Admissions for ventilator setting adjustments or equipment servicing were reported to be infrequent, or were not reported. However, reporting methods and disease states differed significantly precluding any meaningful quantitative synthesis by the review authors.(106) Healthcare resource utilisation was assessed in seven included studies. Home care costs were found to be substantial in these studies, with out-of-pocket expenses (for example, privately funding home care, medications or equipment) also reported to be significant. The review identified four studies with cost estimates for the provision of domiciliary ventilation services, of which one related specifically to patients with SCI.(108) Adjusting to 2022 Irish Euro Prices, the annual cost of home care per ventilator-dependent individual with SCI was estimated to be €586,707 increasing to €778,030 once all equipment costs were accounted for.(106, 108)

The impact on family carers was assessed in eight included studies.(106) Family carers in these studies (n=555) were predominantly younger than the ventilator user, female (76%) and of spousal (54%) or parental (31%) relation. Fewer than half of the family carers were in active employment. Carer burden was assessed in three of these studies; all three highlighted significant burden in the domains of financial strain, negative impact on employment, and insufficient time for oneself and for personal relationships. Satisfaction among family carers with regards to their decision to care for their loved one was generally high (average of 80% positive), though varied depending on the population and relationship to the individual.

3.4.2. Sison et al.: systematic review

Sison et al. conducted a systematic review (searches up until March 2020) with the aim of determining the outcomes of individuals who are chronically invasively ventilated outside of ICU settings.(109) A total of 60 studies were included which involved a range of settings; these were categorised as either 1) ICU 2) step-down units/in-patient wards, or 3) home/nursing home settings. While the indications for mechanical ventilation were not reported in this review, the authors noted that there was considerable clinical heterogeneity among included studies. The authors found that though mortality rates within and between care settings differed vastly, mortality outcomes of individuals in non-ICU studies were crudely comparable to those in ICU. Mortality rates ranged from 13.7% to 77.8% in ICUs (n = 17 studies), 7.8% to 51.0% in step-down units and inpatient wards (n = 26 studies), and 12.0% to 91.8% in home or nursing home settings (n = 19 studies). Importantly, studies with longer follow-up durations in home or nursing home settings reported higher mortality rates.

Ten out of 19 homecare/nursing home studies included in this review reported on hospitalisation and complication rates.(109) The proportion of chronically mechanically ventilated patients in home care settings requiring at least one hospitalisation ranged from 44.1% to 60.0%.(109) However, hospitalisation rates were generally observed to be low in this population. When examined for a follow-up period of one year, two of the included studies estimated a mean number of hospitalisations per patient per year of only 0.82 (± 0.98)(110) and 1.7 (± 2.9),(111) respectively, and another study reported that individuals were admitted to the hospital for an average of 3.3 days (± 6.5) annually.(112) Among included studies in this review, ventilator malfunction was a stated reason for hospitalisation in 7.7% to 28.6% of home care individuals, though the authors noted that mortality rates due to ventilator failure were low in home settings, with none of the studies reporting ventilator malfunction as the primary cause of death.(109) Based on the totality of the evidence in this review, the authors concluded that non-ICU settings should be viewed as suitable and safe alternative care settings for medically stable, ventilator-dependent individuals.(109)

3.4.3. Geiseler et al.: HTA

A HTA on the topic of home invasive mechanical ventilation was published in 2010 by the German Institute for Medical Documentation and Information.(113) The HTA covered a series of domains including medicine/nursing, economics, social, ethical and legal aspects, and was based on a systematic review conducted in 2008. The authors concluded that invasive mechanical ventilation was associated with an improvement in the symptoms of hypoventilation. While an increase in life expectancy was considered likely, this could not be definitively confirmed through the conduct of an RCT, as such a study would be unlikely to occur for ethical reasons. Complications (such as pneumonia and blocked cannulas) were reported to be rare among included studies, indicating the safety of care in the home setting. Included studies reported domiciliary ventilation to be cheaper than ICU ventilation with cost savings ranging from 62% to 74% across included studies. However, domiciliary invasive ventilation was found to be particularly costly and substantially more so than domiciliary NIV due to the greater equipment and consumable requirements and the need for 24/7 care from highly qualified staff.

With regards to HRQL, evidence was limited and mostly qualitative in nature. The HRQL of individuals receiving domiciliary ventilation was mostly self-reported as 'good'.(113) However, the impact of domiciliary ventilation on carers was much more mixed with both positive and negative impacts reported across included studies.

3.4.4. King: literature review

A non-systematic literature review published by King in 2012 (no literature search date provided) examined a broad range of issues pertaining to long-term home mechanical ventilation in the US.(80) The author outlined advantages from the patient's perspective of domiciliary ventilation compared with ICU ventilation, which include a quieter, more familiar environment, greater access to family and friends, greater personal space and privacy, as well as greater independence.(80) The author also pointed to evidence that domiciliary ventilation is associated with reduced healthcare costs,(114) as well as improved quality of life and community engagement.(92)

However, the author highlighted that there may be additional risks associated with providing highly complex care at home (for example, ventilator failure).(80) One included study of 150 individuals availing of domiciliary ventilation in the US recorded 189 reported home ventilator failures in a one year period, equivalent to one ventilator failure for every 1.25 years of continuous use.(115) In this study, the most common causes of home ventilator failure were reported to be "defective equipment or mechanical failure" (39%) followed by "functional equipment improperly used by caregivers" (30%). The most common remedial action undertaken was to provide a replacement ventilator (44%). Of note, hospitalisation was only necessary in 1% of failure reports. No adverse outcomes, deaths, or serious injuries were associated with home ventilator failure in this study.(115) Another study from the UK evaluated the nature of calls to a support line for individuals availing of domiciliary ventilation. The population comprised 1,211 individuals predominantly receiving NIV (99%). The authors reported a total of 188 emergency calls, with ventilator failure accounting for 28% of emergency visits to the home.(116) The review author noted that while deaths and other serious consequences have occurred at home due to ventilator failure and or alarm malfunction, these events tended to be rare.(80)

3.4.5. Relevant primary research studies

Two important primary research studies, identified as part of the overarching search strategy (Appendix 1), have been published since the search dates of the above discussed evidence syntheses.(117, 118)

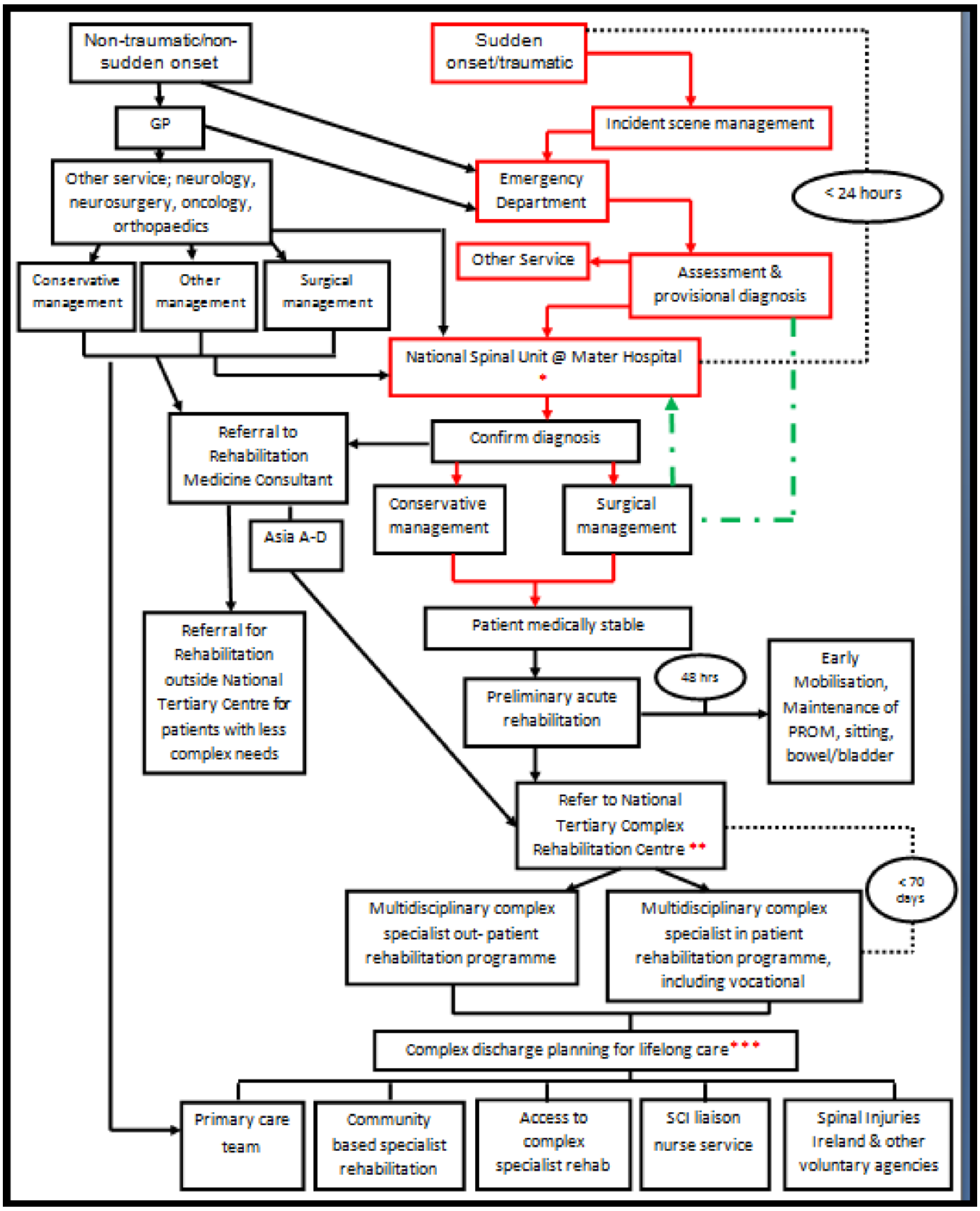
Ribeiro et al. describe the findings of a five-year longitudinal cohort study conducted in Portugal between January 2016 and January 2022, evaluating the change in HRQL over time in individuals receiving domiciliary ventilation.(118) A total of 104 participants were included in this study, 51 of whom (49%) had COPD; it is unclear how many, if any, patients with SCI were included. Participants were on domiciliary ventilation for a median of 43.5 months (95% CI, 22-88.5), and were found to have good overall adherence to ventilation therapy (median 8 hours per day (95% CI, 6-9) daily hours). At five years, 57 participants (54.8%) were still alive. HRQL scores remained relatively stable over the course of the study in those that still alive after five years. It was also observed that HRQL was significantly higher at baseline in those that lived beyond five years than in those that died within five years of domiciliary ventilation (median summary scores according to the Severe Respiratory Insufficiency (SRI) questionnaire; 59.6 vs 48.7; $p = 0.004$). It is important to note however that no participants in this study were treated with invasive ventilation, so the applicability of these findings to patients with SCI availing of domiciliary invasive ventilation is unclear.

Klingshirn et al. describe the findings of a mixed methods study conducted in Germany (between 2019-2020) that aimed to examine differences in the quality of care provided to individuals receiving domiciliary ventilation at home versus in shared living communities.(117) Of the 46 individuals receiving ventilation who participated in the survey, 37 (80%) availed of invasive ventilation, 28 (61%) lived in a private home, and four (9%) had SCI. Participants living in a private home were on average almost 20 years younger than those living in a shared living community (average: 46.86 ± 15.40 years vs. 65.07 ± 11.78 years; $p = 0.001$). No significant difference was found in HRQL between those living in private homes and those living in shared living communities. In both cohorts, the lowest score in the SRI questionnaire was in the 'physical functioning' domain. Carer burden among 18 participating carers was found to be 'moderate' on average, as measured by the short version of the Burden Scale for Family Caregivers (BSFC-s), with no significant differences observed between those caring for someone at home versus in a shared living community. Qualitative findings from the semi-structured interviews (individuals on ventilation: $n = 13$; family carers: $n = 18$) highlighted that, though challenging, person-centred care is possible in both settings. The main limitation of this study was the small number of individuals included in the survey. The authors noted that the COVID-19 pandemic made recruitment into this study a particular challenge.

3.5. Current integrated care pathway in Ireland

An integrated care pathway for patients with SCI has been developed in Ireland by the HSE's National Clinical Programme for Rehabilitation Medicine, in line with its model of care.(2, 7) Integrated care pathways are structured multidisciplinary care plans which detail essential steps in the care of patients with a specific clinical problem.(119) The care pathway for the management of SCI extends from pre-hospital care through to discharge and lifelong care (Figure 3.2).

Figure 3.2: Full pathway of care for patients with spinal cord injury in Ireland



Source: HSE's National Clinical Programme for Rehabilitation Medicine(2)

As part of this integrated care pathway, patients with SCI are transferred to the National Rehabilitation Hospital (NRH) for post-acute rehabilitation once they are deemed to be ready to engage in rehabilitation. In 2019, the NRH commenced admission of ventilator-dependent patients with SCI. Prior to this, ventilator-dependent SCI patients in Ireland were transferred to hospitals in the UK to receive inpatient rehabilitation care under the HSE's Treatment Abroad Scheme (TAS). The new pathway, within the broader integrated care pathway, was developed by the Mater National Spinal Cord Injury team and the NRH Spinal Cord System of Care programme. The first patient was admitted in 2019; this followed the appointment of an intensivist and a rehabilitation SCI care coordinator, and the development of an education programme which addressed staff training and competencies in the management of patients with high SCI and ventilator dependency.

Discharge planning for individuals who are ventilator-dependent remains a huge challenge for a range of reasons, including the current fragmented funding model for community services as well as the lack of suitably trained homecare staff in the community. There may also be regional variation in the availability of the necessary resources to enable the homecare of these individuals. As a result, patients with SCI who are ventilator dependent often remain hospitalised for a prolonged period of time, often several years, before being discharged home.(2) These delayed discharges can have a detrimental impact on the individual, their family and carers, management of bed capacity in the NRH and other hospitals, and the wider healthcare system due to the significant cost of treating these patients in hospital.

3.6. Discussion

High cervical SCI can be catastrophic for an individual. These injuries can result in permanent tetraplegia and respiratory failure among a range of other life-changing consequences, rendering these individuals completely dependent on others. Patients with lesions C4 or higher generally require mechanical invasive ventilation to survive given the lack of neural control of breathing, though the majority of these individuals (particularly those with C4 injuries) will eventually be weaned off the ventilator.

Hypercapnic chronic respiratory failure can result from a number of underlying diseases such as ALS, OHS and SCI. It is this inability to adequately remove carbon dioxide from the blood that requires an individual to be ventilated to ensure efficient gas exchange. In general, a person requires a ventilator until such a point as they can breathe spontaneously on their own. The respiratory management of patients with SCI who are ventilator dependent is complex. Clinicians need to carefully consider how best to manage the chronic respiratory failure in these individuals in the long term (for example, invasive ventilation or phrenic nerve stimulation).

Domiciliary ventilation is growing in prevalence internationally, though this increase is driven largely by the increased use of NIV as opposed to invasive ventilation. The prevalence of domiciliary ventilation varies between and even within countries and there may be several factors influencing this (such as the centre’s location, size and experience, as well as differing levels of adoption of the service among hospital groups). Domiciliary ventilation offers those who require long-term mechanical ventilation a suitable alternative to ICU ventilation. Given the rarity of domiciliary invasive ventilation, particularly in SCI populations, evidence of its clinical effectiveness and safety is weak or absent. Based on limited evidence, individuals availing of domiciliary ventilation generally reported improvements in HRQL outcomes; however, the evidence for family carers is mixed, with both improvements and disimprovements reported. While domiciliary ventilation has been used and recommended for a range of conditions under specific circumstances (for example, where the patient is medically stable, correct supports are in place, the patient's family is supportive, and other relevant criteria are satisfied), it is important to note that there are some safety concerns associated with the provision of such complex care in the home setting. There are reports in the literature of ventilation failure, hypoxic brain injuries and deaths associated with the use of domiciliary ventilation, though these are rare. In the absence of important risk mitigation factors, such as, a rigorous clinical governance system, proper training, sufficient staffing, resources and supports, there is the potential for serious consequences where care is provided in the home setting. Additionally, from the patient's perspective, there is often a preference to return home once the right supports and equipment are in place, even if this does entail some added risk. Importantly, the alternative is to spend an indefinite amount of time in an ICU, which may not be in anyone's best interest (the patient, the family or the healthcare system). Regardless of the setting in which care is provided for these patients, it is important to note that an element of risk will always be involved given the reliance on a ventilator to stay alive.

There are important limitations to note with regard to the underlying evidence base. The studies were generally small and of heterogeneous study design, and so it is challenging to determine whether there are differences in outcomes between populations. It is also important to consider that there may be inherent differences in the populations that are discharged for domiciliary ventilation compared with those that remain as inpatients (for example, those that are more unwell may not be considered suitable for domiciliary ventilation); these inherent differences may be difficult to control for in analyses given the small sample sizes involved. An integrated care pathway for patients with SCI has been developed in Ireland by the National Clinical Programme for Rehabilitation Medicine. This care pathway extends from pre-hospital care to discharge and lifelong care. However, there are challenges (such as funding and staffing) with the implementation of this care pathway, particularly at the discharge stage. As a result of these barriers, patients with SCI who are ventilator dependent might have to remain hospitalised for a prolonged period of time, often several years, before being discharged home.(2)

4. Scoping review of international practice and standards

Key points

- Patients with high cervical cord injuries requiring invasive mechanical ventilation have complex health care needs, and require substantial support from formal and informal caregivers in a home setting.
- A scoping review of international practice and standards relevant to the provision of permanent domiciliary invasive ventilation in adults with spinal cord injury was undertaken. The search included guidelines, position papers, protocols and standards (collectively referred to as 'guidance documents') that related to, or were applicable to a population with SCI.
- Seventeen guidance documents from Australia, Canada, Germany, Italy, New Zealand, Poland, Switzerland, the Netherlands, the UK, and the US, were identified for inclusion. Ten contain SCI-specific guidance regarding domiciliary invasive ventilation while seven contain more general, non-condition specific guidance.
- In relation to domiciliary invasive ventilation guidance, detailed recommendations are provided for the following topics:
    - discharge process
    - communication and monitoring post-discharge
    - equipment
    - staffing.
- Patients with high cervical cord injuries generally have multiple, complex healthcare needs, often as a direct or indirect consequence of the SCI. Recommendations are provided in relation to the management of the following issues:
    - respiratory complications
    - bladder complications
    - bowel complications
    - skin integrity
    - sexual health
    - venous thromboembolism

    - pain
    - autonomic dysreflexia
    - emotional wellbeing, mental health and substance abuse
    - cardiometabolic disease and nutrition
    - bone health
    - palliative care.
- In relation to broader issues of SCI care pathways and supports, guidance is issued in relation to:
    - education and training
    - governance structures and care pathways
    - ethics and safeguarding.
- In terms of governance, guidance documents discuss the importance of having clear roles and responsibilities for the domiciliary ventilation service. Individuals receiving domiciliary ventilation and their families should be clear about who to contact for different reasons (for example, acute illness, equipment malfunction or patient transfers). There should be a clear understanding among the various clinicians involved as to who is ultimately responsible for the care of these individuals in the community.
- There is consistency across guidance documents that 24/7 care, involving at least one trained homecare staff who has demonstrated competency for the tasks required, is essential for these individuals due to their complex healthcare needs. However, there is a lack of consensus on the requirement for nursing qualifications in the provision of care to these individuals.
- In general, the guidance documents all discuss the importance of appropriate preparation and timing of the transition to the home setting; anticipation of present and future requirements in terms of environment and training of those providing care; and anticipation of disease progression.
- A comprehensive, holistic and person-centred approach to care, delivered from a well-coordinated interdisciplinary team, which respects the preferences of the individual, is advocated in these guidance documents.
- While international standards and practice in this area may vary, there are many common recommendations that may be useful to inform a national provision of service in Ireland.

4.1. Introduction

The delivery of domiciliary invasive ventilation services differs from jurisdiction to jurisdiction. The aim of this Chapter is to describe a scoping review that was undertaken of international practice and standards relevant to the provision of permanent domiciliary invasive ventilation in adults with spinal cord injury.

4.2. Methods

The methods undertaken in this review are detailed in a separate protocol document which has been provided to the EAG.

4.2.1. Scoping review methodology

Given the broad nature of this topic, a scoping review was undertaken. A scoping review can be defined as a form of evidence synthesis that addresses an exploratory research question with the aim of mapping key concepts, types of evidence, and gaps in research related to a defined field; this is accomplished by systematically searching, selecting, and synthesising existing evidence.(120) Scoping reviews are generally preferable to systematic reviews when the purpose is to provide a comprehensive overview of a broad topic, rather than to determine the efficacy or effectiveness of a specific intervention.(121) Importantly, scoping reviews still provide a high standard of rigour and transparency.(121, 122)

This review adhered to the Arksey and O'Malley six-stage framework for conducting a scoping study.(123) This framework follows the main systematic reviewing principles; however, it allows for more flexibility in terms of inclusion and exclusion criteria, pays less attention to quality appraisal and is more focused on presenting a thematic overview of findings rather than determining any definitive effect estimate.

4.2.2. Research question

The following research question was formulated according to the Population, Area of Interest and Context (PICo) framework (as shown in Table 4.1):

- What practice and standards do international, national or regional guidelines, recommendations, position papers and standards specify for the provision of permanent domiciliary invasive ventilation in adults with spinal cord injury?

For the purpose of this scoping review, the umbrella term 'guidance documents' is used to describe the heterogeneous collection of guidelines, position papers, recommendations and standards identified for inclusion in this review. These guidance documents are used to infer practice and standards in their respective jurisdictions.

Table 4.1 PICo for Scoping Review of International Practice and Standards

Population	Individuals with SCI receiving permanent domiciliary invasive ventilation
Interest	Practice and standards for provision of care including, but not limited to: ▪ Pre-transition ▪ Patient assessment ▪ Room and environment requirements ▪ Training requirements ▪ Staffing requirements ▪ Informed consent processes ▪ Funding process and requirements ▪ Transition phase ▪ Process of transition ▪ Transportation requirements ▪ Liaison and other support staff ▪ Technical requirements ▪ Ventilators ▪ Oxygen ▪ Hoist ▪ Wheelchair ▪ Bed ▪ Other equipment ▪ Management of SCI complications ▪ Ventilator management ▪ Respiratory care and function ▪ Autonomic dysreflexia ▪ Emergency management ▪ Bowel management ▪ Bladder management ▪ Medication administration ▪ Nutrition and feeding ▪ Skin integrity ▪ Stoma care ▪ Waste management ▪ Telemedicine ▪ Infection prevention and control ▪ Psychosocial care ▪ End of life/palliative care ▪ Rehabilitation ▪ Education and training ▪ Governance, quality assurance and legal issues ▪ Governance structures and accountability

Context	▪ Escalation pathways ▪ Care and referral pathways ▪ Organisations responsible for providing care, funding and oversight ▪ Safeguarding ▪ Ethics ▪ Quality assurance processes ▪ Other legal issues
	Guidance documents: ▪ Guidelines (international, national or regional) ▪ Position papers ▪ Recommendations ▪ Standards

4.2.3. Search strategy

The search strategy was developed by a librarian in conjunction with the other members of the evaluation team, and was peer-reviewed by a second librarian using the Peer Review of Electronic Search Strategies (PRESS) tool.(124) There were no language or date restrictions. The finalised search strategy is outlined in Appendix 1.

A search of the following electronic databases was conducted between 29 August 2022 and 6 September 2022: Medline (EBSCO), Embase (OVID), The Cochrane Library, ClinicalTrials.gov, CINAHL (EBSCO), and APA PsycInfo (EBSCO). This was supplemented by a grey literature search of the following sources conducted on 29 and 30 August 2022: TRIP, BMJ Best Practice, UptoDate, Guidelines International Network, International HTA Database, Core, Google, Google Scholar, the Physiotherapy Evidence Database (PEDRO) and the Agency for Healthcare Research and Quality (AHRQ).

A manual website search of the following international societies and agencies was conducted on 5 October 2022: the World Health Organization (WHO), European Respiratory Society (ERS), American Thoracic Society (ATS), Canadian Thoracic Society (CTS), American Association for Respiratory Care (AARC), American Spinal Injury Association (ASIA), Paralyzed Veterans of America (PVA), Multidisciplinary Association for Spinal Cord Injury Professions (MASCIP) UK, Thoracic Society of Australia and New Zealand (TSANZ), British Association of SCI Specialists (BASCIS), Respiratory Information for Spinal Cord Injury (RISCI), and the International Spinal Cord Society (ISCOS).

Bibliographies of included and otherwise relevant documents were screened. Additional ad-hoc Google searches were also conducted on 4 October 2022 to identify other potentially relevant documents.

4.2.4. Selection of studies

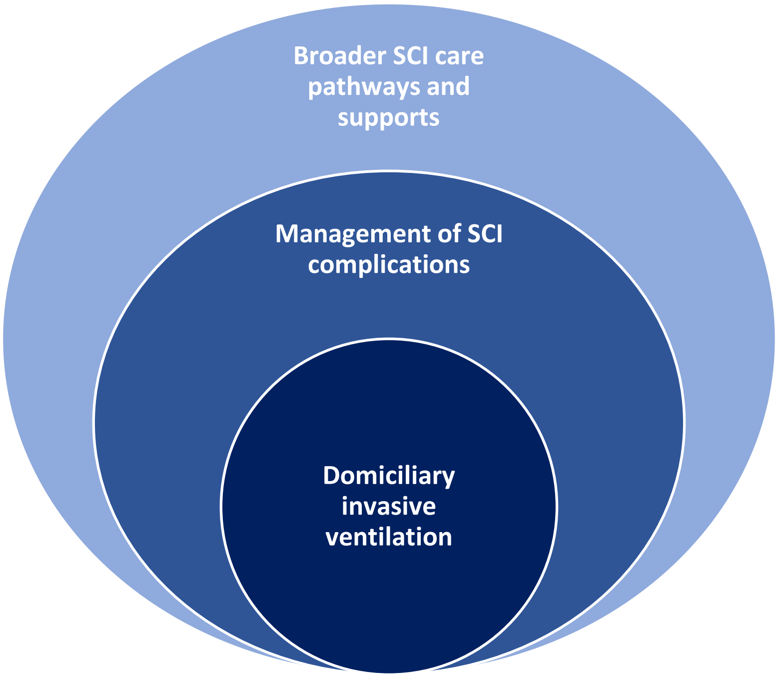
The results of the search were exported to Covidence (www.covidence.org) after de-duplication in Endnote. Two reviewers independently reviewed the titles and abstracts and, subsequently, full texts of the identified records. Those that met the inclusion criteria for this scoping review (as per Table 4.2), were included in this Chapter. Any disagreement regarding the eligibility of documents was resolved through discussion, and by a third reviewer where necessary.

Table 4.2: Inclusion and exclusion criteria

Inclusion Criteria	Exclusion Criteria
▪ Guidance is provided regarding the provision of care to those availing of invasive mechanical ventilation at home. ▪ Guidance is generally applicable to all invasively ventilated patients regardless of underlying diagnosis **and or** is specific to patients with spinal cord injuries ▪ Guidance is evidence-based (meets at least one of the following criteria: – based on a literature review – based on expert consensus methods, which are described in the document – affiliated with a recognised society/organisation.)	▪ Guidance is specific to another patient group, or a paediatric population, with very limited, or no applicability to adults with spinal cord injuries ▪ Hospital or long-term care based ventilation ▪ Acute or post-acute settings ▪ Older guidance that has since been updated (or synthesised into a newer guidance document) ▪ Guidance specific to an individual hospital/specialist centre/care provider ▪ Guidance is focused on the management of SCI complications only, without reference to domiciliary ventilation ▪ Guidance is focused on the broader structures of SCI care pathways and supports without reference to domiciliary ventilation.

Inclusion and synthesis was framed around three main concepts (Figure 4.1). Domiciliary invasive ventilation guidance was considered as essential for inclusion and as such was the primary outcome of interest for this scoping review. Guidance relating to the management of SCI complications was considered a secondary outcome. Guidance in relation to the broader SCI care pathway and supports was considered a tertiary outcome.

Figure 4.1 Conceptual framework for the Scoping Review



4.2.5. Data extraction

A data extraction tool was developed and piloted before implementing. Data extraction was performed by one reviewer and double-checked by another. Google translate was used to obtain translations of non-English language documents. As this was a scoping review aiming to provide an overview of a diverse range of standards and practice, no quality appraisal was undertaken.

4.2.6. Reporting

This scoping review is reported in accordance with the Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) Checklist.(125)

4.3. Results

4.3.1. Search results

A total of 1,246 records were identified via databases and registers, with 108 records identified via other methods (Figure 4.3). After removal of duplicates, 1,218 records (from all sources) were screened by title and abstract, of which 1,116 were excluded. One hundred and two reports underwent full text screening, of which 17 reports (hereafter referred to as 'guidance documents') were included in this scoping review.(79, 88, 90, 126-139) Four of the included 17 guidance documents were identified from databases or registers(79, 131, 134, 139) and the remaining 13 were identified from other sources.(88, 90, 126-130, 132, 133, 135-138). Excluded reports and their reasons for exclusion are outlined in Appendix 2.

4.3.2. Characteristics of included guidance documents

Relevant guidance documents containing information regarding the provision of domiciliary invasive ventilation were identified from 10 different countries (Figure 4.2). Three guidance documents were retrieved from Italy;(127, 129, 136) two each from the US,(128, 132) the UK,(130, 137) Germany,(79, 139) Poland(131, 135) and Canada;(90, 134) and one each from Switzerland,(88) Australia,(133) New Zealand(126) and the Netherlands.(138) Of the 17 included guidance documents, four were regional (Apulia,(127) Piedmont(136) and Campania(129) in Italy, and New South Wales(133) in Australia) and the remaining 13 were national; no relevant international guidance document was identified.

Importantly, not all guidance documents identified are specific for SCI, though all contain guidance on domiciliary invasive ventilation. Of the 17 included guidance documents, 10 contain SCI-specific guidance regarding domiciliary ventilation(79, 88, 90, 126, 130, 132-134, 137, 139) while seven contain more general, non-condition specific guidance.(127-129, 131, 135, 136, 138) The characteristics of the 17 included guidance documents are outlined in Table 4.3

Figure 4.2: Countries with relevant guidance documents

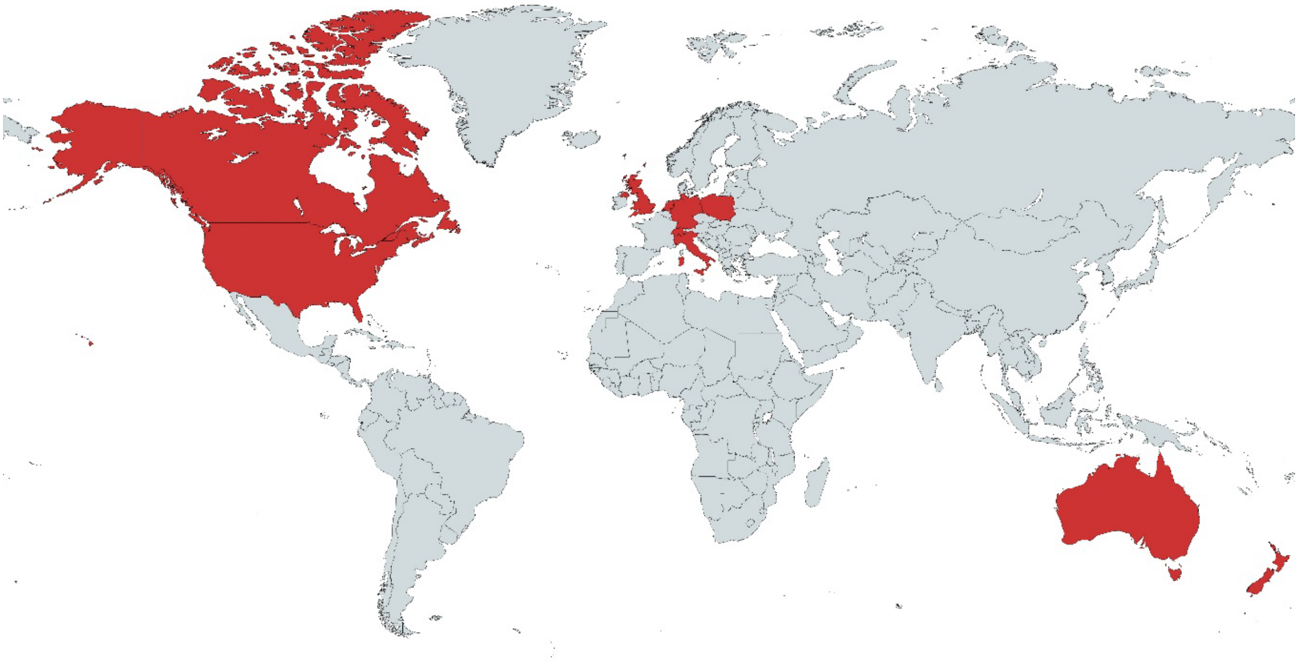


Figure 4.3 PRISMA flow diagram of included guidance documents

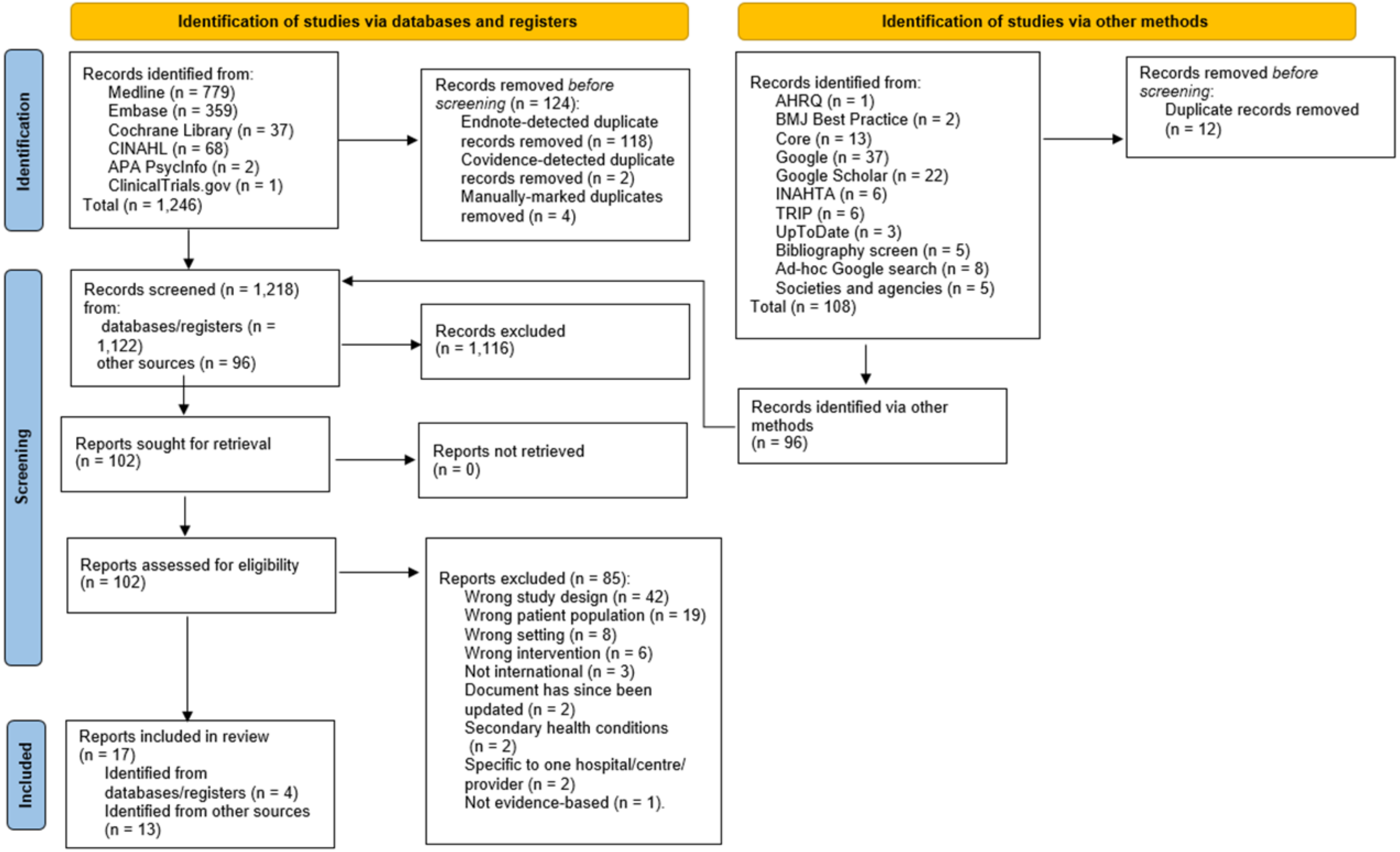


Table 4.3 Table of characteristics for included guidance document

Guidance document	Organisation/Author	Year	Level (National /Regional)	Adapted from previous Guidance	Evidence synthesis conducted	Expert Consensus	Funding
Australia							
Guidance on the support needs for adults with spinal cord injury (3rd Ed)(133)	Insurance & Care New South Wales (icare)	2017	Regional (New South Wales)	Yes	Yes	Yes	icare Lifetime Care
Canada							
Canadian Spinal Cord Injury Practice Guideline (Can-SCIP)(90)	Praxis Spinal Cord Institute, Spinal Cord Injury Research Evidence (SCIRE) & KITE (Knowledge, Innovation, Talent and Everywhere)	2021	National	Live guidance	Yes	Yes	Praxis Spinal Cord Institute, Vancouver, British Columbia
Home mechanical ventilation: A Canadian Thoracic Society clinical practice guideline(134)	Canadian Thoracic Society/ McKim *et al.*	2011	National	No	Yes	Yes	From multiple organisations
Germany							
German National Guideline for Treating Chronic Respiratory Failure with Invasive and Non-Invasive Ventilation: Revised Edition 2017 – Part 1 & Part 2(79, 139)‡	Lead society: German Society for Pneumology and Respiratory Medicine (DGP)	2018	National	Yes	Yes	Yes	DGP

Guidance document	Organisation/Author	Year	Level (National /Regional)	Adapted from previous Guidance	Evidence synthesis conducted	Expert Consensus	Funding
Italy							
Recommendations – Guidelines for long term home mechanical ventilation (Piedmont Region)[136]	Piedmont Region/Regional Commission for the coordination of the Pneumological Clinical Network of the Piedmont Region	2021	Regional	NR	NR	Yes	NR
Regional guidelines: Home Mechanical Ventilation. (Apulia Region)[127]	Apulia Regional Health Agency (A.Re.S.)	2013	Regional	NR	Not Clearly specified	Yes	NR
Regional guidelines: home mechanical ventilation. Adults and pediatrics (Campania Region)[129]	Regional Health care council (Campania)	NR	Regional	NR	NR	Yes	NR
Netherlands							
Chronic Ventilation[138]	Dutch Association of Physicians for Pulmonary Diseases and Tuberculosis, and the Association for Respiratory Support. Authorised by a range of Dutch associations and societies	2021	National	Yes	Yes	Yes	NR
New Zealand							

Guidance document	Organisation/Author	Year	Level (National /Regional)	Adapted from previous Guidance	Evidence synthesis conducted	Expert Consensus	Funding
Spinal cord injury guidelines(126)	Accident Compensation Corporation (ACC)	2017	National	Yes	Yes	Yes	NR
Poland							
Practical aspects of nursing care provided to patients diagnosed with amyotrophic lateral sclerosis receiving home mechanical ventilation & Amendment of the Regulation on guaranteed benefits in the field of nursing and care services as part of long-term care(131, 135) †‡	Butna et al & Minister of Health (Poland)	2014	National	N/A	N/A	N/A	N/A
Switzerland							
Long-Term Mechanical Ventilation: Recommendations of the Swiss Society of Pulmonology(88)	Swiss Society of Pulmonology/ Janssens *et al.*	2022	National	Yes	Yes	Yes	NR
United Kingdom							
Service Specification: Spinal Cord Injury Clinical Network – Standards of Care 2022(130)	British Association of Spinal Cord Injury Specialists	2022	National	N/A	No	Yes	N/A

Guidance document	Organisation/Author	Year	Level (National /Regional)	Adapted from previous Guidance	Evidence synthesis conducted	Expert Consensus	Funding
Carer training – Standards expected in terms of knowledge and skills Consensus statement on behalf of RISCI (GBI)(137)	Respiratory Information in Spinal Cord Injury	2013	National	N/A	NR	NR	NR
USA							
AARC clinical practice guideline. Long-term invasive mechanical ventilation in the home--2007 revision & update(128)	American Association for Respiratory Care (AARC)	2007	National	Yes	NR	NR	NR
Respiratory Management Following Spinal Cord Injury: A Clinical Practice Guideline for Health-Care Professionals(132)	Consortium for Spinal Cord Medicine	2005	National	No	Yes	Yes	Administrative and financial support provided by Paralyzed Veterans of America

† This document which focused on amyotrophic lateral sclerosis (ALS) was included as it had criteria that were relevant for invasive mechanical ventilation regardless of diagnosis

‡ For these reports, two related guidance documents were combined. Therefore 17 guidance documents are referred to within 15 unique reports.

Key: DGP - German Society for Pneumology and Respiratory Medicine NR: Not Reported; N/A: Not Applicable

Of note, one of the included guidance documents (the Canadian Spinal Cord Injury Practice (Can-SCIP) Guideline)(90) is based on a synthesis of 41 previous SCI clinical practice guidelines. Many of these 41 clinical practice guidelines, including international guidance by the WHO,(4) were identified as part of the literature search for the current review. In order to avoid duplication, the evaluation team excluded any clinical practice guideline already included in the Can-SCIP guideline. An exception was made for one particular guideline by the Canadian Thoracic Society(134) on the topic of home mechanical ventilation, given the specific relevance of this topic to the current review, and the potential for additional relevant guidance that may not have been captured as part of the Can-SCIP recommendations. The remaining 40 clinical practice guidelines contained within Can-SCIP were screened, but none contained the primary outcome of interest (that is, domiciliary invasive ventilation guidance), and hence the risk for missing key guidance was considered low. Guidance that was relevant for this review was extracted from among the 585 recommendations contained within the Can-SCIP guideline.

4.3.3. Overview of guidance

Relevant guidance from across a broad range of domains was extracted from all 17 guidance documents. Of note, the two Polish documents were extracted as a single report given the significant overlap in reporting.(131, 135) Similarly, the two German documents were extracted as a single report, as they comprised two parts of one large guidance document.(79, 139) Therefore, 15 discrete reports of 17 guidance documents are described in this scoping review. While the Can-SCIP guideline covers a large number of domains, it is important to remember that this guideline is based on a synthesis of 41 previous clinical practice guidelines; therefore caution is advised when directly comparing against the other included guidance documents.(90) Additionally, the Can-SCIP guideline aimed to provide an overview of guidance across the entire care continuum for patients with SCI and was not focused on community-based mechanical ventilation guidance per se. Therefore the Can-SCIP guideline, while comprehensive and methodologically rigorous, should not necessarily be viewed as the gold standard for the purpose of this scoping review. A summary of the topics discussed within each discrete report is outlined in Tables 4.4-4.6.

For the purpose of this Chapter, a high level overview of the guidance is discussed for each of the three main concepts:

- domiciliary invasive ventilation
- management of SCI complications
- broader SCI care pathways and supports.

More detailed information on all of the guidance is available in Appendix 3.

Table 4.4: Overview of domains addressed across included guidance documents (domiciliary invasive ventilation guidance)

	Domiciliary invasive ventilation							
Guideline Name	**Pre-transition phase**	**Transition phase**	**General principles**	**Technical requirements**	**Staffing**	**Monitoring**	**Infection prevention and control**	**Other guidance**
Australia								
Guidance on the support needs for adults with spinal cord injury (3rd Ed)(133)	✓		✓	✓	✓			✓
Canada								
Canadian Spinal Cord Injury Practice Guideline (Can-SCIP)(90)*	✓		✓					
Home mechanical ventilation: A Canadian Thoracic Society clinical practice guideline(134)	✓	✓	✓	✓	✓			
Germany								
German National Guideline for Treating Chronic Respiratory Failure with Invasive and Non-Invasive Ventilation: Revised Edition 2017 – Part 1 & Part 2(79, 139)‡	✓	✓	✓	✓	✓	✓		
Italy								
Recommendations – Guidelines for long term home mechanical ventilation (Piedmont Region)(136)	✓	✓	✓	✓		✓	✓	
Regional guidelines: Home Mechanical Ventilation. Description and acquisition of pulmonary ventilators for hospital and home treatment for chronic respiratory insufficiency and sleep breathing disorders (Apulia Region)(127)	✓		✓	✓	✓			

	Domiciliary invasive ventilation							
Guideline Name	**Pre-transition phase**	**Transition phase**	**General principles**	**Technical requirements**	**Staffing**	**Monitoring**	**Infection prevention and control**	**Other guidance**
Regional guidelines: home mechanical ventilation. Adults and pediatrics (Campania Region)[129]	✓		✓	✓		✓		
Netherlands								
Chronic Ventilation[138]	✓	✓		✓	✓	✓		✓
New Zealand								
Spinal cord injury guidelines[126]	✓				✓			
Poland								
Practical aspects of nursing care provided to patients diagnosed with amyotrophic lateral sclerosis receiving home mechanical ventilation & Amendment of the Regulation on guaranteed benefits in the field of nursing and care services as part of long-term care [131, 135] †‡	✓			✓	✓			
Switzerland								
Long-Term Mechanical Ventilation: Recommendations of the Swiss Society of Pulmonology[88]	✓			✓		✓		
United Kingdom								
Service Specification: Spinal Cord Injury Clinical Network – Standards of Care 2022[130]	✓	✓			✓	✓		

	Domiciliary invasive ventilation							
Guideline Name	**Pre-transition phase**	**Transition phase**	**General principles**	**Technical requirements**	**Staffing**	**Monitoring**	**Infection prevention and control**	**Other guidance**
Carer training – Standards expected in terms of knowledge and skills Consensus statement on behalf of RISCI (GBI)(137)								
USA								
AARC clinical practice guideline. Long-term invasive mechanical ventilation in the home--2007 revision & update(128)	✓		✓	✓	✓	✓	✓	
Respiratory Management Following Spinal Cord Injury: A Clinical Practice Guideline for Health-Care Professionals(132)	✓			✓	✓			

† This document which focused on ALS was included as it had criteria that were relevant for invasive mechanical ventilation regardless of diagnosis

‡ For these reports, two related guidance documents were combined. Therefore 17 guidance documents are referred to within 15 unique reports.

* Based on a synthesis of 41 clinical practice guidelines

Key: AARC - American Association for Respiratory Care

Table 4.5: Overview of domains addressed across included guidance documents (management of SCI complications)

Guideline Name	Management of SCI complication														
	Respiratory	**Bladder**	**Bowel**	**Skin**	**Sexual health**	**VTE**	**Pain**	**Autonomic dysreflexia**	**Emotional wellbeing, mental health and substance abuse**	**Cardiometabolic and nutrition**	**Bone Health**	**Palliative care**	**Rehabilitation**	**Wheeled mobility**	**Other guidance**
Australia															
Guidance on the support needs for adults with spinal cord injury (3rd Ed)(133)															
Canada															
Canadian Spinal Cord Injury Practice Guideline (Can-SCIP)(90)*	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
Home mechanical ventilation: A Canadian Thoracic Society clinical practice guideline(134)	✓														
Germany															
German National Guideline for Treating Chronic Respiratory Failure with Invasive and Non-Invasive Ventilation: Revised Edition 2017 – Part 1 & Part 2(79, 139) ‡												✓			
Italy															
Recommendations – Guidelines for long term home mechanical ventilation (Piedmont Region)(136)															
Regional guidelines: Home Mechanical Ventilation. Description and acquisition of pulmonary ventilators for hospital and home treatment for chronic respiratory															

Guideline Name	**Management of SCI complication**														
	Respiratory	**Bladder**	**Bowel**	**Skin**	**Sexual health**	**VTE**	**Pain**	**Autonomic dysreflexia**	**Emotional wellbeing, mental health and substance abuse**	**Cardiometabolic and nutrition**	**Bone Health**	**Palliative care**	**Rehabilitation**	**Wheeled mobility**	**Other guidance**
insufficiency and sleep breathing disorders (Apulia Region)(127)															
Regional guidelines: home mechanical ventilation. Adults and pediatrics (Campania Region)(129)															
Netherlands															
Chronic Ventilation(138)	✓											✓		✓	
New Zealand															
Spinal cord injury guidelines(126)														✓	✓
Poland															
Practical aspects of nursing care provided to patients diagnosed with amyotrophic lateral sclerosis receiving home mechanical ventilation & Amendment of the Regulation on guaranteed benefits in the field of nursing and care services as part of long-term care(131, 135) †‡															
Switzerland															
Long-Term Mechanical Ventilation: Recommendations of the Swiss Society of Pulmonology(88)	✓														
United Kingdom															

Guideline Name	**Management of SCI complication**														
	Respiratory	**Bladder**	**Bowel**	**Skin**	**Sexual health**	**VTE**	**Pain**	**Autonomic dysreflexia**	**Emotional wellbeing, mental health and substance abuse**	**Cardiometabolic and nutrition**	**Bone Health**	**Palliative care**	**Rehabilitation**	**Wheeled mobility**	**Other guidance**
Service Specification: Spinal Cord Injury Clinical Network – Standards of Care 2022(130)															
Carer training – Standards expected in terms of knowledge and skills Consensus statement on behalf of RISCI (GBI)(137)															
USA															
AARC clinical practice guideline. Long-term invasive mechanical ventilation in the home--2007 revision & update(128)															
Respiratory Management Following Spinal Cord Injury: A Clinical Practice Guideline for Health-Care Professionals(132)	✓				✓	✓	✓		✓						✓

† This document which focused on ALS was included as it had criteria that were relevant for invasive mechanical ventilation regardless of diagnosis

‡ For these reports, two related guidance documents were combined. Therefore 17 guidance documents are referred to within 15 unique reports.

* Based on a synthesis of 41 clinical practice guidelines

Key: AARC - American Association for Respiratory Care; SCI - Spinal Cord Injury; VTE – Venous Thromboembolism

Table 4.6: Overview of domains addressed across included guidance documents (broader SCI care pathways and supports)

Guideline Name	Broader SCI care pathways and supports							
	Governance structure	Funding model	Care pathways	Education across the continuum of care	Safeguarding and ethics	Other supports	Quality assurance processes	Other guidance
Australia								
Guidance on the support needs for adults with spinal cord injury (3rd Ed)(133)	✓	✓		✓	✓	✓		✓
Canada								
Canadian Spinal Cord Injury Practice Guideline (Can-SCIP)(90)*			✓	✓	✓	✓		
Home mechanical ventilation: A Canadian Thoracic Society clinical practice guideline(134)		✓	✓	✓	✓	✓		
Germany								
German National Guideline for Treating Chronic Respiratory Failure with Invasive and Non-Invasive Ventilation: Revised Edition 2017 – Part 1 & Part 2(79, 139)	✓		✓	✓	✓			
Italy								
Recommendations – Guidelines for long term home mechanical ventilation (Piedmont Region)(136)	✓			✓		✓		
Regional guidelines: Home Mechanical Ventilation. Description and acquisition of pulmonary ventilators for hospital and home treatment for chronic respiratory insufficiency and sleep breathing disorders (Apulia Region)(127)	✓	✓				✓		

Regional guidelines: home mechanical ventilation. Adults and pediatrics (Campania Region)(129)	✓	✓	✓					
Netherlands								
Chronic Ventilation(138)	✓	✓	✓	✓	✓			✓
New Zealand								
Spinal cord injury guidelines(126)								
Poland								
Practical aspects of nursing care provided to patients diagnosed with amyotrophic lateral sclerosis receiving home mechanical ventilation & Amendment of the Regulation on guaranteed benefits in the field of nursing and care services as part of long-term care (131, 135) †		✓		✓				
Switzerland								
Long-Term Mechanical Ventilation: Recommendations of the Swiss Society of Pulmonology(88)	✓			✓				
United Kingdom								
Service Specification: Spinal Cord Injury Clinical Network – Standards of Care 2022(130)			✓	✓				
Carer training – Standards expected in terms of knowledge and skills Consensus statement on behalf of RISCI (GBI)(137)				✓				
USA								
AARC clinical practice guideline. Long-term invasive mechanical ventilation in the home--2007 revision & update(128)								
Respiratory Management Following Spinal Cord Injury: A Clinical Practice Guideline for Health-Care Professionals(132)		✓		✓	✓	✓		

† This document which focused on ALS was included as it had criteria that were relevant for invasive mechanical ventilation regardless of diagnosis

‡ For these reports, two related guidance documents were combined. Therefore 17 guidance documents are referred to within 15 unique reports.
* Based on a synthesis of 41 clinical practice guidelines

Key: SCI - Spinal Cord Injury; AARC - American Association for Respiratory Care

4.3.4. Domiciliary invasive ventilation guidance

4.3.4.1. Discharge process

Sixteen of the 17 included guidance documents discuss the critical importance of the discharge process.(79, 88, 90, 126-136, 138, 139) The British Association of Spinal Cord Injury Specialists (BASCIS) advise that the discharge process and planning should start from the time of diagnosis, throughout the individual's initial rehabilitation, and should be conducted in a collaborative manner with involvement of the individual, family and friends (as desired), local services and specialists.(130) The Canadian Thoracic Society advise that *"once a ventilated patient has been identified as suitable for home mechanical ventilation (HMV), the in-hospital care routines should be reviewed and changes implemented as quickly as possible. This review is to be considered from the perspective of limited professional caregiver supervision and intervention in the community."*(134) The individual needs to be in a stable medical condition prior to discharge and should be transferred to, and be comfortable with, the ventilation machine that will ultimately be used in the home setting.(79, 139) A thorough assessment of the individual's living situation may be necessary prior to medically approving discharge given the safety risks associated with domiciliary invasive ventilation.(134, 138) It is important that the individual and their family are fully informed of the risks (and benefits) of domiciliary invasive ventilation. A quality indicator within the German National Guideline is that domiciliary invasive ventilation is only established after consent is given by the extensively informed patient and/or their legal guardian.(79, 139)

There are very clear recommendations from the Canadian Thoracic Society regarding the requirements for a person to be discharged home for invasive ventilation. They state that, before discharge can happen, the candidate for domiciliary invasive ventilation:(134)

- should be medically stable
- must be motivated (along with their family)
- must have an adequate home setting
- must have sufficient caregiver support
- must have access to adequate financial resources
- must have access to equipment appropriate for their needs.

Individuals requiring mechanical invasive ventilation at home need substantial supports, and this is especially true if these individuals also have a spinal cord injury. The icare (insurance and care) guidance document from New South Wales, Australia, advises that decisions surrounding support requirements for Patients with SCI need to take account of the person's goals, context and progress.(133) The icare guidance notes that there are many factors to consider when deciding on the individual's need for support. The key considerations, and action points relating to exploring these considerations, include:

- Why assistance is needed:
    - understand the person
    - understand the person's context
    - understand the person's progress.
- Matching the need for assistance with the supports such as:
    - the type of formal and informal supports that exist within the home and community
    - whether additional assistive technology is required or would assist
    - for whom, how and when assistance is needed.
- Consideration of practical matters including:
    - criteria for funding
    - 'when, how and who' will ensure that all the informal and formal supports (including support workers) are integrated and coordinated
    - each person's circumstances related to the criteria for funding.(133)

Individuals should not be discharged until such a time as the hospital is confident that the ventilation and care can be continued safely, effectively and comfortably at home. The Dutch guidance document recommends that on the day of discharge a liaison nurse visits the individual in their home to check whether the ventilator has been set up properly, to answer any further questions and if necessary, and to provide additional training to informal and or professional care givers.(138)

Given the complexity of the discharge process for these individuals, some guidance documents recommend having an explicit checklist to ensure that all required equipment, training, funding and supports have been obtained prior to the individual being discharged.(79, 88, 129, 136, 139) The German National Guideline specifies that provision of such a checklist to the individual upon discharge from hospital should be considered a quality indicator for that service.(79, 139)

A trial discharge is recommended by the Canadian Thoracic Society, who advise that a night or two at home enables the individual, the carers and health providers to understand gaps in the preparation for discharge to the community. The guideline recommends that once the individual and their families return to the hospital, they should be encouraged to share concerns or difficulties experienced in the home trial with the hospital care providers who will be able to troubleshoot these issues.(134)

4.3.4.2. Communication and monitoring post-discharge

A clear and open line of communication between the discharging hospital, the individual and their family, and the community healthcare providers is advised,(134) with an explicit recommendation in two documents that the hospital be contactable 24/7 to offer professional support, if required.(88, 138) Three guidance documents specify that the transition period must involve close collaboration between the discharging hospital team and the individual's general practitioner (GP) and community-based respiratory physician,(134) with the GP receiving sufficient notice about the individual's complex care needs prior to discharge.(136, 138) BASCIS advise that referrals to the local relevant medical specialists (for example, Respiratory, Ear Nose & Throat (ENT) or Psychiatry), and, if desired, support from SCI charities, should be completed prior to discharge.(130) BASCIS also specify that discharge information must be made available to the primary carer, GP, local nursing team, physiotherapy/occupational therapy teams and psychological team, and the required services and supports organised.(130) Care coordination to ensure that informal and formal supports work properly and well together is noted to be critical for all those involved, and particularly for the person with spinal cord injury.(133)

A comprehensive assessment of the individual in their home should be carried out by an interdisciplinary health care team from the discharging hospital.(134) Recommendations regarding how often domiciliary ventilated-individuals should receive follow-up visits from the hospital healthcare team vary.(79, 88, 128, 130, 134, 136, 138, 139) However, there is a general consensus that the first follow-up visit should occur within the first few weeks or months of discharge, with the visits subsequently becoming less frequent (often every six to 12 months thereafter). An individualised approach to determining the frequency of follow-up visits is advocated by the American Association for Respiratory Care (AARC).(128)

4.3.4.3. Equipment

Eight reports detail the equipment requirements for domiciliary invasive ventilation.(127-129, 131-133, 135, 136, 138) A substantial amount of equipment is recommended for those requiring domiciliary invasive ventilation, and these recommendations vary depending on the individual's underlying diagnosis, level of injury (in the case of SCI) and co-morbidities. For example, individuals who have high cervical cord injuries will have greater equipment requirements than those with lower spinal cord injuries, as the former patient cohort will often have no or limited neural control of breathing, urinating and their bowels.(133) At a minimum, the US-based Consortium for Spinal Cord Medicine recommend supplying the following equipment for individuals living at home who require mechanical ventilation following SCI:(132)

- two portable ventilators and all ancillary supplies and equipment (for example, suction, oxygen, IV therapy, nutritional therapy)
- an electric hospital bed with adjustments and appropriate overlay or mattress
- a reclining padded commode chair for toileting and showering
- a mechanical power lift for safe transfers
- a power wheelchair that allows for patient mobility, independent weight shifts, and portable ventilation
- a backup manual wheelchair.

They also advise that provision of emergency call systems and technology (for example, environmental control system, voice-activated computer and telephone) may enhance the independence of the individual.

Provision of a second/back-up ventilator is recommended if survival is dependent on mechanical ventilation (such as in those with high cervical cord injury) in case of primary ventilator failure, and also to enable mobilisation in a wheelchair.(79, 88, 127-129, 132, 133, 136-139) Five guidance documents also reference the importance of having two active alarms – one for accidental disconnections and the other for high pressure detection.(79, 88, 128, 138, 139) The Dutch guidance further specifies the level of healthcare provider supervision required (ranging from remote assistance to direct supervision) depending on the individual’s care needs (for example, the individual can safely be helped within 30 minutes or the individual must be helped immediately) and alarm capability (for example, device linked to nurse and or doctor call system, or device not linked to any external monitoring system).(138) Any change of ventilator should only occur under medical surveillance in hospital.(79, 139)

The importance of maintaining adequate power supply to the home is emphasised in many of the guidance documents.(79, 88, 128, 129, 131, 133, 134, 136, 138, 139). For example, the AARC recommend the following approach to ensuring an adequate power supply:(128)

- alternating current (AC) is the primary power source for most long term ventilators.
- direct current (DC) by external battery may be used to allow mobility and as an emergency power source. The internal battery of the ventilator should only be for short-term use; it should not be used as a primary source of power.
- a portable generator may be required if frequent power outages occur or if the home is in a remote location.

4.3.4.4. Staffing

It is acknowledged in the Dutch guidance that there is *"no literature available on the standards for staffing in ventilators"*(138) and so recommendations vary. However, there is consistency across guidance documents that 24/7 care, involving at least one trained caregiver, is essential for these individuals due to their complex healthcare needs.(126, 128, 131, 133-135) Recommendations regarding the minimum total estimated caregiver time for high cervical cord injured adults (C1-C3) with no motor function below this level range from 168 hours to 196 hours per week (the latter therefore entailing more than the equivalent of one person providing care on a 24/7 basis for the whole week), with significant assistance required for transfers, care and domestic activities.(126, 133, 134)

Recommendations differ in terms of whether a nurse is required to provide this 24/7 care or whether a suitably trained healthcare assistant/nursing aide under nursing supervision is adequate. The New Zealand guidance states the following in relation to C1-C3 injured adults (ASIA A or B classification): *"If the person is medically stable, attendant care is generally provided under the supervision of a registered nurse, by people who have successfully completed competency-based training. If the treating team identifies that the person is significantly medically unstable (for example, with severe dysreflexia) this situation may best be managed with appropriately skilled registered nurses providing all attendant care. However, there may be some circumstances where this level of care is not available (for example, in remote geographic areas). In all cases, access to a registered nurse for support and advice is required at all times with all programs routinely and regularly reviewed by an appropriately skilled registered nurse."*(126) In contrast, the German guidance explicitly states that only registered nurses with specialised training can provide independent care for individuals who are ventilated: "*Nursing aids, physician assistants, and remedial therapists may only work in the ventilation care branch as a part of a nursing team with nursing specialists (shared living communities or inpatient nursing facilities). Independent specialised nursing care of ventilated individuals is only possible with the [relevant] qualifications."*(79, 139) The German guidance does however advise that the extent of nursing care can be stepped down under certain circumstances (for example, stable medical condition, access to equipment and funding etc.) based on consensus by the entire multidisciplinary team in conjunction with the individual concerned and their family.(79, 139) The US AARC refers to "credentialed" health care professionals (namely, a registered/certified respiratory therapist or a registered nurse) *"capable of providing direct patient care and possessing demonstrated competencies to monitor and assess both the patient and equipment"*. The AARC also advise that lay caregivers, such as family members and other healthcare personnel, can be trained to undertake certain tasks and techniques, though they must be appropriately trained and must demonstrate competency in specific areas.(128) Other guidance documents refer to "trained" or "sufficiently skilled" staff without necessarily stating whether staff need to be registered nurses or not.

The Dutch guidance document refers to national legislation, which differentiates between routine medical acts that can be performed by any professional caregiver and those that are 'reserved', that is, those which carry significant risk and so may only be carried out by certain authorised professions.(138) The guidance documents state that care of individuals requiring invasive mechanical ventilation involves a large degree of 'reserved' acts, such as, changing tracheal cannulas. The legislation details which professions are independently authorised to carry out 'reserved' acts on their own authority (these include doctors, dentists, physician assistants and nurse specialists). Other professionals (for example, other nurses, healthcare assistants) may carry out 'reserved' acts at the request of an independently authorised individual, as long as the person is competent in performing that task, has an action schedule, and can contact the SCI specialist nurse on duty in case of emergency. While the legislation is specifically concerned with professional caregivers (that is, those that are paid for their work), the guidance document states that in the context of non-professional care providers (such as family members and friends), such 'reserved' acts can be performed at home provided that these individuals are sufficiently trained and similar standards of care are applied.

The Dutch guidance also advises that consideration be given to relevant patient-related (for example, the individual's care needs, ventilation-free time, hand function, ability to speak, etc.) and organisational factors (for example, care demands, maximum distance from the individual that staff can be at any given time, availability of technology, etc.) when determining the level of skill required by staff. The preference of the individual should always be taken into account when considering the options.(138)

4.3.5. Management of SCI complications

Patients with high cervical cord injuries generally have multiple, complex healthcare needs, often as a direct or indirect consequence of the SCI.(90) Management of these SCI complications in a home environment presents additional challenges, as multiple specialties are needed to ensure safe and effective care. Care coordination is defined as *"a proactive approach in bringing care professionals and providers together around the needs of service users to ensure that people receive integrated and person-focused care across various settings"*. As previously mentioned, care coordination is critical to ensure that informal and formal supports, from all required disciplines, work effectively together to ensure the best outcomes for the individual with SCI.(133)

The majority of recommendations relating to the management of SCI complications came from the Can-SCIP guideline, which aims to provide comprehensive evidence-based recommendations in all phases of care across an individual's lifetime.(90) Across all included guidance documents, recommendations are provided in relation to the management of the following issues in patients with SCI:

- respiratory complications(88, 90, 132, 134, 138)
- bladder complications(90)
- bowel complications(90)
- skin integrity(90)
- sexual health(90, 132)
- venous thromboembolism(90, 132)
- pain(90, 132)
- autonomic dysreflexia(90)
- emotional wellbeing, mental health and substance abuse(90, 132)
- cardiometabolic disease and nutrition(90)
- bone health(90)
- palliative care.(79, 90, 138, 139)

Guidance documents also provide recommendations regarding wheeled mobility and transport,(90, 126, 138) assistance with daily activities of living,(126, 133) rehabilitation,(90) secondary mild brain injury(132) and a range of therapy options.(90)

4.3.6. Broader SCI care pathways and supports

4.3.6.1. Education and training

The provision of education and training programmes for those providing care is considered critical for the safe and effective care of individuals requiring invasive mechanical ventilation at home.(79, 90, 130-139) Eleven of these guidance documents also outline the competencies that need to be demonstrated, and or the minimum topics that need to be covered in such education and training programmes, before someone is considered competent to care for an individual requiring invasive mechanical ventilation outside of a hospital setting.(79, 90, 130-132, 134-139) For example, the Respiratory Information in Spinal Cord Injury (RISCI) Carer Training guidance document states that the home care provider should evidence that their staff have been trained or have access to training for all of the following five standards, and that this training is updated regularly(137):

1. Ventilation (for example, setting up the ventilator, checking and documenting against prescription and configuration)
2. Interface (for example, understanding specific features of chosen interface)
3. Tracheostomy (for example, providing routine care of the stoma and tracheostomy tube)
4. Management of associated equipment (for example, correctly setting up of all associated devices)
5. Emergency and urgent care needs (for example, understanding the need for, and having access to 24 hour support).

Providing education, information and support to the individual with SCI and to their family, as early as possible and throughout the individual's life course, is also recommended in seven guidance documents.(79, 90, 130, 132, 134, 138, 139) A holistic and individualised approach to the education of the individual and their family is recommended, in which their specific social, emotional, educational, and cultural needs are met.(132) The importance of centrally involving the individual with SCI in the education and training is highlighted in the Canadian Thoracic Society guidance, where they recommend that these individuals "…*should be taught to direct and or perform their own care, given that occasionally the individual may find themself in situations where they have to guide an untrained bystander to perform care"*.(134) BASCIS recommend providing a 'relative's day' once a year, given that the education required will change over time and this new information will need to be available in order to provide lifelong support.(130) Can-SCIP recommend that *"individuals with SCI should have timely access to local peer support services and community-based programs to increase the quality of life and community participation after injury across their lifespan."*(90)

4.3.6.2. Governance structures and care pathways

Eleven of the 17 included guidance documents report on the governance structure and or care pathways for individuals who are invasively ventilated at home.(79, 88, 90, 127, 129, 130, 133, 134, 136, 138, 139) It is important to emphasise that nine of these 11 guidance documents are not specific to individuals with SCI, and so are relevant for any individual requiring domiciliary invasive ventilation regardless of the underlying diagnosis.(79, 88, 127, 129, 133, 134, 136, 138, 139)

The Dutch guidance provides detail about the organisation and governance of domiciliary ventilation services in the Netherlands.(138) The document outlines how there are four designated centres for chronic ventilation (called home ventilation centres) in the country, with each centre serving a different region (Groningen, Utrecht, Rotterdam and Maastricht). Each home ventilation centre is affiliated with a teaching hospital, and provides specialised care to individuals with chronic respiratory insufficiency regardless of the underlying disease (for example, amyotrophic lateral sclerosis (ALS) which is a sub-set of motor neurone disease, obesity hypoventilation syndrome (OHS) and SCI). Referrals to these home ventilation centres can be made electively from the outpatient setting or from the inpatient setting following an acute event. It is acknowledged that there are many different specialties involved in the care of such individuals and that the provision of care is particularly complicated when the person transitions home. The Dutch guidance specifies that when the individual is set up in their home, their GP becomes their main practitioner, but the specialist doctor from the discharging home ventilation centre is a 'co-practitioner' and remains closely involved in the person's care. The guidance states that *"in order to prevent ambiguity and therefore dangerous situations for the patient, clear agreements must be made about main and co-practitionership, consultancy and about which information is essential to transfer. It must also be clear to the patient and their carers who, in each situation, is the contact person to be addressed."* The home ventilation centre should be contactable 24/7 to provide any required medical advice to the GP. The guidance document also outlines roles and responsibilities in relation to other specific scenarios such as hospital and nursing home admissions, equipment purchase, failure and maintenance, ventilator adjustments, and transfers.

Similar recommendations are outlined in the German guidance document.(79, 139) Home mechanical ventilation (HMV) centres exist in Germany where specialised teams set up, monitor and optimise home ventilation therapy for individuals with chronic respiratory failure (regardless of underlying cause). The German guidance states that *"the outpatient treating physician must undertake the responsibility of outpatient treatment of a patient receiving home mechanical ventilation therapy,"* and that "*the supervising home mechanical ventilation centre must become involved where necessary and be available for advice."* The guidance specifies that *"the [outpatient] physician should therefore be able to demonstrate experience in out-of-hospital ventilation and should carry out home visits. With the relevant qualifications, this task can be taken on, for example, by a general practitioner, similarly by a respiratory specialist, an anaesthetist, a paediatrician, a neurologist, an internal medicine specialist, or a specialist with extra training in intensive care medicine and hence mechanical ventilation. If the required medical expertise is not available, the supervising HMV centre should become involved on an advisory basis."* The guidance recommends that physiotherapy, speech therapy, and occupational therapy should form an inherent part of the outpatient treatment programme for individuals requiring home ventilation, and that these therapists should demonstrate familiarity with the ventilators and associated equipment if they are working with the individual in the absence of a nurse competent in this area.(79, 139)

The importance of a comprehensive, robust and round-the-clock equipment maintenance service is discussed in four guidance documents.(79, 127, 138, 139) The Dutch guidance document recommends that *"the equipment provider must carry out an initial briefing on the ventilation device in accordance with medical product laws and guarantee permanent availability with a prompt, needs-based service."* Similarly, the German guidance also refers to this need for checks by the equipment provider.(79, 139)

4.3.6.3. Ethics and safeguarding

Issues relating to ethics and safeguarding are discussed in seven of the included guidance documents.(79, 90, 132-134, 138, 139) The importance of respecting the individual's autonomy, dignity and confidentiality is emphasised. In particular, undertaking advanced care planning (ACP) or advanced health care directives (AHCD) as early as possible, in conjunction with the medical team, and re-evaluating regularly, is recommended.(79, 132, 134, 138, 139) The Canadian Thoracic Society advises that doctors *"proactively counsel capable patients and establish clear advanced directives (regarding issues such as crisis management and end-of-life care) in a timely manner, ensuring that patients fully understand and appreciate the reasonably foreseeable outcomes of their decisions. Physicians must work with patients to help prioritise their values, interests and preferences."(134)* In relation to individuals that lack decision-making capacity, the Canadian Thoracic Society advises that *"substitute decision-makers and clinicians must incorporate the patient's advance care directives in the decision-making process or, where there are no known advance care directives, to act in the patient's 'best interests'."(134)* Providing additional supports for individuals who are vulnerable (due to physical, economic, social, or emotional vulnerability) is discussed in the Can-SCIP guidelines.(90)

Withdrawal of mechanical ventilation, thereby allowing a person to die, is discussed in the German guidance document in the context of German law.(79, 139) The guidance states that "*if there is no hope even for the stabilisation of quality of life that corresponds to the expectation of a home-ventilated end-of-life patient, it is ethically justified to discuss and (if necessary) undertake a change in therapy goals in the form of therapy limitation or withdrawal. Once mechanical ventilation has been started it should neither be forcefully or automatically continued nor must it be potentially "terminated" or "withdrawn." Instead, mechanical ventilation therapy should, like other treatments (medication, nutrition, infusion therapy, etc.), be checked regularly (e.g., daily), with strict consideration of the patient's will for its ongoing justification/indication. Termination of mechanical ventilation in such a situation is designated as passive euthanasia in Germany. This is legally allowed and* *ethically justified [in Germany]."*(79, 139) Withdrawal of mechanical ventilation therapy is not specifically addressed in any of the other included guidance documents.

4.4. Discussion

In this Chapter, a scoping review of international practice and standards relevant to the provision of permanent domiciliary invasive ventilation in adults with spinal cord injury was undertaken. A broad range of practice and standards was identified across 17 guidance documents from 10 different countries, covering three main areas: domiciliary invasive ventilation; management of SCI complications; and broader SCI care pathways and supports.

Patients with high cervical cord injuries requiring invasive mechanical ventilation have complex health care needs, and require substantial support from formal and informal caregivers in a home setting. While not all included guidance documents were specific for patients with SCI, there were many commonalities identified in terms of recommendations for a safe and effective domiciliary invasive ventilation service. In general, the guidance documents all discuss the importance of: appropriate preparation and timing of the transition to the home setting; anticipation of present and future requirements in terms of environment and training of those providing care; and anticipation of disease progression. A comprehensive, holistic and person-centred approach to care, delivered from a well-coordinated interdisciplinary team, which respects the preferences of the individual, is advocated in these guidance documents. Given the substantial amount of equipment and staff required to provide a 24/7 domiciliary invasive ventilation service, there is clear guidance that adequate funding and acquisition of these resources must be obtained prior to discharge. Clear communication and collaboration across disciplines and settings, with a clear understanding of roles and responsibilities and the overarching governance structure, is recommended throughout. Advanced care planning is also recommended so that the wishes of the individual are understood by all involved in an emergency or end of life situation.

While there is substantial agreement between these guidance documents in relation to the core components of a safe and effective domiciliary ventilation, some differences in practice are noted. For example, the German guidance document emphasises that only appropriately qualified nurses should be providing care to individuals receiving domiciliary invasive ventilation.(79, 139) Whereas the New Zealand guidance acknowledges that, where possible for a medically unstable individual, a suitably qualified nurse should be providing direct patient care, but otherwise care may be provided "*by people who have successfully completed competency-based training.*"(126) Most included guidance documents do not specify the requirement for nurses, but rather focus on staff (of whatever qualification) being required to demonstrate competence to care for an individual requiring invasive mechanical ventilation outside of a hospital setting.

Despite the comprehensive search and broad inclusion criteria, only a limited number of guidance documents were identified. Only 10 of the 17 included documents contain SCI-specific guidance regarding domiciliary invasive ventilation, and no single document was found to address all relevant aspects of care.(79, 88, 90, 126, 130, 132-134, 137, 139) An evaluation of the quality of published SCI clinical practice guidelines, performed by the Can-SCIP guideline expert panel, highlighted this paucity of guidelines specific for the community setting.(140) This suggests that there is a gap in the literature regarding guidance documents specifically for patients with SCI living at home who require invasive mechanical ventilation. In particular, gaps were identified in the provision of advice (in that it was either absent or scarce) regarding infection prevention and control measures, safeguarding and ethics, governance and funding, and quality assurance processes. Future guidance documents should include these elements to enable a quality domiciliary invasive ventilation service that considers all aspects of care delivery.

4.4.1. Strengths and limitations

A strength of this scoping review is that a comprehensive search, without date or language restriction, and inclusive of grey literature, was undertaken. This resulted in the identification of a number of relevant guidance documents – each with a substantial volume of information. A limitation of this scoping review is that, despite the broad search strategy adopted, the included guidance all originated from high income countries in Europe, North America and Australia, with no relevant guidance identified from Asia, South America or Africa. It is important to note that the fact that no guidance document was identified from these other regions does not necessarily mean that these regions do not provide domiciliary invasive ventilation services, but rather that their documents may not be easily identifiable. Searching for grey literature is a known challenge and often requires screening a substantial amount of alternative sources (for example, websites, books and guidance documents),(141) and hence the grey literature search undertaken was limited by the resources available to the team. Additionally, while there were no language restrictions within the search strategy, the use of English terms only may have limited the ability to detect relevant guidance published in other languages. Hence, there may be a bias in the findings towards English speaking countries. It must be acknowledged that there are differences across countries in terms of the healthcare systems and available resources to support domiciliary ventilation of patients with SCI. Given that the countries from which the identified guidance originated are all high income countries, it is likely that the healthcare systems are broadly similar to that in Ireland. However, differences in terms of staff resources and the geographic distribution (of staff, patients and SCI centres) may influence recommendations.

Additionally, given the comprehensive search strategy that was adopted, which included a supplementary manual search of relevant agencies and societies, the evaluation team is reasonably confident that the majority of important guidance documents have been identified. Further, given the largely similar nature of much of the guidance located within these documents, it is likely that a 'saturation point' has been reached, and additional documents may not provide any new relevant information.

4.4.2. Conclusion

Provision of care to adults with spinal cord injuries who require permanent mechanical invasive ventilation in a home setting is complex and requires careful consideration of a broad range of issues. While international standards and practice in this area may vary, there are many common recommendations that may be useful to inform a national provision of service in Ireland.

5. Budget Impact Analysis

Key points

- A budget impact analysis (BIA) was undertaken to estimate the costs of providing a comprehensive homecare package to a patient with spinal cord injuries (SCI) who requires invasive mechanical ventilation, as well as the costs of developing a bespoke trainer and support role on a national footing.
- The analysis adopted the perspective of the Irish publicly-funded health and social care system, namely the Health Service Executive (HSE). Only direct medical costs to the HSE were estimated. The BIA projected costs over a five-year time horizon consistent with national guidelines.
- A 'typical' HSE funded homecare package was costed based on international guidance and current practice with input from EAG members.
- For the purpose of this BIA, the comparator was the patient remaining in hospital for five years instead of being discharged home when clinically suitable to do so. Four different costing methods were used to estimate the hospital care costs in the comparator group.
- Costs included inpatient care, equipment and consumables, medication, labour, healthcare service use, and trainer and training costs. Equipment and consumable requirements and costs were obtained through discussions with clinical experts, HSE managers and HSE procurement staff who had oversight of national contracts, invoices and quotes, and were able to provide estimates. Where cost estimates were unavailable, best guess estimates were provided by clinical experts.
- Assuming 196 hours of paid formal care per week (that is, one staff member providing 24/7 care over the course of a week with 28 hours of overlap with a second staff member), the total budget impact for domiciliary ventilation care provision per patient was estimated at €3.4 million over five years. The majority of expenditure over a five-year time horizon was associated with the cost of labour (€2.8 million, 83%).
    - In the event that a patient is provided with 336 hours of paid formal care per week (that is, one nurse and one HCA each providing 24/7 care), over a five-year time horizon, the total budget impact of this

level of care provision was estimated at €4.8 million per patient, which is 42% higher than the base case.

- The total cost of establishing and implementing a new bespoke role in the community that would provide ongoing training and support to patients, families and homecare workers, was estimated at €0.45 million over a five-year period.
- There was substantial uncertainty regarding the five-year incremental budget impact of providing a comprehensive homecare package compared with hospital care for patients, with estimates ranging from €0.58 million cheaper to €1.41 million more costly. It is therefore unclear whether homecare would cost more or less than hospital care for these patients.
- Uncertainty relating to nursing costs was found to contribute most to uncertainty in the total budget impact analysis (lower bound: €2.9 million, upper bound: €3.9 million). Uncertainty in this parameter was associated with 30% variation in the total budget impact over a five-year time horizon.
- Given the substantial contribution of labour to the total costs of a homecare package, scenario analysis was undertaken to examine the impact on the five-year total budget impact of altering the ratio of nursing to healthcare assistant (HCA) care. As the ratio of nursing care to HCA decreases the total budget impact also decreases. Relative to the basecase, in a scenario where it was assumed that there was 28 hours of nursing care and 168 hours of HCA care per week, the total budget impact was reduced by €0.7 million (21%) over five years.
- If there is an expectation that one new patient will require this service every second year, and accounting for the bespoke training and support role, then the resulting five-year total budget impact for the HSE overall is estimated to be between €6.7 million and €9.2 million, with the higher estimate reflecting the cost for three consecutive patients who each require 336 hours of paid care per week. In contrast, assuming a worst case scenario of two new patients every year, the total cost to the HSE overall is estimated to be between €21.2 million and €29.8 million over five years.
- There are other substantial costs that fall on patients and families, as well as other government departments that were not considered as part of this BIA. For example, there are large upfront costs such as housing and car adaptations that, while heavily subsidised by the State, still incur substantial out-of-pocket expenses for patients and families. The uncertainty surrounding medical card approval is another source of worry for many patients and families.

- A limitation of this BIA was the high degree of uncertainty in relation to the estimated hospital care costs. The evaluation team was also unable to determine the costs of providing long-term care for these patients in residential care settings given the lack of available data.

5.1. Introduction

This Chapter describes the budget impact analysis (BIA) undertaken to estimate the costs of providing, on a national footing, a comprehensive homecare package to a patient with SCI who requires invasive mechanical ventilation, as well as the costs of developing a new bespoke role in the community that would provide ongoing training and support to patients, families and homecare workers, over a five-year period. The need for this role was informed by the findings of Chapter 4, in conjunction with feedback from members of the EAG, and is described in detail in Section 5.2.4.

5.2. Methods

The analysis described in this Chapter was conducted in line with the national HTA guideline for the conduct of BIA of Health Technologies in Ireland,(142) using Microsoft Excel 2013.

5.2.1. Study objective

The purpose of this BIA was to estimate the potential financial cost of providing a comprehensive homecare package at the individual patient level, and to estimate the cost of establishing and implementing a new bespoke role in the community that would provide ongoing training and support to patients, families and homecare workers, over a five-year period.

5.2.2. Target population

As outlined in Chapter 2, high cervical traumatic SCI in Ireland are rare, with only 18 such cases (an incidence of 3.6 per million) discharged from the National Spinal Injuries Unit (NSIU) in 2021. Moreover, only 3-5% of patients with tetraplegia require permanent mechanical ventilation.(28, 29) To-date, the clinical experience in Ireland has been that every one to two years, there is, on average, one new patient with SCI who requires permanent mechanical ventilation.(143) In other words, in some years there are no new patients, and in other years there may be one or two. Expert clinical opinion is that there are between five and ten adults with SCI currently availing of domiciliary invasive ventilation in Ireland.(69) For the purpose of this BIA, the analysis was undertaken from a 'per patient' perspective with the aim of informing the potential costs and resources required to provide a 'typical' comprehensive homecare package for a patient with high cervical SCI who requires permanent mechanical invasive ventilation.

5.2.3. Components of a 'typical' homecare package

While it is acknowledged that the care needs for each individual patient will vary substantially depending on the level and extent of their injuries, their age, comorbid conditions, social factors (for example, level of family and financial support, accommodation suitability) and access to services etc.,(133) a 'typical' HSE funded homecare package was costed based on international guidance (Chapter 4) and current practice with input from EAG members. The homecare package developed for the purpose of this BIA is outlined in Table 5.1. However, given the substantial variation in homecare packages that are possible, the costs and resource use estimates presented in this Chapter should be viewed as indicative rather than definitive.

Table 5.1: Components of a 'typical' homecare package for patients with high cervical SCI who require mechanical invasive ventilation

Equipment and consumables
Nebuliser & associated equipment and consumables
2 x portable mechanical ventilators & associated equipment and consumables
Cough assist device & associated equipment and consumables
Phrenic nerve stimulator
2 x Portable suction machine & associated equipment and consumables
A power chair with alternative access
Communication assistive technology & environmental control units
Hi-Lo electric hospital bed
Scan turn mattress
Hoist and tracking hoist
Shower trolley
Oxygen saturation monitor
Capnography monitor
Physiotherapy tilt table
MotoMed
Emergency tracheotomy kit
Ambu Bag
Tracheostomy consumables
Colostomy consumables
Urinary catheter consumables
Personal protective equipment
Healthcare and support staff
Nurse
Healthcare assistant (HCA)/ healthcare support assistant (HCSA)
Clinical Psychologist
Occupational Therapist
Physiotherapist

Medications
Macrogol
Nebulisers - Saline (Hypertonic 7%)
Salbutamol Nebulisers
Carbocisteine
Baclofen
Gabapentin
Vitamin D supplementation
Co-codamol (Paracetamol and codeine)
Healthcare service use
Outpatient visit

5.2.4. Bespoke national training and support community nurse

Informed by the findings of Chapter 4, and in conjunction with feedback from members of the EAG, indicative costings are outlined for a bespoke national training and support nurse role in the community. The aim of such a role would be to provide ongoing training and support to patients availing of mechanical invasive ventilation and their families and homecare workers in the community. The lack of continuity of care in the community and the potential risk this presents to patient care was raised as an important issue by both patients (Chapter 7) and clinicians.(144) From a clinical governance point of view, it was not clear who was responsible for providing training to new homecare providers, particularly in the years and months after the patient has been discharged from the National Rehabilitation Hospital (NRH).(144) This new role could potentially fill this gap and provide a level of consistent training to those providing care to these patients in the community and provide ongoing support to patients and families.

Given that there are many different causes of chronic respiratory failure that may lead to the requirement for mechanical invasive ventilation (for example, motor neurone disease, amyotrophic lateral sclerosis, spinal muscular atrophy, Duchenne muscular dystrophy, post-polio syndrome and other muscular diseases), it is important that such a role is not specific for any one particular condition. Evidence from a national register of domiciliary invasive mechanical ventilation patients in Finland, conducted between 2015 and 2019, found that of the 141 patients availing of this service nationally, 41 had motor neurone disease (29%) and 28 had SCI (20%).(97) These findings highlight that domiciliary invasive mechanical ventilation is not unique to patients with SCI. Providing a critical volume of care may also be important for maintaining technical skills given that domiciliary invasive mechanical ventilation is rare (2 per 100,000 inhabitants in Finland).(97) There may also be other benefits to such an integrated role in terms of knowledge exchange across National Clinical Programmes. The possible components of a bespoke training and support community nurse are outlined in Table 5.2. However, the specifications for this role would need to be clearly defined.

Table 5.2: Possible components of a bespoke national training and support role

Training and support role
Nurse (including travel costs)
Laptop
Online module development and update
Training manual printing
Trainer devices (nebuliser, cough assist, portable suction machine, ventilator and associated equipment and consumables)
Emergency tracheotomy kit
Ambu Bag
Tracheostomy consumables
Personal protective equipment

5.2.5. Comparator

For the purpose of this BIA, the comparator was the patient remaining in hospital for five years instead of being discharged home when clinically suitable to do so. While in practice these patients would rarely spend five full years in hospital awaiting discharge, some patients have spent over three years in hospital in light of the challenges outlined in Section 3.5.(2) It is important to note that there is currently no alternative accommodation type (for example, a nursing home or a step down unit) that provides care for ventilator-dependent SCI patients in Ireland, and so long term residential care was not included as a comparator due to the lack of data available.

5.2.6. Study design

A BIA over a five-year time horizon was undertaken in accordance with national HTA guidelines.(142) The focus was on estimating the total rather than incremental cost, as the purpose is to inform budget and resource planning rather than reimbursement decisions. As discussed in Section 3.5, it is already standard practice to discharge these patients home once safe to do so, however uncertainty remains over the total cost of a 'typical' homecare package.(2) The model included costs relating to equipment and consumables, healthcare and support staff, medications and health service use as outlined in Table 5.1.

However, for illustrative purposes, the incremental cost per patient, defined as the difference in average annual costs between the provision of homecare and hospital care, was also estimated. Separately the total budget impact of the national training and support role was also estimated based on costs as outlined in Table 5.2.

5.2.7. Perspective and time horizon

The analysis adopted the perspective of the Irish publicly-funded health and social care system, namely the Health Service Executive (HSE). Only direct medical costs to the HSE were estimated. Indirect costs such as productivity losses associated with morbidity and mortality, and out-of-pocket expenses incurred by patients and families were not included in the model. However a descriptive summary of the major out-of-pocket expenses incurred by patients and families is provided based on information collated by Spinal Injuries Ireland (SII) – a support and service agency in Ireland for patients with SCI.(145) Costs falling on other government departments (for example Department of Social Protection) are also outlined.

The base case analysis estimated the costs to the HSE of providing a comprehensive homecare package to patients with high cervical SCI who are dependent on mechanical invasive ventilation. The BIA projected costs over a five-year time horizon consistent with national guidelines.(142)

This analysis also estimated the total five-year cost to the HSE of providing domiciliary ventilation services to an estimated three patients, based on a conservative expectation that one new patient will require this service every second year (that is a second and third patient avail of the service, on years three and five respectively), alongside the creation of a bespoke national training and support role.

5.2.8. Model input parameters

Costs and resource use were estimated from a variety of published and unpublished sources including salary scales,(146) drug prices and other HSE reimbursement and payment information,(147-149) quotes and invoices for equipment and consumables,(150) supplemented by input from clinical and technical experts and HSE managers where necessary.(150-155) Given the rare occurrence of this particular type of care in the context of a complex funding system without a centralised cost register, parameter inputs and the underpinning model assumptions were heavily reliant on expert opinion.

Value added tax (VAT) was included in the BIA model (where appropriate depending on the item),(156) and discounting or depreciation were not applied, to reflect the actual expenditure by the HSE in each year reported.

As outlined in Chapter 3, the evidence regarding the clinical benefits and harms of domiciliary ventilation is relatively scarce and is based on heterogeneous populations, study designs and methodological approaches, as well as small sample sizes.(80, 106, 109, 113) Very limited data pertain specifically to patients with SCI availing of domiciliary invasive ventilation, given the rare occurrence of this condition and the ethical challenges of conducting comparative research in this population. Therefore, it is unclear whether domiciliary ventilation leads to better (or worse) outcomes for patients with SCI compared with hospital care. As discussed in Chapter 7, patients are discharged home, when clinically suitable and safe to do so, in respect of patients' preference to reside in their own home. For the purpose of this BIA, it was assumed that 'treatment effects' for these patients were similar between hospital and home care.

Assumptions underlying the input data are outlined in Appendix 4.

5.2.9. Cost inputs

In accordance with national HTA guidelines, all costs are presented in 2022 Irish Euro (€).(142) Costs included inpatient care, equipment and consumables, medication, labour, healthcare service use, and trainer and training costs.

For the comparator, the long-term inpatient care costs for treating patients with SCI requiring invasive ventilation in the NRH were estimated using four different approaches; this was conducted given that the NRH is not currently funded through the Activity Based Funding (ABF) model and there are no per patient cost estimates available.(157) For two of the approaches, acute care costs were used as a proxy for costs incurred by the NRH even though it is a post-acute rehabilitation hospital as opposed to an acute hospital. In the first method, a fixed intensive care unit (ICU) bed day cost from an acute hospital was obtained confidentially from the Healthcare Pricing Office (HPO) based on 2018 cost estimates (and inflated to 2022 prices).(158) In the second method, based on discussions with clinical coders in NRH,(154) it was assumed that the patient was coded under the relevant diagnosis related group (DRG) A06A ("TRACHEOSTMY/VENT>=96HRS, MAJC"). Hospital inpatient costs were calculated for this DRG code as per the methods outlined in the 2022 HPO Admitted Patient Price List.(149) In the third method, an average NRH bed day of approximately €1,400 was assumed.(159) Notably, no average bed day is available specifically for ventilated patients in the NRH. In the fourth method, it was assumed that 6.5 whole time equivalents (WTE) of nurses (comprising an average hourly cost of Enhanced Nurse (General) and Enhanced Nurse Senior (General) grades) were required to provide 24/7 care in a full year (accounting for annual leave, sick leave and training leave).(159) For all methods, it was assumed that the patient spent 1,826 days (five years) in hospital. The two extreme estimates provide the upper and lower bounds for the actual cost.

Equipment and consumable requirements and costs were obtained through discussion with clinical experts, HSE managers and HSE procurement who had oversight of national contracts, invoices and quotes and were able to provide estimates.(150, 152, 160, 161) Where specific data on costs were unavailable, estimates were provided by clinical experts. An annual maintenance cost of 10% of the original purchase price applying from year two was assumed.(162)

Medication and resource use estimates were informed by discussions with clinical experts.(151) It was assumed that the patient would be provided with a medical card and would be prescribed the following medication on a regular basis:

- macrogol 3350 13.8g sachet powder for oral solution
- hypertonic sodium chloride 7% solution for nebulisation
- salbutamol 2.5mg/2.5 ml nebuliser solution
- carbocisteine 250mg/5ml oral solution
- baclofen 5mg/5ml oral solution
- gabapentin 300mg capsules
- vitamin D supplementation (oral solution 25,000 international units (IU)) for eight months of the year only)
- paracetamol and codeine 500mg/30mg effervescent tablets
- sennoside B 7.5mg/5ml syrup
- bisacodyl 10mg suppositories

Dosage and frequency of administration were inferred by the summary of product characteristics (SPC) for each medication as per the Health Products Regulatory Authority (HPRA) medicines database and published literature.(163, 164) Liquid formulations or capsules/tablets that can be dispersed in water were selected in case of swallowing difficulties in the patient. It is acknowledged that these formulations generally incur a slightly higher cost. Of note, sennoside B and bisacodyl medicinal products are not reimbursed by the Primary Care Reimbursement Service (PCRS) and must be paid for out-of-pocket by the patient, and so were not included in the costs accrued to the HSE. It was assumed that all medicines would be collected from the patient’s community pharmacy under the general medical card scheme (GMS) on a monthly basis (with the exception of sennoside B and bisacodyl which would be paid for privately). The PCRS reimbursement prices for the included medicines were obtained from the PCRS list of reimbursable items.(147) The total costs of the medicines to the HSE were then estimated in accordance with the National Centre for Pharmacoeconomics (NCPE) Guidelines for Inclusion of Drug Costs in Pharmacoeconomic Evaluations.(165) Data from the PCRS were correct as of 11 March 2023.

National and international guidelines recommend a minimum of 196 hours of paid homecare for ventilator-dependent patients with complete C1-C3 SCI.(2, 133) However, local experts have confirmed that some patients in Ireland are currently receiving 336 hours of paid care per week, comprising 24/7 nursing and 24/7 HCA care.(166) In a scenario analysis, the cost of providing 336 hours of formal care instead of 196 hours as per the base case, is examined (see below).

In addition, as discussed in Chapter 4, there is disagreement among guidelines as to whether domiciliary ventilation care should be fully nurse-led care or whether HCAs can provide this care with nursing supervision. In the base case it was assumed that one nurse was providing care 24/7 and this was supplemented by 28 hours of HCA support per week.(143) In scenario analyses (see below) the ratio of nursing to HCA care was varied and the subsequent impact on the total budget impact examined. Nursing and HCA pay costs additionally included premium pay at 30% of total salary cost to factor in Sundays, public holidays and overnights,(159) and travel costs at 10% of total salary cost.(153)

In terms of primary care, current practice is that when a patient is referred to a health and social care professional, each episode of care generally lasts for a maximum of six sessions and then the patient is discharged from their books, though the patient can be re-referred back to the health and social care professional, if needed.(167) However the number of referrals will largely depend on the individual patient's needs as well as service availability locally in the context of long community waiting lists.(168) A range of assumptions with regard to the number of health and social care professional visits the patient would receive in their own home were applied; these were based on individual discussions with occupational therapy (OT), physiotherapy and clinical psychology EAG members (Appendix 4).(169, 170)

Based on discussions with clinical experts, it was assumed that the patient would visit their GP twice a year (as per normal GP practice for issuing six-monthly prescriptions) and attend two outpatient visits per year.(151) It was assumed that since the patient has a medical card and the GP is reimbursed on a capitation basis for all eligible patients, there would be no additional cost to the HSE for each visit by the patient.(148) The outpatient cost was inferred from the HSE’s cross border directive whereby the maximum one can currently be repaid by the HSE for an outpatient consultation abroad is €178, which implies that this is the cost in the Irish system.(171)

Based on discussions with Spinal Injuries Ireland, the importance of a driver was emphasised to enable the patient to engage in society to the fullest possible extent, though it is likely that some patients will be able to avail of supports from family and friends and so may not need a designated paid driver.(172) Hence, in the base case it was assumed that family or friends would be able to provide transport assistance to the patient, but in a scenario analysis one whole time equivalent HSE driver was costed using the HSE Consolidated Salary Scales.(146)

As highlighted in Chapter 4, and emphasised by various EAG members,(143, 144, 172) ongoing training and support in the community is considered crucial to providing continuity of care. A new training and support nurse in the community and associated training equipment and consumables were therefore costed, with cost assumptions inferred from another HSE national training programme(153) along with previously collected equipment and consumables costs.(150)
Based on EAG discussions, the time spent by NRH nurses and health and social care professionals providing training in advance of patient discharge, and also their time spent undertaking home visits, were also factored in (Appendix 4).(169, 170)

Table 5.3: Cost inputs†^

Parameter	Units per year	Mean unit cost (Deterministic value) ‡	Lower bound	Upper bound	Source
Homecare package					
Equipment and consumables					
Nebuliser and kit	1	€1,295	€1,036	€1,554	NRH(150)
Replacement consumables for nebuliser (from year 2)	0.5	€825	€660	€990	NRH(150)
Communication assistive technology	1	€8,362	€6,690	€10,034	NRH(150)
Cough assist device (rental)	12	€595	€476	€714	NRH(150)
2x Portable suction machines	2	€869	€695	€1,043	NRH(150)
Manual wheelchair	1	€7,071	€5,657	€8,485	NRH(150)
Electric wheelchair	1	€12,930	€10,344	€15,516	NRH(150)
Modifications for electric wheelchair	1	€10,791	€8,633	€12,949	NRH(150)
2x Ventilators plus consumables (rental)	12	€1,305	€1,044	€1,567	NRH(150)
Consumables to support discharge (3 month's supply)¤	4	€3,303	€2,643	€3,964	NRH(150)
Ongoing consumable supplies (from year 2)§§	1	€50,000	€40,000	€60,000	Expert Opinion(150)
PNS – fitting and conditioning	1	€58,000	€46,400	€69,600	NRH(150)
PNS - maintenance	1	€7,754	€6,203	€9,305	NRH(150)

Parameter	Units per year	Mean unit cost (Deterministic value) ‡	Lower bound	Upper bound	Source
Bariatric electric profiling bed with standard foam mattress (purchase)	1	€1,585	€1,286	€1,902	HSE(173)
Scan turn mattress	1	€6,500	€5,200	€7,800	NRH(150)
Hoist	1	€2,000	€1,600	€2,400	HSE(152)
Tracking hoist	1	€3,878	€3,102	€4,654	HSE(152)
Shower trolley	1	€4,640	€3,712	€5,568	HSE(152)
SpO_2 oxygen monitor (portable and 'vital signs')	1	€1,220	€976	€1,463	HSE(174)
Capnography monitor	1	€2,000	€1,600	€2,400	Clinical Engineer(160)
Physiotherapy tilt table	1	€3,000	€2,400	€3,600	Physiotherapist(161)
MotoMed	1	€4,500	€3,600	€5,400	Physiotherapist(161)
Labour					
Nurse (hourly)	8,803.5±	€46	€37	€55	HSE Salary Scales(146)
HCA (hourly)	1,461	€32	€25	€38	HSE Salary Scales(146)
Clinical psychologist (hourly)	7±	€67	€54	€81	HSE Salary Scales(146)
Occupational therapist (hourly)	13±	€51	€41	€62	HSE Salary Scales(146)
Physiotherapist (hourly)	11.5±	€51	€41	€62	HSE Salary Scales(146)
Medications					
macrogol 3350	12	€27	€21	€32	PCRS(147)
hypertonic sodium chloride	12	€42	€34	€51	PCRS(147)

Parameter	Units per year	Mean unit cost (Deterministic value) ‡	Lower bound	Upper bound	Source
salbutamol	12	€17	€14	€20	PCRS(147)
carbocisteine	12	€16	€12	€19	PCRS(147)
baclofen	12	€52	€42	€63	PCRS(147)
gabapentin	11	€34	€27	€40	PCRS(147)
vitamin D supplementation	8	€14	€11	€17	PCRS(147)
paracetamol and codeine	11	€25	€20	€31	PCRS(147)
Healthcare service use					
Outpatient visit	2	€178	€142	€214	HSE cross border directive(171)
Hospital care§					
Cost estimate using DRG code A06A (Year 1)	1	€476,730	€381,384	€572,0756	ABF 2022 Admitted Patient Price List(149)
Cost estimate using DRG code A06A (from Year 2)	1	€379,401	€303,521	€455,282	ABF 2022 Admitted Patient Price List(149)
National training and support					
Nurse (annual)	1	€72,217	€57,773	€86,660	HSE Salary Scales(146)
Laptop	1	€643	€514	€771	Online retailer(175)

Parameter	Units per year	Mean unit cost (Deterministic value) ‡	Lower bound	Upper bound	Source
Online module development	1	€35,000	€28,000	€42,000	HSE(152)
Online module content update (from Year 2)	1	€2,500	€2,000	€3,000	HSE(152)
Training manual printing	1	€447	€358	€537	HSE(152)
Nebuliser and kit	1	€1,295	€1,036	€1,554	NRH(150)
Cough assist device	1	€4,580	€3,664	€5,496	NRH(150)
Portable suction machine	1	€869	€695	€1,043	NRH(150)
Ventilator plus humidifier plus 2 breathing circuits	1	€515	€412	€618	NRH(150)
Consumables ¤	1	€3,304	€2,643	€3,964	NRH(150)

Key: ABF – activity based funding; HCA – healthcare assistant; HSE – Health Service Executive; NRH – National Rehabilitation Hospital; PCRS – Primary Care Reimbursement Service; PNS –phrenic nerve stimulator.
†Costs are presented in 2022 Irish Euro. Uncertainty in cost parameters is represented by 20% variation in the mean
‡All mean unit costs are exclusive of VAT and 10% maintenance cost (where applicable)
§Fixed ICU bed day mean unit cost not reported as confidential
^Salaries are based on mid-point of scale adjusted for pension, pay related social insurance (PRSI) and overheads (such as office space, heating and lighting) as per National HTA guidelines
*Excludes travel costs at 10% of total salary cost
Excludes premium pay at 30% of total salary cost for Sundays and public holidays **and travel costs at 10% of total salary cost
††Based on the mean salary scales for basic and senior staff grades
¤ Equivalent to a 3 month's supply includes personal protective equipment, and consumables for tracheostomy, suction, colostomy, urinary catheterisation and an Ambu-bag
§§ Ongoing consumables includes items for tracheostomy, ventilator, PNS, cough assist, suction, nebuliser, colostomy, urinary catheterisation and other items. Estimated cost only.
±Represents year 1 units. Hours decrease in subsequent years with additional hours required in year 1 to provide in-house training and in-home visits.

5.2.10. One-way sensitivity analysis

Given the lack of plausible estimates of uncertainty for the included cost parameters, uncertainty was represented by 20% variation in the mean value. The impact of extreme variation in single input parameters on the model output was presented on a tornado plot.

5.2.11. Scenario analyses

Scenario analyses were conducted to assess uncertainty in the model. In each scenario, model assumptions were changed and a base case parameter was replaced with an alternative estimate.

Members of the EAG confirmed that some patients in Ireland are currently receiving 336 hours of paid care per week, comprising 24/7 nursing and 24/7 HCA care.(166) In a scenario analysis, the cost of providing 336 hours of formal care instead of 196 hours as per the base case, is examined.

The EAG advised that one of the main contributors to the cost of these homecare packages is labour and that the proportion of care provided by nurses relative to HCAs influences these costs. In the base case it was assumed that a nurse provides 24/7 care to the patient, and this is supplemented by an additional 28 hours of HCA care. In scenario analyses the ratio of nursing to HCA care (out of a total possible 196 hours of care in a week) was altered from 6:1 in the basecase right through to 1:6 (Table 5.4). No less than 28 hours per week of nursing care was considered in any given scenario, as guidelines recommend that direct nurse supervision and involvement may be required for certain high risk tasks (for example, changing tracheal cannulas).(2, 138)

Table 5.4: Input parameters used in scenario analyses

Scenario	Nursing hours per week	HCA hours per week
Basecase (6:1)	168	28
Scenario 1 (5:1)	163.3	32.7
Scenario 2 (3:1)	147	49
Scenario 3 (2:1)	130.7	65.3
Scenario 4 (1:1)	98	98
Scenario 5 (1:2)	65.3	130.7
Scenario 6 (1:3)	49	147
Scenario 7 (1:5)	32.7	163.3
Scenario 8 (1:6)	28	168

Key: HCA – healthcare assistant

Even though the NRH is not currently funded under the ABF model, and it is not possible to determine per patient costs, in scenario analyses the hospital care costs for these patients were calculated as per the methods outlined in the 2022 HPO Admitted Patient Price List,(149) using the average NRH bed day cost, and assuming 6.5 WTE nursing per year.(159) The effect of using different methods for estimating the hospital care costs on the incremental budget impact was examined.

Given that different grades of nurses may be involved in the care of these patients, in another scenario analysis, a 50:50 mix of Enhanced Nurse and Enhanced Nurse Senior was assumed (as per practice in the NRH),(159) as opposed to a 50:50 mix of Staff Nurse and Clinical Nurse Specialists as per the base case.

There is uncertainty about how many of these patients might be discharged home each year. The basecase analysis used plausible estimates derived from historical patterns. However, as it is a rare outcome, a higher incidence may occur in one or several years. A worst case scenario was modelled, in which the total five-year cost to the HSE of providing this service to two new patients every year (and so a total of 10 new patients within five years) was estimated.

In a final scenario analysis, the financial impact of providing a designated driver (1 WTE) to the patient was examined. In a fifth scenario analysis, travel costs for homecare staff were excluded in the context of the patient's home being considered as the employee's 'normal place of work' for revenue purposes.(176)

5.2.12. Quality assurance

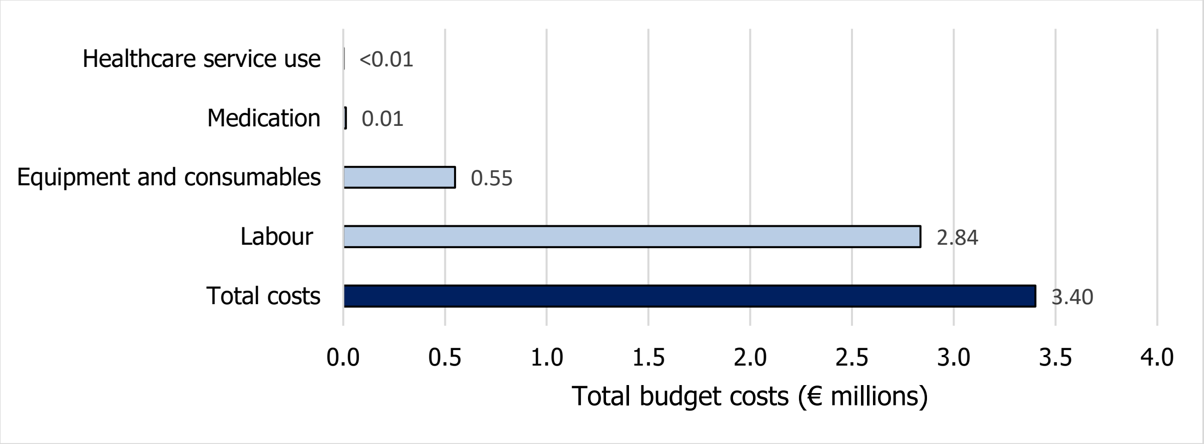
The BIA was developed in accordance with national HTA guidelines,(142) and quality assured in accordance with the HTA quality assurance framework. All model inputs and outputs were reviewed by a second member of the evaluation team. Input parameters and assumptions underpinning this BIA were reviewed and endorsed by the EAG.

5.3. Results

5.3.1. Total budget impact

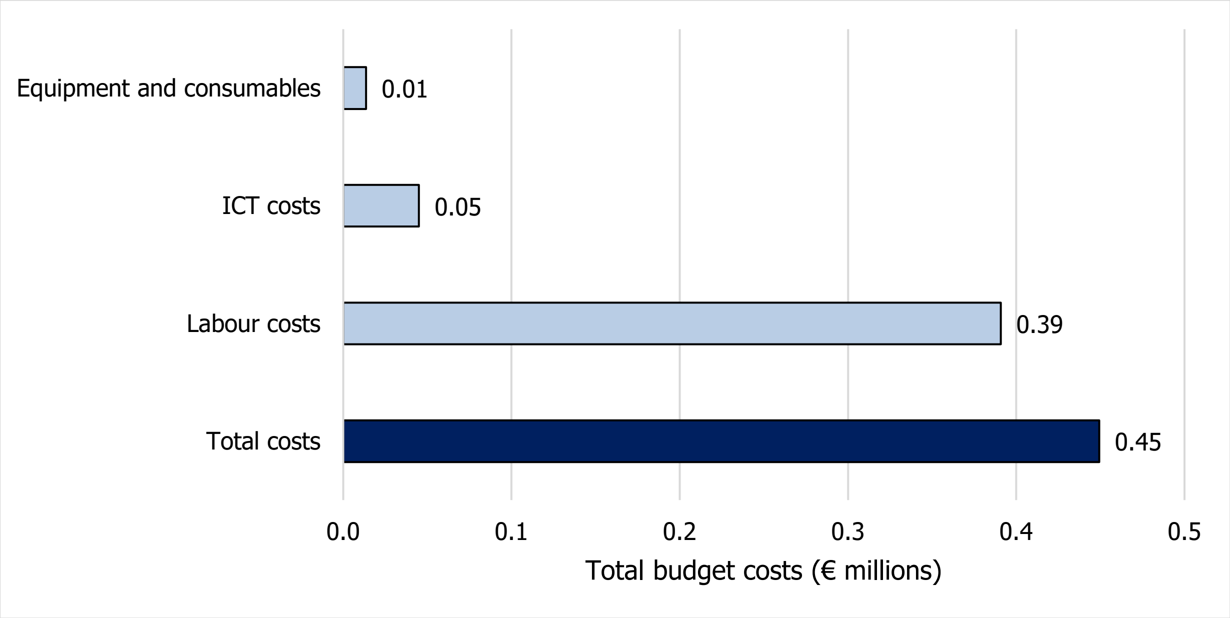
Over a five-year time horizon, the total budget impact of a comprehensive homecare package for a patient with SCI requiring mechanical invasive ventilation was estimated at €3.4 million per patient. The majority of expenditure over a five-year time horizon was associated with the cost of labour (€2.8 million, 83%) (Figure 5.1).

Figure 5.1: Itemised five-year total budget impact of a comprehensive homecare package for a patient with spinal cord injuries requiring mechanical invasive ventilation



The total budget impact for establishing and implementing a new bespoke role in the community that would provide ongoing training and support to patients, families and homecare workers, was estimated at €0.45 million over a five-year period (Figure 5.2). The majority of expenditure for this training and support role was also associated with the cost of labour (€0.39 million, 87%).

Figure 5.2: Itemised five-year total budget impact of a national training and support role

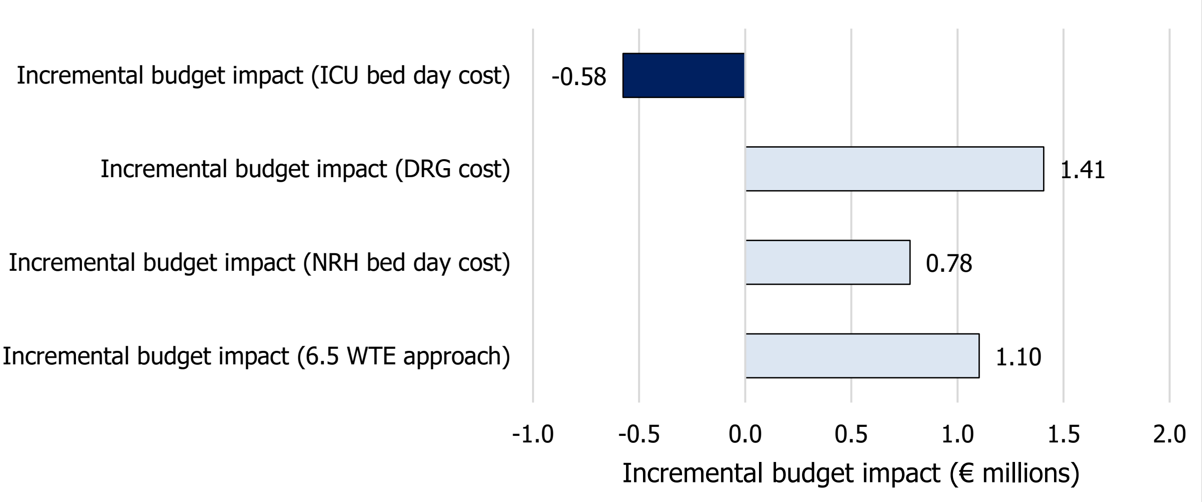


Key: ICT – information and communications technology

5.3.2. Incremental budget impact

There was substantial uncertainty regarding the five-year incremental budget impact of providing a comprehensive homecare package, compared with hospital care, for each patient, with estimates ranging from €0.58 million cheaper in the base case to €1.41 million more costly one scenario analysis (Figure 5.3). Of note, three of the four methods resulted in a net budget impact. However, given the notable disparity in cost estimates, driven by differences in costing methodologies, particular caution is advised in interpretation of the incremental budget impact. It is unclear whether homecare would cost more or less than hospital care for these patients.

Figure 5.3: Incremental five year budget impact analysis



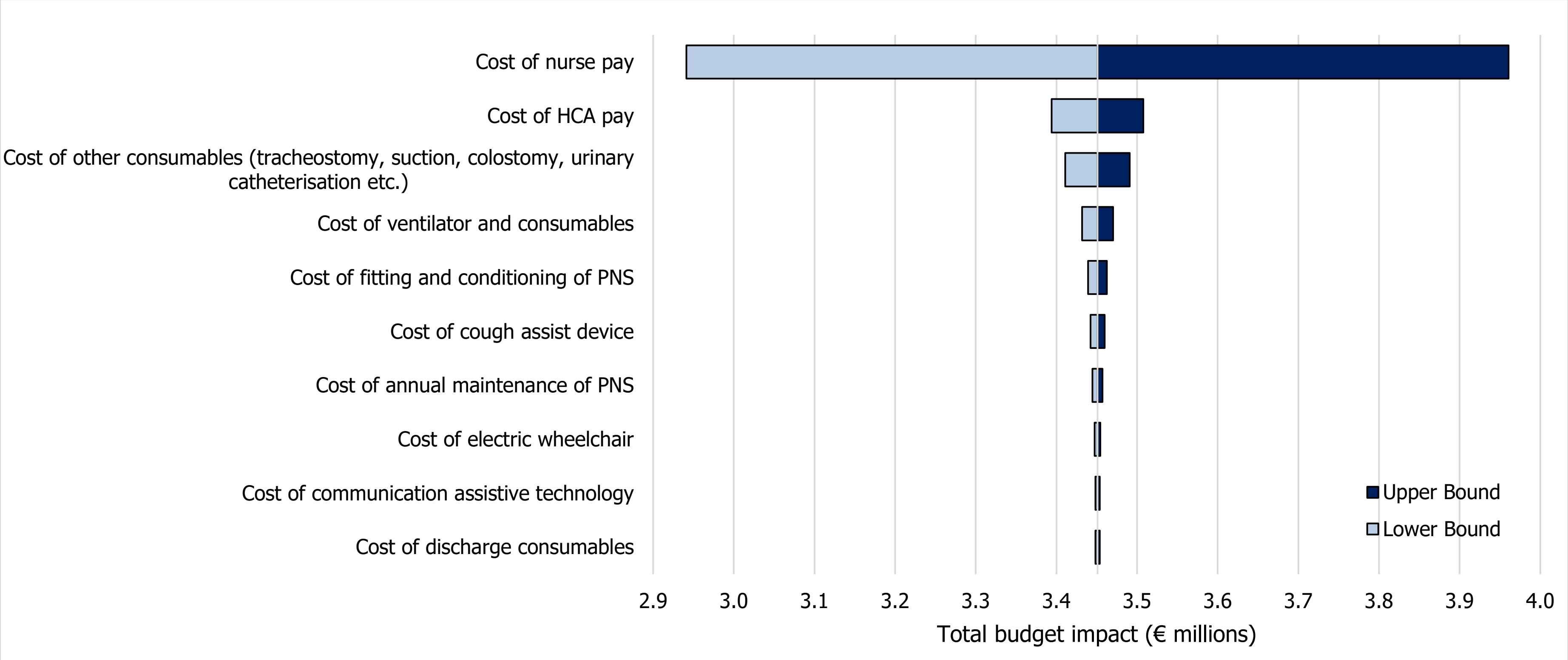
Key: DRG – diagnosis related group; ICU – intensive care unit; NRH – National Rehabilitation Hospital; WTE – whole time equivalent.

5.3.3. One-way sensitivity analysis

In the one-way sensitivity analysis (OWSA), input parameters were varied and ranked in order of increasing influence on the total budget impact. The result is presented as a tornado plot which provides a visual representation of the sensitivity of the model to the uncertainty associated with individual parameters (Figure 5.4). Although all parameters were varied in the analysis, only the ten most influential parameters are presented.

Uncertainty relating to nursing costs was found to contribute most to uncertainty in the total budget impact analysis (lower bound: €2.9 million, upper bound: €3.9 million). Uncertainty in this parameter was associated with 30% variation in the total budget impact over a five-year time horizon. Other influential parameters included the cost of HCA pay, and the cost of consumables relating to the ventilator as well as those needed for other aspects of care (for example, tracheostomy, suction, colostomy, urinary catheterisation etc.)

Figure 5.4: Tornado plot of one-way sensitivity analysis for the five-year total budget impact analysis of comprehensive homecare package



Key: HCA – healthcare assistant; PNS – phrenic nerve stimulator

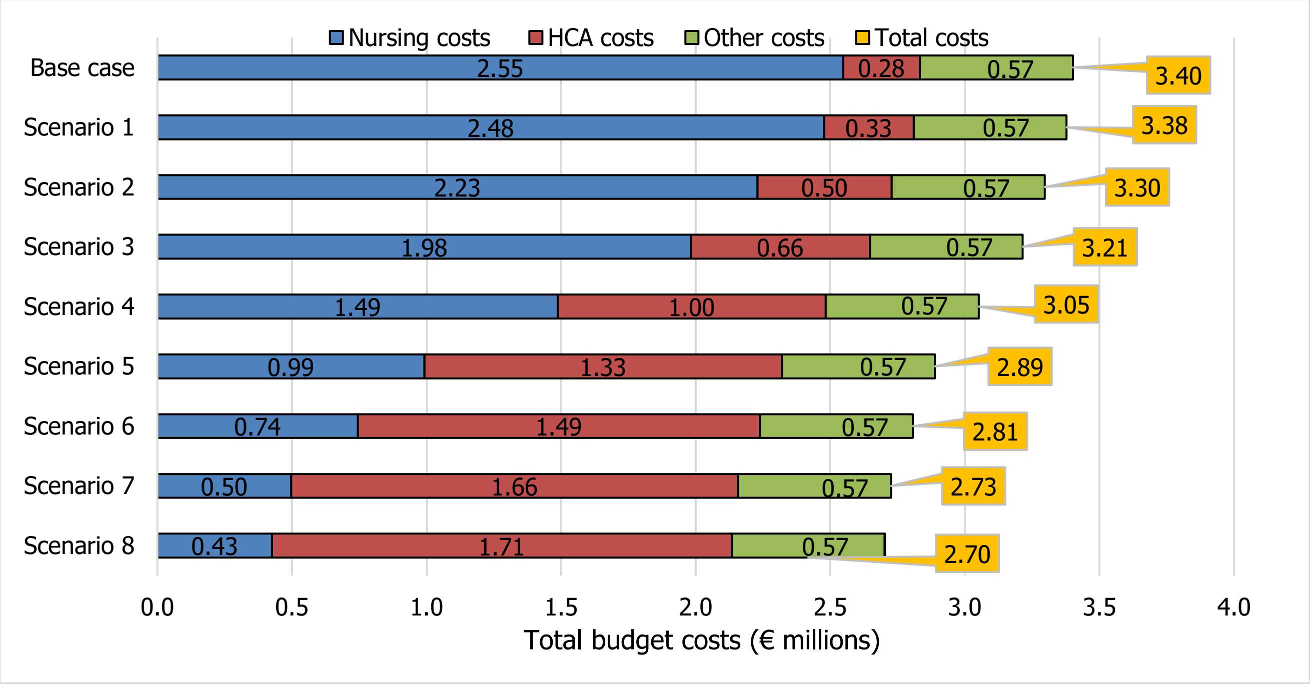
Note: Uncertainty represented by 20% variation in the mean value

5.3.4. Scenario analyses

As some patients currently avail of 336 hours of paid formal care per week (that is, one nurse and one HCA each providing 24/7 care), a scenario analysis was undertaken assuming this intensive level of care provision. Over a five-year time horizon, the total budget impact of this intensive level of care provision was estimated at €4.8 million per patient, which is 42% higher than the base case.

Given the substantial contribution of labour to the total costs of a homecare package, scenario analysis was undertaken to examine the impact on the five-year total budget impact of altering the ratio of nursing to HCA care. Figure 5.5 illustrates that, as the ratio of nursing care to HCA decreases, the total budget impact also decreases. Relative to the basecase, in scenario 8 where it was assumed that there was 28 hours of nursing care and 168 hours of HCA care per week, the total budget impact was reduced by €0.7 million (21%) over five years.

Figure 5.5: Results of scenario analysis examining the impact of alternative nurse to healthcare assistant (HCA) staffing ratios on the five-year total budget impact



Note: **Hours of care per week per profession.** *Basecase* -168 hours nursing, 28 hours HCA (6:1); *Scenario 1* – 163.3 hours nursing, 32.7 hours HCA (5:1); *Scenario 2* - 147 hours nursing, 49 hours HCA (3:1); *Scenario 3* – 130.7 hours nursing, 65.3 hours HCA (2:1); *Scenario 4* - 98 hours nursing, 98 hours HCA (1:1); *Scenario 5* – 65.3 hours nursing, 130.7 hours HCA (1:2); *Scenario 6* - 49 hours nursing, 147 hours HCA (1:3); *Scenario 7* – 32.7 hours nursing, 163.3 hours HCA (1:5); *Scenario 8* - 28 hours nursing, 168 hours HCA (1:6).

In another scenario analysis, the financial impact of alternative nursing grades was examined. The overall impact on the five-year total budget impact was minimal, decreasing from €3.40 million in the base case to €3.35 million in the scenario analysis. This indicates that the base case assumption of a 50:50 mix of staff nurse and clinical nurse specialist grades results in broadly similar cost estimates as a 50:50 mix of enhanced nurse and enhanced nurse senior grades.

In the final scenario analysis, the financial impact of providing a designated driver (1 WTE) to the patient was examined. The recruitment of a driver increased the five-year total budget impact by €252,503 (6.9%) relative to the base case. In the fifth scenario analysis, travel costs for homecare staff were excluded. This reduced the five-year budget impact by €184,845 (5.4%) relative to the base case.

5.3.5. Overall cost to the HSE

If there is an expectation that one new SCI patient will require domiciliary invasive ventilation every second year (that is a second and third patient availing of the service, on years three and five respectively), and accounting for the bespoke training and support role, then the resulting five-year total budget impact for the HSE overall is estimated to be between €6.7 million and €9.2 million, with the higher estimate reflecting the cost for three consecutive patients who each require 336 hours of paid care per week. Based on an assumption of two new patients per annum, effectively a worst case scenario, the total cost to the HSE overall is estimated to be between €21.2 million and €29.8 million over five years.

5.3.6. Other costs that may be relevant for the HSE

This BIA focused on the costs associated with a ‘typical’ homecare package for patients with SCI availing of invasive mechanical ventilation, and also a bespoke national training and support role. There may be other costs that are potentially relevant for the HSE depending of the nature of the complexity of care required for the patient, and also depending on the clinical governance structure that is ultimately implemented by the National Clinical Programme for Rehabilitation Medicine and others. With regards to the patient, other possible costs for the HSE that were not included in this BIA may include:

- Administration costs associated with private homecare providers
- Emergency department visits and hospital admissions
- Ambulance transfers involving the Mobile Intensive Care Ambulance Service
- Community Intervention Team (CIT) visits and associated treatment

- Admission to residential care settings
- Use of respite services
- Enteral/parenteral feeding and/or administration of medicines
- Additional prescriptions
- Involvement of other health and social care professionals (for example, speech and language, dietetics etc.).

In relation to changes to the governance structure, there may be expenditure in terms of recruitment of administrative staff and purchase of ICT. There would also be considerable opportunity cost associated with the involvement of existing consultants and other clinicians in setting up and implementing a new governance system.

5.3.7. Costs falling on patients and families

While the perspective of this BIA is that of the HSE, there are considerable and ongoing out-of-pocket expenses for these patients and their families associated with the provision of complex healthcare in a home setting. The following substantial costs have been reported by Irish patients with SCI availing of domiciliary ventilation services and their families:(145)

- Medical
    - Physiotherapy – often paid for privately
    - D-mannose supplement for prevention of urinary tract infections
    - Items not covered on medical cards (for example, certain skin creams)
    - Counselling costs
- Housing
    - Electricity costs
    - Housing adaptations
    - Increased heating costs, ensuring availability of hot water (noted as being particularly important as patients with SCI are more sensitive to cold conditions)
    - Cleaners and household help beyond what is funded by the HSE
- Transport
    - Provision of an accessible car

  - Taxis
  - Parking charges in hospital carparks for family members
- Family costs
  - Reduced income as family members reduce employment or retire to care for patients.

The patient representative noted that one major area of concern for patients with SCI and families is that medical cards are not guaranteed. Although most patients receive an emergency card as an inpatient, this is then means-tested post discharge. It was reported that this uncertainty causes huge stress for families and also acts as a barrier for the patient to return to work due to fear of losing their medical card. It was considered that this factor may contribute to patients with SCI being unemployed.(145) If a patient doesn't qualify for a medical card, there would be substantial out of pocket expenditure involved in providing their care. Of note, obtaining a power chair and environmental control units/communication assistive technology, would be particularly expensive.(177)

5.3.8. Costs falling on other government departments

While the perspective of this BIA is that of the HSE, it is important to acknowledge the substantive costs that fall on other government departments with regard to the delivery of a comprehensive homecare package for patients with SCI requiring mechanical invasive ventilation. The relevant grants, allowance and tax relief schemes are as follows (Appendix 5):

- Housing adaptation grant for people with a disability(178)
- Drivers and passengers with disabilities: tax relief scheme(179)
- Disability Allowance(180)
- Carer's Allowance.(181)

5.3.9. Protocol deviation

In the protocol for this HTA, it was planned that the cost of domiciliary invasive ventilation would be compared with the cost of long-term invasive ventilation in both hospital and residential care settings.(13) However, given that no residential units in Ireland currently accept ventilated SCI patients and hence there is a lack of available data,(182) the evaluation team was unable to provide cost estimates for the provision of care in residential care settings. It was the expert opinion of the EAG that it is more challenging to find a suitable residential care placement than it is to send the patient home, given that the care requirements far exceed what is usual in the vast majority of residential care settings, and that this is an important gap in service. While it is difficult to assess how costs may differ between a specialised residential setting and a home environment, certain equipment such as beds, hoists, ventilators, cough assist, oxygen monitor, nebulisers, suction etc. may already be available in the residential care setting and so there may be a reduction in terms of initial set up costs. However the need for 24/7 specialised one-to-one nursing/HCA care remains regardless of the setting which incurs significant costs.

5.4. Discussion

This BIA investigated the total costs of providing a comprehensive homecare package to a patient with SCI who requires invasive mechanical ventilation, as well as the total costs of developing a bespoke trainer and support role on a national footing. Over a five-year time horizon, the total budget impact of a comprehensive homecare package was estimated at €3.4 million per patient. Given differences in how acute care can be costed, it is unclear whether homecare would cost more or less than hospital care for these patients. However, the decision on whether to discharge home such a patient should be based on patient, social and ethical factors rather than cost saving ones. Regardless of whether a discharge home represents an actual cost saving or not to the HSE, freeing up of a scarce ICU or high dependency unit (HDU) bed in a hospital will likely increase efficiencies in the health service. Almost all of the expenditure over the five-year period was associated with the cost of labour (€2.8 million, 83%), and equipment and consumables (€0.5 million, 16%). Medications and healthcare service use contributed relatively little to the total budget given that the patient cohort under consideration that are suitable for domiciliary invasive ventilation (typically young males) are largely healthy otherwise. While the budget impact may be quite large at the individual patient level, the impact at a population level would be small given that a maximum of one or two new patients are expected to require this service every year. Assuming that one new patient will require this service every second year, and accounting for the bespoke training and support role, then the resulting five-year total budget impact for the HSE overall is estimated to be €6.7 million in the base case.

International guidance generally recommends a *minimum* of 196 hours of formal care per week(133) and this estimate of 196 hours was assumed in the base case. However, care needs vary from patient to patient and there are reports of ventilator-dependent SCI patients in Ireland with substantially higher care needs availing of 336 hours of formal care per week,(166) resulting in a higher total budget impact (estimated at €4.8 million over five years). It is important that care packages are developed based on an individual's needs, taking into consideration human resource and practical issues (for example staff breaktimes etc.), and that 196 hours of formal care per week should be viewed as a *minimum* rather than a target or a *maximum*. Continuity of care 24/7 is essential for these patients as they must be under the direct supervision of a competent person at all times, and this must factor into staff scheduling.

In the OWSA, uncertainty in nursing costs was found to be a key driver of uncertainty in the total budget impact analysis (lower bound: €2.9 million, upper bound: €3.9 million). In scenario analysis, reducing the amount of nursing care to a minimum of 28 hours per week, and increasing HCA care to a maximum of 168 hours per week, reduced the five-year total budget impact by €0.7 million (21%). While international guidance varies on this matter (Chapter 4), some countries do support a system of HCA-delivered homecare to invasively ventilated patients with nursing supervision. For example, in New Zealand the relevant guidance acknowledges that, where possible for a medically unstable individual, a suitably qualified nurse should be providing direct patient care, but otherwise care may be provided "*by people who have successfully completed competency-based training.*"(126) Greater delegation of roles to competent HCAs may also enable greater access to these services in the community given the current recruitment and retention issue in the homecare sector.(183) However, it is critical that any potential savings and increased access to homecare services do not compromise patient safety. As outlined in Chapter 4, it is crucial that any homecare staff involved in the care of these patients are trained and competent to provide the complex care required and know how to deal with urgent and emergency situations as they arise. There needs to be a clear clinical pathway for these patients in the community underpinned by a strong governance framework. Consultation with the Nursing and Midwifery Board of Ireland (NMBI) may be required to discuss the implications for their scope of practice of delegating high risk tasks to unregulated professionals (See Section 7.3.3.2).(184)

The need for greater levels of ongoing training and support in the community throughout the patient's lifetime, as advocated by patient representatives and clinicians alike,(144, 172) supports the potential need for a bespoke role in the community. The total budget impact for establishing and implementing a new bespoke role in the community that would provide ongoing training and support to patients, families and homecare workers, was estimated at €0.45 million over a five-year period. The majority of expenditure for this training and support role was also associated with the cost of labour (€0.39 million, 87%). While the cost inputs for this role are largely indicative they give an idea of the potential budget impact of such a role, which could potentially be integrated across other National Clinical Programmes (for example, Respiratory and Neurology). Given the rare occurrence of domiciliary invasive ventilation nationally and internationally,(97) a role that provides training and support to all care providers of these patients regardless of underlying diagnosis may help to improve standards and potentially ensure high quality care for these patients.

In relation to keeping these patients with very complex health and social care needs in their homes there are other substantial costs that fall on patients and families, as well as on other government departments. There are large upfront costs such as housing and car adaptations that, while heavily subsidised by the State, still incur substantial out-of-pocket expenses for patients and families. Energy bills are also a cause for worry for many patients and families given the reliance on many equipment on electricity, particularly in light of the current 'cost of living crisis'.(185) There are also concerns that the current maximum grant available of €30,000 for people with a disability to make the required housing adaptions is insufficient given the substantial and increasing costs associated with building.(186) A review of the full suite of Housing Adaptation Grants for Older People and People with a Disability is currently underway by the Department of Housing, Local Government and Heritage.(187)

There is limited economic evidence specifically relating to the cost of providing domiciliary invasive ventilation for patients with SCI, however literature describing the consequences of SCI more generally highlights the associated economic burden. A study conducted in 1996 by Botel et al. estimated the costs of homecare for ventilator-dependent patients with SCI. Adjusting to 2022 Irish Euro Prices, the annual cost of home care per ventilator-dependent individual with SCI was estimated to be €586,707 increasing to €778,030 once all equipment costs were accounted for.(108) A 2006 study by Priebe et al. reported an average annual cost of €943,282 associated with the management of a person with high cervical SCI (C1-C4) in their first year and €168,965 in each subsequent year, with an estimated lifetime cost of €3.7 million for a 25 year old, adjusting to 2022 Irish Euro Prices.(188) Importantly the cost estimates by Priebe et al. do not specifically pertain to a ventilator-dependent patient. Despite considerable differences in the costing methods used, and the contexts in which the studies were undertaken, the total budget impact estimated here in the first year of care (€752,697) is broadly comparable to that reported by both Priebe et al.(188) and Botel et al.(108)

5.4.1. Strengths and limitations

A key strength of this BIA was the involvement of a wide range of stakeholders in the acquisition and contextualisation of cost and resource use data. The scoping review in Chapter 4 provided the evaluation team with additional awareness of international practice and standards in this area, thus informing relevant scenario analyses.

A limitation of this BIA was the high degree of uncertainty in relation to the estimated hospital care costs. Four different methods were used that provided vastly different estimates, making it difficult to determine whether homecare is more or less costly than hospital care. It is also unclear whether the home and hospital care costs are directly comparable given that they include different cost considerations. For example the ABF Price List approach estimates a price for the entire episode of care from admission right through to discharge from the hospital linked to a particular diagnosis related group (DRG). It does not provide pricing information for individual diagnoses, interventions or procedures.(158) This differs from the approach undertaken for costing the homecare package which assumed more of a micro-costing approach. Therefore caution is urged in the interpretation of the estimated incremental budget impact.

Another limitation of this BIA was the inability to determine the cost of providing long-term care for these patients in residential care. As discussed in Chapter 6, the funding system for these patients who require discharge packages is complex and involves multiple budget holders. 'Top-up' funding from Disability services would be required to supplement the 'Fair Deal Scheme' in order to provide adequate funding to provide care and supports for these patients in residential care settings.(189) However, the evaluation team was unable to ascertain the amount of additional funding available for these particular patients given that no residential unit nationally currently accepts these patients.(182)

5.4.2. Conclusion

Providing comprehensive homecare to patients with SCI who require mechanical invasive ventilation is highly costly, although limited to a very small cohort of patients. It should be noted, however, that the cost of homecare is likely to be broadly similar to hospital-based care for these patients. Over a five-year time horizon, this BIA estimated that the total budget impact of a comprehensive homecare package for an individual patient would cost €3.4 million, with 83% of costs associated with nursing and HCA care. While the provision of such a homecare package may be costly at an individual level, it would have a relatively low impact at a population level given the rare occurrence of these events. The total cost for the HSE overall is estimated to be between €6.7 million and €9.2 million over five years assuming one new patient every second year. The total budget impact for establishing and implementing a new bespoke role in the community that would provide ongoing training and support to patients, families and homecare workers, was estimated at €0.45 million over a five-year period. Such a role may help to improve standards and potentially ensure high quality care for patients availing of domiciliary invasive ventilation.

6. Organisational considerations

Key points

- An assessment was undertaken of the necessary organisational changes associated with the provision of domiciliary ventilation to patients with SCI on a national footing.
- While it is already standard practice to discharge these patients home as soon as it is safe to do so, there are substantial organisational barriers to the provision of homecare services for these patients.
    - Currently the funding of homecare packages for these patients with substantial care needs is a challenge as these are funded from various different sources including local budgets; the funding of high dependency patients may overwhelm local budgets in particular.
    - Other barriers include the limited availability of highly trained staff in the community, the lack of a statutory homecare scheme, the lack of an explicit care pathway in the community and the significant out-of-pocket costs for patients and families.
- Appropriately adapted housing is an essential prerequisite for the delivery of homecare services for these patients. However, there are limits to the funding available for housing modifications, and this funding is means tested. In addition, some patients do not own their own house and so provision of alternative accommodation by local authorities may be necessary. A review of the full suite of Housing Adaptation Grants for Older People and People with a Disability is currently underway by the Department of Housing, Local Government and Heritage.
- The organisational considerations for this HTA are discussed at three different levels of the healthcare system (micro-, meso- and macro-level). The micro-level factors relate primarily to patient and family interactions; the meso-level relate to healthcare organisation and the community; and macro-level relate to health policy.
- Micro-level organisational factors for consideration include:

  - development of explicit discharge criteria and protocols to assist with the complex discharge process and so that patients and families can have realistic expectations for the future
  - a more flexible process for obtaining housing adaptation funding that can enable families to carry out necessary renovations, provision of housing for those without their own home, and greater access to step down units to facilitate the transition home and respite services to provide relief for carers and families
  - exploring the barriers that currently exist to providing domiciliary ventilation in long term care facilities and how these might be addressed
  - provision of recommended supports and services to enable the patient to engage in their communities to the fullest possible extent
  - greater access to ongoing face-to-face supports for patients and families in the community.
- Meso-level organisational factors for consideration include:
  - development of a centralised national systematic, dedicated and integrated budget with an associated national tender framework to cover personal care, nursing care and equipment; this would likely alleviate the administrative burden and inequity of the system, and expedite the approval of a fully funded homecare package for these patients
  - development of a model of complex discharge planning similar to that used for paediatric services (as recommended by the HSE Integrated Care Pathway for the Management of SCI)
  - development of an explicit care pathway within the community for these patients, with defined involvement and clear roles and responsibilities for all involved parties, including processes for the transfer of ventilator-dependent children with SCI into adult services.
  - care coordination, in the context of healthcare staffing issues, to ensure that informal and formal supports, from all required disciplines, work effectively together to ensure the best outcomes for the patient with SCI
  - the model of homecare provision, and, specifically, whether healthcare assistants providing care with nursing oversight is feasible and appropriate, and how competency can be assured in those

  providing care; substituting 24/7 nursing care with 24/7 trained HCA care with appropriate nursing oversight would reduce the overall cost of a care package for these patients and would likely increase the availability of staff

  - development of a bespoke training and support role in the community to provide quality assurance of domiciliary invasive ventilation services
  - involving other National Clinical Programmes such as Respiratory Medicine in further developing the care pathway into the community
  - involving clinical engineers with respect to the maintenance of equipment for these patients in the community, and having a clear understanding of the governance of these equipment.

- In terms of the macro-level factors, there is currently a policy initiative for the safe discharge home of these patients with an appropriately funded package of care in the context of an impending statutory homecare scheme.
- A bespoke national training and support nurse was costed in Chapter 5. It is possible that such a role could be developed to provide ongoing support and information to patients and families, and provide training to homecare staff, ensuring that they are competent to provide the level of care required for these patients.
- Should a decision be made by the HSE to implement a national, systematic, dedicated and integrated budget with an associated national tender framework for the provision of homecare to invasively ventilated patients with SCI, this will require substantial organisational cooperation across multiple agencies and programmes. Consideration should be given to organisational factors at the micro-, meso- and macro- levels when coming to a decision.

6.1. Introduction

This Chapter provides an assessment of necessary organisational changes that would be associated with the provision of domiciliary ventilation to patients with SCI on a national footing. This Chapter has been guided by the considerations outlined in the European network of HTA (EUnetHTA) Core Model organisational aspects domain.(190) The organisational considerations for this HTA are discussed at three different levels of the healthcare system (micro-, meso- and macro-level). The micro-level factors relate primarily to patient and family interactions; the meso-level relate to healthcare organisation and the community; and macro-level relate to health policy.(190) This Chapter describes the high-level organisational considerations associated with the national provision of care to these patients; however, the specific details regarding service provision may need to be outlined in a separate implementation plan to be developed by the HSE.

6.2. Micro-level factors

The focus of micro-level organisational factors is on the patient flow through the system. An integrated care pathway for patients with SCI has been developed in Ireland by the HSE's National Clinical Programme for Rehabilitation Medicine, in line with its model of care.(2, 7) This pathway has been described in Chapter 3 but in brief, patients with high cervical SCI who require permanent mechanical invasive ventilation, whom are deemed clinically suitable for home discharge, remain in the National Rehabilitation Hospital (NRH) or another hospital, until the care package and required resources have been secured and the home environment has been appropriately modified.(2) Prior to 2019, ventilator-dependent SCI patients in Ireland were transferred to hospitals in the UK to receive inpatient rehabilitation care under the HSE's Treatment Abroad Scheme (TAS) as the NRH did not have the specialist staff, equipment or pathway in place, to provide inpatient care for ventilated patients.(2) However, since 2019, the NRH commenced admission of ventilator-dependent patients with SCI and so there is no longer a requirement for these patients to receive care abroad.(191)

6.2.1. Discharge to home

As noted within the HSE Integrated Care Pathway document, a safe return to home with an appropriately funded care package would be considered the ideal for many patients with this level of high dependency needs.(2) However, there are many barriers to organising home care for these patients such as the challenges of navigating multiple local budgets, the limited availability of highly trained staff in the community (See Section 6.3), and the significant cost for families to make the necessary housing and transport modifications. Given these challenges, many patients remain in the NRH for a prolonged period as a delayed discharge while awaiting a homecare package to be secured. Sometimes patients may be repatriated to an acute hospital closer to their home during this wait period.(192) Some ventilator-dependent SCI patients in Ireland have spent several years in hospital awaiting to be discharged home.(2) Patients and families have reported that these delays have been very traumatic and have had a hugely negative impact on them.(172) For many of these patients and family members, the excessive length of time spent in hospital awaiting a homecare package was the overriding memory of the entire care pathway.(172) However, it has been the experience of disability case managers in the community that access to a step-down facility, with the required trained staff and equipment, close to the patient's home, provides a good interim solution if substantial delays to suitably adapting the home and organising the care package are anticipated. It also allows time for the staff to get used to the care plan, and for issues to be troubleshooted prior to moving home permanently. However, it is acknowledged that there is currently inequity in the provision of these step-down services for individuals with severe disabilities across the country.(193)

The NRH is responsible for the discharge of these patients. There are currently no explicit discharge criteria specified for these patients in the NRH; patients are assessed on a case-by-case basis reflecting the complexity and uniqueness of each patient's circumstances.(189) However, there are certain implicit criteria that need to be met at a minimum before these patients can even be considered for discharge. Importantly, there must be a willingness on behalf of the patient and their family for a return home. These aspects include whether:(189)

- the patient is sufficiently medically stable
- the home is suitable
- the patient will have sufficient family support at home
- there is sufficient funding in place.

These implicit criteria match broadly to those recommended by the Canadian Thoracic Society.(134)

Patients should not be discharged until such a time as the hospital is confident that the ventilation and care can be continued safely, effectively and comfortably at home. There are inevitably limits to the supports that can be provided and so there may be patients who may never be suitable for a home discharge given their social or medical situation.(189) It is important that appropriate arrangements are put in place for these patients, being mindful of patient's preferences; such arrangements may include transfer to a residential care facility. As outlined in Chapter 4, the Dutch guidance document recommends that on the day of discharge a liaison nurse visits the patient in their home to check whether the ventilator has been set up properly, to answer any further questions and if necessary, and to provide additional training to informal and or professional care givers.(138) Given the complexity of the discharge process for these patients, some guidance documents recommend that the discharging hospital should have an explicit checklist to ensure that all required equipment, training, funding and supports have been obtained prior to the individual being discharged.(79, 88, 129, 136, 139) The German National Guideline specifies that provision of such a checklist to the patient upon discharge from hospital should be considered a quality indicator for that service.(79, 139) Clear criteria for the safe discharge of these patients is needed, given the risks associated with the complex care of these patients at home. A quality indicator within the German National Guideline is that domiciliary invasive ventilation is only established after consent is given by the extensively informed patient and/or their legal guardian.(79, 139) A centralised approach to budgeting and planning (as discussed in Section 6.3.2) may enable a smoother discharge for patients.

In Ireland, before being permanently discharged from the NRH, these patients undergo a trial discharge period whereby they spend a night or two at home before returning to the NRH to troubleshoot any issues they might experience.(143) This approach is recommended by organisations such as the Canadian Thoracic Society, who advise that a night or two at home enables the patient, the family and health providers to understand gaps in the preparation for discharge to the community.(134)

6.2.2. Grants to enable discharge to home

Patients and families have expressed their frustrations when it comes to obtaining grants from local authorities in order to undertake the required housing renovations.(172) It was their experience that the local Council would not approve the grant until the HSE-funded care package is in place; however, often these care packages would not be put in place until the home is fully prepared.(172) Consideration should also be given to provision of the grant even if the patient may never be likely to be fit for permanent discharge, so that their home could be made accessible to enable the patient to spend some time at home.(194)

There are also limits on the funding available for housing modifications, and these are means tested. The Housing Adaptation Grant for People with a Disability scheme provides grant aid *"to assist in the carrying out of works that are reasonably necessary for the purposes of rendering a house more suitable for the accommodation needs of a person with a disability."* The effective maximum grant under the scheme is €30,000, which may cover up to 95% of the approved cost of works.(195) To make the modifications required to accommodate the patient, their bed and other equipment, and also the homecare staff (including a break room), €30,000 is not considered to be sufficient and there is generally a reliance on local communities for fundraising.(172, 189) (194) As discussed in Chapter 5, a review of the full suite of Housing Adaptation Grants is currently underway.(187)

6.2.3. Care needs and supports

SCI patients with C1-C3 injuries, who are dependent on permanent mechanical invasive ventilation require specific additional supports in terms of specialist staff and equipment. These patients are considered 'level 3' (on a three point scale of increasing requirements for assistance) in terms of their care needs, given that they are unable to breathe independently, require permanent mechanical ventilation, and generally require care continuously over a 24 hour period, seven days a week.(2) The third edition of attendant care guidance from New South Wales, Australia(133) (note that the Irish Attendance Care Guidelines developed at the NRH and included in the HSE Integrated Care Pathway document is based upon the second edition of this guidance),(2, 196) recommends that for patients with complete C1-C3 injuries, a minimum of 196 hours of personal care per week (28 hours per day) is required, dependent on personal circumstances. Specifically, the 2017 'icare' guidance states that full support (that is, the patient can only complete ≤25% of the task on their own) for the following activities would be included within the indicative 196 hours per week(133):

- transfers and bed mobility
- moving around the community via modified vehicle
- showering
- grooming
- eating and drinking
- toileting
- dressing
- overnight care
- health needs (for example, medical appointments, exercise, medication management, catheter changes etc.)
- meal preparation
- housework
- shopping
- home and garden maintenance.

Participation in education, employment, social and leisure activities is crucial for patients.(172) It is important to consider that public transport may not suitably meet the needs of many people with severe disabilities, in particular those with SCI who are ventilator dependent.(193) A designated driver (usually a family member, friend or volunteer) who is trained to drive a specially adapted vehicle is generally required to transport the patient around the community.(133, 193) There are important additional considerations with regards to the transport of these patients in adapted vehicles(160):

- There are numerous requirements that must be followed when transporting medical gases, including the correct storage and securing of the oxygen cylinders in the vehicle, and the notification of the insurer that the vehicle is carrying oxygen.

- Given that oxygen aids in combustion there are particular safety hazards associated with its transport. Therefore, smoking (including e-cigarettes) must be avoided throughout the entire journey, and the oxygen should never be used while the vehicle is being refuelled.(197)

- The cylinder size and the quantity of oxygen required for the journey (taking into account possible delays), particularly if the patient is heavily dependent on oxygen and on high oxygen concentrations and or high flows.

- Back-up cylinders may be required in the vehicle which need to be secured safely and should either have built in regulators (CD or XZ cylinders for example) or have regulators attached so the cylinder is ready for use.

- There needs to be consideration into the type of wheelchair the patient has - can it safely hold the weight of the ventilator and can an oxygen cylinder be safely attached?

- The patient's nurse/HCA must be able to reach them and provide assistance at any given moment throughout the journey, for example to clear secretions, and so special seating arrangements are necessary.

- Battery life of devices needs to be considered, especially if the patient is travelling for a long distance.(193)

Additional support, on top of the indicative minimum 196 hours, may be needed to enable the patient to further participate in society. The level of required support will vary depending on the individual and contextual factors (environmental and personal considerations). The following activities generally require full support from carers(133):

- education
- work and employment
- caring for others (for example, children)
- recreation, leisure and community life
- religious or spiritual practices.

6.2.4. Assistive technology

Assistive technology refers to “any piece of equipment, or product, whether it is acquired commercially, modified or customised, that is used to increase, maintain or improve the functional abilities of individuals with disabilities”.(133) Assistive technology requirements for high cervical SCI patients, particularly for those who are ventilator-dependent, are substantial and vary considerably depending on the patient’s context, capacity and performance. Importantly, a patient’s needs for assistive technology may change over time, for example should they develop a secondary health condition.(133) Tables 6.1 and 6.2 outlines disposable and non-disposable assistive technology that may be required for ventilator-dependant patients with no motor function below C1-C3 as recommended by the 2017 icare guidance.(133)

Table 6.1: Disposable assistive technology

Disposable items for patients with no motor function below C1–C3

- Antimicrobial filters suitable for use with ventilator
- Heat moisture exchange (e.g. humidivent)
- Tracheostomy care suction catheters 12Fr/30.5cm length
- Reusable blue swivel connectors
- 15mm x 22mm connectors
- Nebuliser with T piece mouthpiece and connecting bush
- Tracheostomy tubes (identical to current tube type and size prescribed as well as one size smaller as well as one size larger and one size smaller) – check exact type (i.e. LPC, cuffless, fenestrated/non-fenestrated).
- Blue flex tubing lengths
- Disposable Foley adaptors
- 50mL syringes
- Large dressing packs
- 10mL ampoules normal saline
- Xylocaine pre-loaded syringes
- Sterile pipe cleaners
- Cotton and velcro tracheostomy tapes
- Normal saline sterile sachets
- Sterile and non-sterile gloves
- Incontinence sheets
- Plastic disposable aprons
- Alcohol wipes
- Disinfectant handwash lotion
- Disposable Yanker sucker
- Sterile lubricating gel tubes
- Female urinary catheters (for use during bowel care – gender is irrelevant)
- Gauze squares
- 10mL ampoules hydrogen peroxide
- Sterile H_2O nebulisers
- Urinary catheters (identical to current catheter size being used as well as one size larger and one size smaller)

• Quick drain catheter tube taps • Urinary drainage 750mL long tube leg bags • Catheter leg straps 45cm • Night bottle • Urinalysis dipsticks • Micropore tape (2.5cm / 1 inch) • Keyhole drain sponges
Additional respiratory disposables for ventilator dependent patients
• Disposable ventilator circuits • Humidifier chamber • Disposable resuscitation bag (e.g. ambi-bag) • Tracheostomy nebuliser kit with appropriate connectors • Spare inner tracheostomy cannula • Large dressing packs • 10mL syringe • Inner cannula cleaning brush • Suction tubing • Dressing as required for around tracheostomy, e.g. split gauze, Allevyn • Passy Muir/speaking valve • Associated consumables for Cough Assist

Note: Table reused with permission from 'icare' New South Wales.(133)

Table 6.2: Non-disposable assistive technology

Non-disposable items for C1–C3 ventilator dependent patients
• Motorised, height-adjustable bed, with appropriate controls and equipment as prescribed including: relevant controls, bolsters, footboard, trendelenburg, head and foot raise, knee break, side/grab rails, linked to environmental control unit; partner bed to be supplied where appropriate • High-care pressure relief mattress, full air replacement with pump • Twenty-four hour drinking system accessible from bed and wheelchair • Mattress overlay for emergency use/when travelling • Power tilt-in-space wheelchair with features as prescribed: head/chin/breath control, postural support devices, power recline and leg raise if required; may include attendant control system • Replacement battery charger • Tyre compressor/pump • Pressure relief wheelchair cushions (2) and covers (2) • Back-up manual or power wheelchair as prescribed • Powered ceiling hoist, and portable electric hoist, hoist charger and batteries (2) • Hoist slings (2 sets) • Shower commode chair with prescribed features or shower trolley • Portable shower hose as required • Over-bed table, height adjustable • Portable lightweight ramps • Slide sheets • Vehicle modifications for attendant-operated wheelchair accessible vehicle with safety (e.g. head and postural support, automatic tie downs) and prescribed features (e.g. air-conditioning) • Independently activated environmental control systems accessible from wheelchair and bed, to include: door opener and intercom, call buzzer/intercom, monitoring system, 'back-to-base' monitored personal alarm, temperature control (reverse cycle air-conditioning at a minimum in the bedroom and living area, fan, heater, blinds), lights, bed, TV/music

- Communication and information assistive technology devices including hardware and software with hands-free access features such as mouth stick, speaker phone, voice activation, eye-operated; devices may include computer/tablet/mobile phone and telephone landline
- Adjustable desk
- Exercise equipment as prescribed
- Splints as prescribed
- Ventilators as prescribed (2)
- Back-up /power source for all powered devices for use when power fails
- Ventilator breathing circuits (3) (specific to ordered ventilator)
- Air Viva resuscitator (2)
- Mains operated suction unit for use in the home
- Evacuation equipment
- Portable suction unit, battery operated
- Breathing circuits as indicated for use with oxygen and air cylinder (2)
- Cough Assist, BiPAP and/or CPAP machine
- Blood pressure monitor
- Thermometer
- Medical grade sheepskin sliding mat, boots and backrest
- Abdominal binders as indicated
- Anti-embolic or compression stockings and gloves

Note: Table reused with permission from 'icare' New South Wales.(133)

6.2.5. Education, information and other supports

As outlined in Chapter 4, providing education, information and support to the patient and to their family, as early as possible and throughout the patient's life course, is recommended across several guidance documents.(79, 90, 130, 132, 134, 138, 139) Patients' needs can change as time progresses and it is important that education, information and supports are available, when needed, to enable patients to reintegrate into society and live a meaningful life. The Canadian SCI Practice Guideline (Can-SCIP) recommend that *"individuals with SCI should have timely access to local peer support services and community-based programs to increase the quality of life and community participation after injury across their lifespan."*(90) Irish SCI patients and their families have called for greater access to ongoing face-to-face supports, including counselling, post discharge.(172) Given the known burden on carers,(198) it is important that supports are readily available, which should include the option of respite care.(129, 134, 137) Respite services may provide relief for families and carers and support their health and well-being,(199) however there is currently no facility in nursing homes or other residential care settings in Ireland to facilitate the short-term care of this patient cohort with such complex care needs.(182) As the patient cohort ages it is important that there are some nursing homes in the country that are prepared to accept ventilator-dependent SCI patients in the near future. Provision of this specialist service may also enable respite care for other patients who are receiving full time care in the home. Consideration should also be given to 'shared care' models whereby patients may spend part of the week at home and the remainder of their time in a hospital or other residential setting.(200) Consideration should be given to exploring the barriers that currently exist to providing domiciliary ventilation in long term care facilities and how these might be addressed. For some patients, where discharge home may not be feasible, this may offer an alternative setting for their long term care.

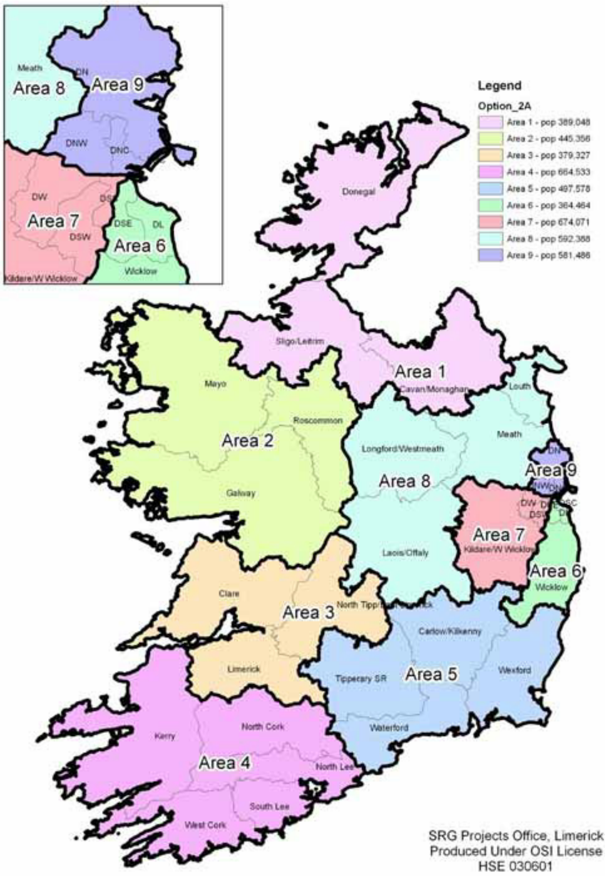
A bespoke national training and support nurse was costed in Chapter 5. The aim of such a role would be to provide ongoing training and support to patients availing of mechanical invasive ventilation and their families and homecare workers in the community. This role would not necessarily be specific for patients with SCI, but would provide training and support to all patients availing of domiciliary invasive ventilation regardless of underlying diagnosis. The total budget impact for establishing and implementing a new bespoke role in the community, was estimated at €0.45 million over a five-year period.

6.3. Meso-level factors

6.3.1. Community Healthcare Organisation and Regional Health Areas

The focus of meso-level organisational factors is on the structure of healthcare organisation. The HSE has established nine Community Healthcare Organisations (CHOs) across the country. These CHOs provide community healthcare services which are defined as *"the broad range of services that are provided outside of the acute hospital system and includes Primary Care, Social Care, Mental Health and Health & Wellbeing Services."* These services are funded by the HSE and other HSE-funded agencies. Within each CHO, a Chief Officer leads a local management team (comprising Head of Primary Care, Head of Social Care, Head of Mental Health, Head of Health & Wellbeing, Lead Quality & Professional Development, GP Lead and Business Management) that focuses on the delivery of all specialist services in their area.(201)

Figure 6.1: Nine CHO areas



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As part of the HSE's Enhanced Community Care (ECC) programme there is a coordinated approach to enhance and increase community healthcare services and reduce pressure on hospital services. The implementation of the ECC is ongoing and is not yet fully embedded across the country. The ECC programme comprises the establishment of 96 Community Healthcare Networks (CHNs), 30 Community Specialist Teams for Older People, 30 Community Specialist Teams for Chronic Disease and 21 Community Intervention Teams across Ireland.(202) Each CHO operates through an average of 10 CHNs, each CHN comprising GPs, nurses and health and social care professionals. A designated GP lead represents GPs at a Network level. Each CHN serves a population of approximately 50,000 people, and consists of between four to six individual primary care teams. The CHNs are further supported by Community Specialist Teams for Older Persons and Chronic Disease.(203)

In line with Sláintecare reforms, there is ongoing implementation of six Regional Health Areas (RHAs), which aims to align hospital and community healthcare services at a regional level, based on defined populations and their local needs (Figure 6.2). This is considered key to delivering on the Sláintecare vision of an integrated health and social care service.(204) The plan is that the nine CHOs and six

Hospital Groups plus Children's Health Ireland will eventually be realigned into these six RHAs. As the current organisation of health and social care does not align geographically nor overlap in terms of geographies, management, clinical oversight, or budgets for defined populations, this significantly hinders the delivery of integrated care.(204) It is envisaged that alignment of acute and community services will have a positive impact on patient care.

Figure 6.2: Six Regional Health Areas

6.3.2. Funding process

There are substantial organisational barriers to the provision of homecare services for ventilator-dependent patients with high cervical SCI. Currently the funding of homecare packages for these patients with substantial care needs is a challenge as these are funded out of various different budgets including local budgets; the funding of high dependency patients may overwhelm local budgets in particular. The budget impact analysis (BIA) in Chapter 5 outlines the substantial funding required for a comprehensive homecare package for patients with SCI who require mechanical invasive ventilation. The BIA estimated that such a package would cost the HSE an estimated €3.4 million per patient over five years. Currently, there is no single source of funding for these patients and so it is not possible to audit spending. There is also no consistent national response to requests for care in the community, which can significantly delay or even prevent the transition of such patients to their home.(2) While there is an overarching integrated care pathway which aims to enable these patients to return to their home with the appropriate supports,(2) there is currently no explicit care pathway within the community for these patients, and there is no mandatory involvement or clear roles and responsibilities for those involved.(189, 205) Consequently, there is no requirement for community teams to provide care, and this leads to inequity of service provision across the country.(189, 205) This presents challenges in that the organisation of care packages is on a case-by-case basis depending on the availability of services in each local region, with no consistency in response.(189, 205)

The funding process is ad-hoc, complicated and can take months if not years to finalise. For example, an assessment of needs is completed by the NRH and if a ventilator-dependent SCI patient requires greater than 56 hours of personal care per week post-discharge (which is highly likely), then an application is made for funding this aspect of their care package through HSE Specialist Services for Older People (regardless of the patient's age).(206) If approved, the HSE can then invite tender submissions from interested parties (that is, private homecare providers) for the provision of home support services, if HSE directly employed staff are not available to deliver such services. The successful parties will then enter a service level agreement with the HSE to provide the services for a defined period. However, neither nursing care nor equipment are covered under the current Older Persons Home Support Tender and require separate funding pathways. Nursing care requests go through local Primary Care and Disability teams.(206) If there is insufficient funding at the local level, the request is escalated initially to CHO level where a business case is made for the individual patient. If there is sufficient funding at the CHO level, then the Chief Officer approves the funding. Otherwise, the request for funding is escalated again to the national HSE office of Disability Operations, where another attempt is made to fund the care package. Disability services do not currently have a national procurement process for any aspects of home support.(206) Separately, equipment in the community falls under the remit of Primary Care. There is a specific procurement on a patient-by-patient basis for specialised equipment (for example, ventilators and consumables), with tendering for the provision of respiratory homecare services (for example, maintenance of ventilators by clinical engineers and provision of consumables etc.).(207) Phrenic Nerve Stimulators (PNS) require additional specialist maintenance, which is undertaken by a company in Finland, in accordance with a structured maintenance programme.(208) As part of this maintenance programme there is an annual visit from the company's engineer at a substantial cost to the HSE. Efficiencies may be gained by the HSE if these engineer visits are coordinated for all PNS users in Ireland, while overview of the timing and need for replacement parts could be delegated to a local clinical engineer.(208, 209)

Generic equipment (for example, beds and hoists) are dealt with under national procurement processes by the HSE.(152) Patients may also be discharged with certain equipment to ensure continuity of care (for example, ventilators, wheelchairs) which come out of the acute hospitals budget as these then need to be replaced by the hospital.(155) Development of a centralised national systematic, dedicated and integrated budget with an associated national tender framework, to cover personal care, nursing care and equipment, would likely alleviate the administrative burden and inequity of the system, and expedite the approval of a fully funded homecare package for these patients. This would also likely assist with budgeting and planning at a national level.

6.3.3. Regulation of homecare service provision

There are important regulatory issues that need to be considered regarding the provision of homecare services in Ireland. The delivery of homecare services in Ireland is complex with a mixture of public (HSE), private, voluntary and not-for-profit entities providing such services.(183) While private, voluntary and not-for-profit homecare providers are subject to monitoring by the HSE via a tendering and service agreement mechanism, the HSE in and of itself is not. This creates a situation where the HSE is not independent of the process. Furthermore, homecare providers that are not delivering homecare on behalf of the HSE (that is, outside of tender or service arrangements) in theory do not need to adhere to any standards, though they may be audited by their national organisation. In this regard, there is no control over who can provide such services and how they are monitored. The lack of independent oversight and regulation in the provision of homecare is problematic, though the Irish Government has indicated that it intends to develop an appropriate, bespoke regulatory framework for this setting.(183) In addition, there is currently no statutory scheme for homecare in Ireland, and so there is no obligation on the HSE to provide 'home help' services, which makes the provision of domiciliary ventilation a challenge (though there is a commitment to implement such a statutory scheme as part of the Sláintecare reforms).(183) This is in contrast to other countries; for example, Finland enacted legislation in 1992 to guarantee equal rights for home patients dependent on invasive mechanical ventilation regardless of the patient's socioeconomic status, religion or underlying diagnosis.(97)

It is worth noting that patients with spinal injuries receiving invasive ventilation support to enable them to live at home in their community will not be included as part of the care or support being regulated by HIQA under the proposed new home support legislative framework. It is essential therefore, that the clinical oversight and governance of the care and support for these patients ensure that all patients are supported through safe, quality care by experienced staff.

6.3.4. Healthcare staff capacity in the community

Extensive case conferences and negotiations are often required between the NRH and community services in terms of organising the required level of homecare for these patients, prior to discharge, even if funding is approved.(189, 205) Recruitment, retention and training of homecare staff is a particular challenge.(183) It was also noted that there are ongoing pay parity issues between homecare staff working in disability and those working in older person’s services, which is possibly further exacerbating retention issues in the homecare disability sector in particular.(193) Given the 24/7 care required from specialist trained nursing and other homecare staff (a minimum of 196 hours of assistance per week is recommended for these patients)(133) along with the regular involvement of other members of the primary care team to keep these patients in their own home, there is a critical need for continuity of care. Continuity of care 24/7 is essential for these patients as they must be under the direct supervision of a competent person at all times, and this must factor into staff scheduling. A protective reserve of trained staff is important to ensure 24/7 care can be maintained in instances where staff are unable to work at short notice. There are also long waiting lists for critical community services such as occupational therapists, physiotherapists and clinical psychologists.(168) For example, at the end of the third quarter of 2022 (July-September), 63,838 people were on the assessment waiting list for physiotherapy, of whom 14,088 had been waiting over a year. Similarly, 35,826 people were on the assessment waiting list for occupational therapy, of whom 9,551 had been waiting over a year. In terms of treatment waiting lists, a total of 15,255 were waiting for psychology, 5,518 of whom had been waiting over a year.(168) As discussed in Chapter 4, care coordination is critical to ensure that informal and formal supports, from all required disciplines, work effectively together to ensure the best outcomes for the patient with SCI.(133) Any gap in, or poor quality of, service provision may result in harm to the patient and or readmission to hospital or residential care.(2)

6.3.5. Primary care services

In terms of the referral of these patients to health and social care professionals in the community, initially the NRH or another acute hospital would liaise with these disciplines. Once the patient is finished with this episode of care (for example, the requirement for a hoist) the patients is ‘discharged’ from their books. However, once a patient in the community has nursing needs, they cannot be discharged from Public Health Nursing. The Public Health Nurse would make the referrals episodically, as required to physiotherapy, occupational therapy etc. for a usual maximum of six sessions, and they would then be discharged from their books.(167) Other parties, including the patient themselves, can also make referrals (for example, GP, hospital), and often with more complex cases there is a case manager who acts as a conduit. With the establishment of the new CHNs, there will be a network manager within each CHN which should help with all the liaisons.(167) However, given the ongoing need for care from these services,(90) the referral process can be administratively challenging and burdensome.

The role of the GP in relation to the care of these patients in their own home is important to consider. Given the rarity of these occurences, many GPs may have limited, if any, experience in the provision of such complex care. There are also important issues with regards to the clinical governance given that these patients may be located a substantial distance away from an acute hospital and the GP may not be readily available to provide care at short notice, particularly out of hours. Given the complex care associated with SCI patients, the capitation fee paid to GPs may not reflect the workload of patient management. It is important to acknowledge the workforce challenge, the workload and the responsibility for GPs who are caring for these patients with complex care needs in the community.(210) A bespoke national training and support nurse as costed in Chapter 5, could have an important role in providing education and training to GPs involved in the care of these patients, however the workforce challenges facing GPs are an ongoing issue. It should be borne in mind that these patients will have 24/7 care provided by competent homecare staff, which may alleviate some, but not necessarily all, of the demand for GP.

As discussed in Chapter 4, various international guidance documents emphasise the need for an ongoing treatment programme for the patient in their home delivered by an interdisciplinary community-based team (for example, physiotherapy, occupational therapy, clinical psychology).(79, 134, 139) However, it is important to acknowledge that in Ireland no such community-based team currently exists specifically for this patient cohort. When such interdisciplinary assessments are facilitated, these are usually done by hospital-based specialists out of inpatient time, which has a knock-on impact on inpatient services. Greater access to a specialist outreach interdisciplinary team may be beneficial for the patient and may reduce the need for the involvement of the primary care team who may have limited expertise in the management of this patient cohort.(161) The lack of SCI expertise among primary care staff has also been noted as a particular challenge when the NRH recommend the purchase of specialised equipment for community-based patients.

Upskilling of primary care colleagues with regards to the use of these equipment is another important consideration in the provision of a national service.(182)

6.3.6. Care model

As discussed in Chapter 4, there is a lack of consensus internationally on the requirement for nursing qualifications in the provision of care to these patients. Some countries, such as Germany, explicitly require trained nurses to provide this 24/7 care,(79, 139) whereas others, such as New Zealand, refer to oversight by nurses with direct patient care being provided by 'competent' carers.(126) Similarly, it is reported that in the UK, trained healthcare assistants (HCAs), who are deemed competent by the relevant authorities, provide care to these patients with supervision by nurses, rather than a 24 hour nursing care model.(2, 143) Substituting 24/7 nursing care with 24/7 trained HCA care with appropriate nursing oversight would reduce the overall cost of a care package for these patients and would likely increase the availability of staff (Chapter 5). However, careful consideration would need to be given to clinical governance and implications for nurses' scope of practice (Chapter 7),(184) should such changes be recommended. Another important consideration is with regard to staff breaks to ensure that there is always at least one trained staff member watching over the patient at all times.(193) As discussed, a bespoke national training and support nurse was costed in Chapter 5. It is possible that such a role could be developed to provide ongoing training to homecare staff, ensuring that they are competent to provide the level of care required for these patients. Provision of competency-based training and specialised further education and training (FET) courses for HCAs to upskill in this area may be important given the global shortage of nurses.(211)

Consideration also needs to be given to the delivery of maintenance treatments (for example, stretching and passive movements, application of splints etc.) as part of the home care package.(161) International guidance documents recommend that *"individuals with SCI be referred to community-based exercise programs with periodic reassessment by an SCI clinician to maintain their fitness and wellness and optimise and monitor their function."*(90) Patients with complete C1-C3 injuries are entirely dependent on others to move their limbs and so involvement of homecare staff and physiotherapy is important. However, clarity is needed as to whether these treatments can be viewed as 'routine care' and so can be provided by a trained HCA or nurse on duty, rather than provided by the physiotherapist only. Given the limited availability of community physiotherapists, reliance on physiotherapists to deliver these maintenance treatments may be restrictive, with clinical opinion suggesting that many of these maintenance treatments can be effectively delivered by trained homecare staff.(161)

6.3.7. Clinical governance

There are also some uncertainties regarding the clinical governance for these patients in the community. While the integrated care pathway for the management of SCI was developed by the National Clinical Programme for Rehabilitation Medicine,(2) there may be benefits from involving other Clinical Programmes such as Respiratory Medicine in further developing the care pathway into the community, given that these ventilated SCI patients have chronic respiratory failure and require oversight from that clinical perspective, as is common in other jurisdictions.(88, 134, 138) For example, in the Netherlands there are four national centres that are responsible for all patients who need domiciliary ventilation, regardless of underlying diagnosis,(11) all four of which are affiliated with a teaching hospital.(138) Patients must be referred into one of these centres before domiciliary ventilation will be funded by the Dutch Healthcare authority.(11) These four centres are also responsible for the ongoing education and training needs of carers and healthcare professionals, and are available 24/7 to provide medical advice if required.(11, 138) In the Dutch system there is also a clear division of tasks and responsibilities between all relevant parties during the care pathway for these patients, including discharge, at home, and re-admission (Appendix 3).(11, 138) It is important that the Irish care pathway is clear and that patients, families and homecare workers all know who to contact and when during different scenarios, particularly during emergency/crisis situations which may require the explicit involvement of local acute hospital services. Consideration should be given to having an identified clinician, linked to the National Clinical Programme for Rehabilitation Medicine, assigned with overall clinical responsibility for these patients. It is also important that the overarching clinical governance structure is suitably aligned to the relevant National Clinical Programmes to support the provision of high quality care to these patients.

6.3.8. Paediatric service organisation

The current HTA is focused on adults with spinal cord injuries. However, a different model of complex discharge planning is in operation for paediatric services in Ireland, which may have some relevance for adults with cervical SCI given the current ad hoc nature of this process. Within paediatric services, complex discharge planning is a six-stage process, overseen by a governance group and a complex child care group. The complex child care group has representation from all key stakeholders including specialist hospital services, local hospital services and community based organisations, with clear roles and responsibilities for all parties. The complex child care group has access to a ring-fenced budget from which funding of home care packages is provided. Each child is assessed by a national standard assessment tool to determine their level of complexity. The tool supports clinical decision-making processes and helps develop and tailor services by directing resources. Once complexity is confirmed, the child falls under the remit of the complex childcare system. Exact levels of support are determined by the multidisciplinary team led by a nominated key worker/coordinator. Following this, a business case is developed and submitted to the complex childcare group for consideration and approval. This process is initiated once the lead clinician has determined that home care is the preferred option for the child. The discharge planning stage is a defined process with associated recommended timeframes.(2) The Integrated Care Pathway for the Management of SCI recommends the development of a model of complex discharge planning similar to that used for paediatric services. As is the case with paediatric services, the completion of a specialist rehabilitation needs assessment/prescription should trigger the commencement of a planned release of funding to ensure timely care for those ventilator-dependent patients with high cervical SCI most at need of multiagency input.(2)

While SCI in children are exceedingly rare (estimated annual cumulative incidence of 1.4 per million per year for traumatic SCI and 1.6 per million per year from non-traumatic SCI in Ireland between 2000 and 2015), particularly injuries leading to complete tetraplegia,(212) it is important that due consideration is given to ventilator-dependent children with SCI who transition into adult services. The current admission criteria for the inpatient spinal cord system of care programme in the NRH is that patients must be at least 16 years of age. Patients younger than 16 years requiring the services of the Spinal Cord System of Care are admitted under the Paediatric Family-Centred Programme.(192) The Paediatric Family-Centred Rehabilitation Programme is the national medical rehabilitation service for children and adolescents (birth – 18 years) requiring a complex specialised rehabilitation service. The Paediatric Programme is a microcosm of the three adult Programmes at NRH, providing rehabilitation services to children and adolescents who have significant injuries and who require medical rehabilitation as a result of conditions such as a brain injury, stroke, spinal cord injury, neurological disorders or limb absence.(213) Some guidance was identified as part of the Scoping Review in Chapter 4 in relation to transitioning from children to adult SCI services which may be useful in informing the national provision of care, and are detailed in Appendix 3. For example, in relation to transitioning from child to adult home mechanical ventilation services, the Dutch guidance recommends the following(138):

- follow the recommendations of the national Transition Care Quality Standard
- start the transition as early as possible after the 12th year of life with a clear plan
- start the transition with the parents

- make an overview of all healthcare providers involved before and after the transfer
- take time to ensure a smooth transfer to the new main practitioner (usually a general practitioner)
- train peers in ventilation care, so that they can go out with the young people as buddies
- make and communicate clear agreements about who to call in case of medical problems
- identify who will direct the transition
- make and communicate clear agreements about the care plan for young people who are not able to make decisions for themselves.

6.3.9. Maintenance of equipment

The HSE tenders out contracts with private companies for home patient services (for example, maintenance of equipment, and supply of gases).(207) However a lack of clarity has been reported in terms of the overall responsibility for some equipment that has been recommended by the NRH for use in the community by discharged patients, or with regards to equipment purchased by the NRH that has been discharged with the patient into the community.(214) When developing the service level agreement there are certain factors that should be carefully considered in the context of providing home services for this patient cohort, including the(160):

- response time for ventilator management as emergency/out-of-hours call outs may add to service costs
- fact that parts and consumables for certain ventilators may be hard to obtain in a timely manner.
- size of oxygen cylinder used and the frequency of oxygen delivery as this may impact on the type of ventilator purchased and energy costs.
- location of the patient, as this may be a factor in the delivery and availability of gas cylinders and so oxygen concentrators may be required, which have a substantial capital and running cost.

- designation of responsibility for each equipment (including repairs, electrical safety testing and management of field safety notices and recalls) given the broad range of equipment required for these patients, and a clear understanding of responsibilities among care providers.

Clinical engineers play a critical role with respect to the maintenance of equipment for these patients in the community and should be central to these discussions.

6.4. Macro-level factors

Macro-level factors relate to health policy and its role in influencing healthcare organisation. In light of Sláintecare,(8) the Delayed Discharges Independent Expert Review,(215) the Integrated Care Pathway for the Management of SCI,(2) the Ombudsman's report on Younger People in Nursing Homes,(216) and HIQA's Position Paper on the Regulation of Homecare,(217) there is currently a policy initiative for the safe discharge home of these patients with an appropriately funded package of care in the context of a statutory homecare scheme.

6.4.1. Sláintecare and the Delayed Discharges Independent Expert Review

In line with Sláintecare reforms, there is an ambition to achieve a universal single-tier health and social care system, which provides equitable access to services based on need, and not ability to pay. One of the key objectives of Sláintecare reforms is about delivering the right care, in the right place, at the right time, by the right team.(8) Specifically, a key aspect of the reform is the shift of care out of acute hospitals into the community and closer to a person's home, where safe to do so. In line with international best practice and guidance, domiciliary ventilation is considered the standard of care for ventilator-dependent patients with SCI where clinically appropriate.(9-12) Given the high occupancy levels that Irish hospitals currently experience, combined with the harms that a delayed discharge can cause the patient (for example, deconditioning and susceptibility to infection) and the cost to the healthcare service, as outlined in the Independent Expert Review of Delayed Discharges,(215) there is added incentive to get these patients home safely. Another aspect of Sláintecare is in relation to equitable access to services.(8) Given the current inequity in service provision for these patients, care is largely informed by budget and healthcare staff availability, rather than on patients' needs. Therefore, the return home for these patients as soon as safely possible with an appropriately funded and equitable package of care, would be in keeping with the ideals of Sláintecare.(8)

6.4.2. HSE Integrated Care Pathway for Patients with SCI

As outlined in Chapter 3, an integrated care pathway for patients with SCI has been developed in Ireland by the HSE's National Clinical Programme for Rehabilitation Medicine, in line with its model of care.(2, 7) Integrated care pathways are structured multidisciplinary care plans which detail essential steps in the care of patients with a specific clinical problem.(119) The care pathway for the management of SCI extends from pre-hospital care through to discharge and lifelong care. It is currently policy of the National Clinical Programme to get ventilator-dependent SCI patients home with an appropriately funded care package, however there is an acknowledgement that there are many barriers to this, as previously discussed (for example, complex funding pathways, inequitable response to care requests, significant training and education requirements, home care staff recruitment and retention issues etc.)(2) The Integrated Care Pathway policy document recommends several key steps to enable a more equitable and efficient care pathway for these patients, including(2):

- the development of a centralised funding stream.
- flexible funding determined by patients' needs as they change over time, as opposed to block funding.
- earlier engagement from social worker/case worker in relation to medical card applications and housing adaptation grants.
- homecare delivered by competent healthcare assistants with nursing oversight, rather than the need for 24/7 nursing care.
- further education and training for homecare staff and families, that takes into consideration the rapid turnover rates of staff.
- development of a model of complex discharge planning similar to that used for paediatric services.

6.4.3. Ombudsman's report on Younger People in Nursing Homes

The 'Wasted Lives: Time for a better future for younger people in Nursing Homes' report, published in 2021, describes the findings of an investigation by the Ombudsman into the appropriateness of the placement of people under 65 years old in nursing homes.(216) The HSE reported that as of 30 June 2020, there were 1,320 people under 65 years supported by the Fair Deal Scheme. The report highlighted the challenges faced by these individuals living in congregated settings designed for older people, with the appropriateness of the placement for these people questioned by the Ombudsman. Many of those interviewed as part of this investigation expressed a preference to return home but were not supported to do so. One of the key financial inequities highlighted was that nursing home care is available on a statutory basis but home care is not, which results in some younger people with disabilities ending up in nursing homes long term. Overall, the investigation found that Ireland still has progress to make in advancing from a medical model of disability to a social model, and that various changes need to be made to the system to facilitate a person-centred approach to care and one which is in keeping with the United Nations Convention on the Rights of Persons with Disabilities (UNCRPD) and Ireland's own strategic approach to disability.(216) One of the key recommendations from this report was that *"a ring fenced annual budget should be allocated to each CHO Area in order to improve the quality of life of each of these individuals and to assist them in leaving nursing homes, if that is their preference, and to support and enable them to enter into more appropriate living arrangements."* *(216)* The report strongly advocates that, with the appropriate supports, many young people with disabilities can avoid institutionalisation in nursing home settings.

6.4.4. HIQA Position Paper on the Regulation of Homecare

In December 2021, HIQA published a report outlining its position on the future regulation of homecare services in Ireland.(217) As homecare remains a discretionary but demand-led service where need often outstrips supply, HIQA state that the current homecare system is not sustainable and is not meeting the needs of people. In order to achieve a well-functioning, transparent and equitable homecare system, HIQA emphasises the need to ensure that all stakeholders work with the person receiving homecare within a defined, national and equally applied framework underpinned by legislation. The most recent Programme for Government has committed to introducing a statutory homecare scheme. In the context of this impending legislation, HIQA advocates for a *"needs-led, integrated homecare system where age is removed as an access barrier and all people can receive care in their home on a regular or intermittent basis when required throughout their lifetime."* HIQA further recommends that *"the fundamental principles of a human rights-based approach — in addition to safety and wellbeing, responsiveness and accountability — should underpin the legislation, standards and regulation of this sector."(217)* There is a policy movement towards independent regulation of the homecare sector, which is important to factor into the planning for the delivery of homecare services for ventilated SCI patients.

6.5. Discussion

The purpose of this Chapter was to provide an assessment of necessary organisational changes that would be associated with the provision of domiciliary ventilation to patients with SCI on a national footing. The information presented has been collated based on the scoping review of international practice and standards (Chapter 4). To put findings into an Irish context this was followed by engagement with stakeholders from the National Clinical Programme for Rehabilitation Medicine, the Spinal Cord System of Care Programme, the NRH, the HSE, and a patient representative organisation. It is important to re-emphasise that it is already standard practice to discharge this patient cohort to their homes as soon as it is safe to do so, however there is currently no clear pathway into the community, no explicit discharge criteria and substantial challenges to securing the required funding and staffing.(2) The organisational analysis undertaken in the Chapter therefore addresses the overall aim of organising and delivering high quality, fully funded domiciliary ventilation for these patients on a national footing, and outlines the associated barriers, enablers, and key considerations otherwise relevant to decision-makers with respect to this aim.

The organisational considerations for this HTA were discussed at three different levels of the healthcare system (micro-, meso- and macro-level). The micro-level factors relate primarily to patient and family interactions; the meso-level relate to healthcare organisation and the community; and macro-level relate to health policy.(190) At the micro-level, consistent with international best practice, important enablers of domiciliary ventilation provision were identified as including:

- development of explicit discharge criteria and protocols to assist with the complex discharge process and so that patients and families can have realistic expectations for the future
- a more flexible process for obtaining housing adaptation funding, that can enable families to carry out necessary renovations, provision of housing for those without their own home, and greater access to step down units to facilitate the transition home and respite services to provide relief for carers and families
- provision of the recommended supports and services to enable the patient to engage in their communities to the fullest possible extent (see Chapter 5)
- greater access to ongoing face-to-face supports for patients and families in the community.

Similarly, at the meso-level, there are a number of important organisational factors that decision-makers need to consider such as:

- development of a centralised national systematic, dedicated and integrated budget with an associated national tender framework to cover personal care, nursing care and equipment; such a budget would likely alleviate the administrative burden and inequity of the system, and expedite the approval of a fully funded homecare package for these patients.
- development of a model of complex discharge planning similar to that used for paediatric services.
- development of an explicit care pathway within the community for these patients, with defined involvement and clear roles and responsibilities for all involved parties, including processes for the transfer of ventilator-dependent children with SCI into adult services.
- care coordination, in the context of healthcare staffing issues, to ensure that informal and formal supports, from all required disciplines, work effectively together to ensure the best outcomes for the patient with SCI.
- whether healthcare assistants providing care with nursing oversight is feasible and appropriate, and how competency can be assured in those providing care
- development of a bespoke training and support role in the community to provide quality assurance of domiciliary invasive ventilation services
- involving other Clinical Programmes in further developing the care pathway into the community
- involving clinical engineers with respect to the maintenance of equipment for these patients in the community, and having a clear understanding of the governance of these equipment.

As outlined in Chapter 5, there is a substantial budget associated with the provision of a comprehensive package of care for these patients, estimated at €3.4 million per patient over five years, along with the creation of a bespoke role in the community that would provide ongoing training and support to patients, families and homecare workers costed at €0.45 million over five years. In addition, substantial reorganisation of funding and clinical governance processes would be necessary to implement the advice from this HTA, which may come at a substantial organisational opportunity cost. While there are many challenges to the implementation of an equitable and streamlined mechanism for the funding and delivery of homecare packages for these patients as outlined above, there are also important policy enablers largely centred on one of the key aims of Sláintecare to shift care out of acute hospitals into the community and closer to a person's home, where safe to do so.(8) Implementation must be within the context of a finite budget and, where possible, in line with evidence-based practice. The decision-maker should recognise the extensive care and support needs of the individual patient while balancing this with providing care to all those who need it in the wider community.

In summary, should a decision be made by the HSE to implement a national systematic, dedicated and integrated budget with an associated national tender framework for the provision of homecare to invasively ventilated patients with SCI, this will require substantial organisational cooperation across multiple agencies and programmes. Consideration should be given to organisational factors at the micro-, meso- and macro- levels when coming to a decision.

7. Patient, social and ethical considerations

Key points

- This Chapter outlines the potential patient, social and ethical considerations relating to the provision of domiciliary invasive ventilation services for adults with spinal cord injuries (SCI). The considerations outlined are framed both in the context of the experiences of the patient and their family, and also through an ethical lens.
- Living at home on a ventilator with high cervical SCI is associated with substantial challenges for both the patient and their family. Patients have expressed how these life-changing injuries are catastrophic and can often require a prolonged period of adjustment. The complications arising from the injuries are profound and can have a hugely negative impact on patients requiring round-the-clock care from homecare staff.
- Before the patient can be discharged with a home care package, the home environment must be adapted to meet the medical and accessibility needs of the patient, which can be a considerable financial burden to patients and their families. Fundraising and the charitable support of the wider community has been noted to be an important source of financing for affected families.
- Families have expressed their strong desire to provide the best possible care for their loved ones, but often to their own detriment (physically, emotionally and financially). Carer burden is particularly problematic among these family carers, given the high level of dependency. Greater provision of paid care, professional support, and respite care have been recommended in the literature to alleviate carer burden and make care provision more sustainable.
- The role of the homecare worker in the patient’s home is complex and challenging. Homecare workers have a difficult balancing act of managing the intricate care needs of the patient while respecting the privacy of the family, which can sometimes lead to tensions when there are conflicting goals. Good communication between the patient, family and homecare worker is important for establishing trust.
- Issues such as high staff turnover and training insufficiencies represent serious risks to achieving sustained high quality care for these patients. Patients need ongoing support from competent homecare workers with an understanding and

knowledge of the issues related to living with a spinal cord injury and requiring ventilator care.

- However, these patients have also demonstrated resilience in adapting to their 'new normal' and a willingness to live a fulfilling life despite the obstacles. Provision of the required supports and services for these patients and their families, on an ongoing basis, may alleviate some of the hardship and make the care provision more sustainable. The importance of a good relationship with family and friends is considered an important factor that motivates patients and adds value to their lived experience.
- The four principles approach to biomedical ethics (otherwise known as principlism) was used as a framework for the ethical analysis: respect for autonomy (that is, respecting the decision-making capacities of an autonomous person); non-maleficence (that is, avoidance of harm), beneficence (that is, providing benefits and balancing benefits against risks and costs); and justice (that is, fairness in the distribution of benefits, risks and costs).
- In terms of respect for autonomy, provision of a national delivery of care for these patients should be viewed as supportive of patient autonomy where a clear preference to move home is expressed. However, it is crucial that patients and families are fully informed of the benefits and risks of domiciliary ventilation, and that a true choice of options is provided to the patient that respects the preference of the individual.
- In terms of the benefit-harm balance, consideration needs to be given to the potential for the benefits of this national provision of care to be outweighed by its associated harms. Some of the key benefits are that it would respect patients' preferences to receive care at home, would likely free up scarce intensive care unit (ICU)/high dependency unit (HDU) beds and may improve patients' quality of life. However, this needs to be weighed against the increased safety concerns associated with providing complex care at home, the substantial burden on families, and the opportunity cost to other patient groups.
- From the perspective of justice and equity, provision of a systematic nationwide service may improve equity of access for these patients given the current geographical disparities in funding and service provision. Creation of a nationwide ring-fenced budget could prevent local budgets being depleted, allowing funding for other important community services to be preserved. However, it is associated with a significant opportunity cost given the substantial nursing requirements. It could also be argued that provision of a dedicated nationwide service exclusively to invasively ventilated patients with

C1-C3 SCI may not be equitable to other patient groups, who may have the potential to benefit from access to this service.

7.1. Introduction

There are many factors that need to be considered prior to the implementation of a health technology. This Chapter outlines the potential patient, social and ethical considerations relating to domiciliary invasive ventilation for adults with SCI. Patient and social aspects are grouped together and described in section 7.2, while ethical considerations are discussed in section 7.3.

7.2. Patient and social considerations

7.2.1. Introduction

The purpose of this section is to discuss the patient and social aspects that should be considered in relation to the national delivery of care for adults (including young adults ≥ 16 years old) with spinal cord injuries (SCI) who require permanent invasive mechanical ventilation and in whom discharge home is deemed clinically appropriate. In the context of this section, the term 'patient aspects' refers to issues such as the burden, experiences and expectations that are relevant to patients, individuals and carers. The term 'social aspects' refers to issues experienced by groups or communities of patients or individuals that may be relevant to the topic under assessment.(190) The focus of this HTA is on an individual's private (domestic) home, although it is acknowledged that other care settings can be relevant to this cohort of individuals, including residential care settings.

A review of the published literature was conducted to inform this section on patient and social considerations. This was supplemented by an interview with a patient representative from Spinal Injuries Ireland conducted on 16 January 2023, discussing the patient and family experience of domiciliary ventilation in Ireland.(172) The analysis of this information was guided by the patient and social domain of the EUnetHTA HTA Core Model Version 3.0.(190)

7.2.2. The experience of living with a cervical spinal cord injury and impact of requiring ventilation

7.2.2.1. Living with a spinal cord injury

As described in Chapter 2, high cervical SCI can be catastrophic for the patient, with injury to the spinal cord resulting from either acute trauma or a non-traumatic cause. The medical management for the patient will depend on the type, level and completeness of injury. These injuries can be extensive and can include: permanent tetraplegia; respiratory failure requiring continuous mechanical invasive ventilation, often by tracheostomy; and a range of other medical and daily care requirements such as bowel and bladder dysfunction, and skin pressure injury prevention. Consequently, these patients are typically entirely dependent on others for their medical and daily care needs, making them a highly vulnerable group.(218)

Through their life-changing consequences, high cervical SCI can be seen to present a sudden and traumatic interruption to the lifecourse of the patient. Thus, in addition to their immediate medical requirements and rehabilitation needs, it is important that patients are supported in coming to terms with their new reality, and receive appropriate information about how to manage their condition.(219) A study exploring patients' perspectives on life following a traumatic SCI found that patients have expressed a need for more guidance around daily care needs, such as bowel, bladder and pressure injury management, medication, and proper use of the urinary catheter.(219) Patients described having some combination of self-motivation, spiritual connection and support from friends and family members as critical in helping them to cope with their injuries and live as normal a life as possible.(219) The importance of a good relationship with family and friends has previously been described as the major factor that motivates patients and adds value to their lived experience.(220)

Evidence supports the provision of early and intensive rehabilitation programmes to individuals affected by traumatic injury.(74) According to the National Institute for Health and Care Excellence (NICE), rehabilitation should be started as soon as possible after a traumatic injury, when the person is ready and able to engage.(75) During this period of rehabilitation and care in hospital, information and support is offered to the patient as part of the care pathway.(2) However, this is a difficult and emotionally overwhelming time for the patient.(220) It may therefore be challenging for the patient to take in and process this information during this time. Provision of supports and information may be beneficial if also provided later in the care pathway and for patients in community settings, when they may be in a better frame of mind to process such information.(172, 219) In addition to being provided at appropriate times and regular intervals through the care journey, it is important that care decisions are tailored to comprise realistic and patient-centred treatment goals.(117)

7.2.2.2. The impact of requiring ventilation

The need for continuous invasive mechanical ventilation has a profound impact on the daily life of patients with high cervical SCI. A 2018 systematic review highlighted the importance of information provision and counselling for patients and carers, before and after receiving tracheostomy.(221) Negative experiences may leave patients and carers feeling scared, overwhelmed, unsupported and powerless. Long-term invasive mechanical intervention may negatively impact a patient's ability to communicate as it can weaken their voice and lead to a dependency on others for communication.(222) However, patients may experience feelings of relief, in that the ventilator support allowed them to continue to breathe following their injury.(220) In this sense, the literature describes a dual aspect to how patients may perceive ventilation support. As described in multiple publications by Lindahl et al., capturing patients' experiences of mechanical ventilation, the ventilator may be perceived as both a source of freedom and imprisonment.(223, 224) Initial feelings of vulnerability arising from a dependence on technology may eventually subside as patients view it as a vital force enabling them to rest and live at home. Being on a ventilator was reported to be associated with patients demonstrating resilience, patience and bravery, and described like overcoming an obstacle.(223) Comfort and confidence in the technology has been described as a gradual process, facilitated by gaining knowledge, care and time.(224) In essence, the patient has a psychological relationship with the ventilator and it is important that trust in its use is not undermined. This may be particularly important in situations where there is potentially high turnover of staff and this could affect this relationship.

This dependency also highlights the importance of the relationships that patients have with carers and health professionals.(117) According to a study of ventilated individuals in Germany, patient comfort is based on trusting relationships between staff, patients and their families.(117) Care should be person-centred and participants highlighted the importance of "*access to the right support, from the right person, at the right time*".(117) These values align with one of the key objectives of Sláintecare reforms to the Irish health and social care system.(8) Specifically, a key element of these reforms is the movement of care from acute hospitals to the community and closer to a person's home, where safe and appropriate to do so, as outlined in Chapter 1.

7.2.3. The experience and expectations relating to domiciliary ventilation

7.2.3.1. Preparation and discharge home

According to the National Clinical Programme for Rehabilitation Medicine's integrated care pathway for the management of SCI, a return to home with an appropriately funded care package would be considered the optimal outcome for many patients with significant dependency needs.(2) Domiciliary ventilation facilitates a more normal everyday life than that permitted from a hospital or residential setting. The provision of domiciliary ventilation has been reported as being an important option for patients, with living at home being valued and contributing to sense of control in everyday life.(218) On the other hand, it is a difficult life situation involving major psychological, physiological, social, and existential challenges.(218, 220) Dependency on technology and others can contribute to feelings of uncertainty and anxiety for patients.(218) It has been reported in the literature that patients can sometimes feel pressured to accept domiciliary ventilation,(218) however there is no evidence that this has occurred in Ireland. The decision for the patient to move to domiciliary ventilation requires careful and extensive assessment, training, education and preparation with respect to the patient, carers and other family members, and health professionals.(2, 218) In this way, healthcare services should be patient-centred with a clear focus on patient involvement and cooperation. An important component of this is providing patients and carers with appropriate information, in non-technical language, at the beginning phase and transition to domiciliary ventilation.(218) It is critical that fully informed consent is provided by patients given the risks that are associated with providing complex care at home (Section 7.3.3.1)

Before the patient can be discharged with a home care package, the home environment must be adapted to meet the medical and accessibility needs of the patient, which may impact on the patient and family experience. These aspects are described in more detail in Section 6. Depending on the home in question, this may require adaptation and refurbishment, which can be a considerable financial burden to patients and their families and may act as a barrier to providing domiciliary ventilation,(91) particularly for those from a lower socioeconomic group.(225) Fundraising and the charitable support of the wider community has been noted to be an important source of financing for affected families in Ireland.(172)

A family member, typically a spouse or parent, will often assume caregiving responsibilities for the patient. This may require this individual to reduce or stop their employment, contributing to the financial burden they experience.(117) Financial uncertainty may also result in delays in moving the patient home. In addition, patients may experience an extended length of time spent in hospital waiting for homecare workers to be trained in ventilation management.(2) In discussions with a representative of Spinal Injuries Ireland to inform this HTA, it was noted that while patients in Ireland have generally reported satisfaction with the care packages they received, they and their family members noted the length of time waiting to receive the care package to have negatively impacted their experience.(172)

7.2.3.2. Living at home

The provision of domiciliary ventilation services permitting the patient to live at home is an important option for patients and family.(218) It has been noted that patients appreciate the improvement to their quality of life brought about by domiciliary ventilation. However, the daily life situation for these patients is profoundly complex and the benefits of living at home are counterbalanced by both the increased safety risks (for example, ventilator failure) and complications to care (for example, dealing with medical emergencies) compared with being in a hospital setting,(2) and the continuous difficulties patients can experience in coming to terms with their personal situation and the challenges they face.(218)

Transformation of the home environment

Domiciliary ventilation requires a transformation of the home environment into a more medicalised setting, with the presence of medical equipment, supplies and homecare workers in the family's private dwelling. As a consequence, this can alter relationships and change what it means to be at home for the patient and their family.(226) At the same time, the one space can be experienced as a home, by the patient and their family, and as a workplace, by the homecare workers and family carers.(226) Patients and family carers may experience a loss of privacy due to the medicalisation of their home and presence of homecare workers.(227) This change in the meaning of home by the presence of technology and homecare workers has been found to be challenging for patients and their families.(226)

Interpersonal relationships

As has been stated, those with high cervical SCI (C1-C3) are often dependent on ventilator support due to compromised respiratory capacity. They require 24/7 care all year round.(2) Homecare workers and family carers are responsible for looking after the medical and daily care needs of the patient. The need for extensive surveillance to monitor the wellbeing of the patient can give rise to a sense of undermined autonomy and self-determinism and loss of privacy.(218) It can be difficult for patients to adjust to having people around them all the time in their own home. Therefore, collaboration between patients and those providing care can be challenging. Patients can experience a more positive relationship with those providing care when there is consideration of their needs and preferences about daily living, and feelings of safety and trust in the competency of the person providing care.(218) According to Martinsen et al., 'good helpers' act in ways that are responsive and sensitive to the expressed needs of the patient. Helpers, be they homecare workers or family carers, can facilitate the patient experiencing creative participation in daily life through organised projects and everyday activities. Patients may recognise aspects of themselves through the actions of a helper performing a task as close as they can to how the patients would have done it themselves.(228) Qualitative interviews with 19 individuals receiving domiciliary ventilation in Sweden highlighted the importance of communication as a way for the patient to express their preferences to others, thereby experiencing a sense of control and empowerment.(222) These positive outcomes highlight that it is critical that helpers practice patience and are supported to give time to listen to what patients have to say.(117)

Although the provision of domiciliary ventilation provides patients with independence and has the potential to improve their quality of life, it may be accompanied with a substantial burden on families.(229) In a qualitative study, family carers described a sense of duty to take care of loved ones, but suffered a significant restriction of their own time with a negative impact on their physical and mental health. This particular study found that the initial transfer home was the most stressful part of the process for families.(229) Carer burden is further discussed in Section 7.2.3.5.

The role of the paid homecare worker is complex and challenging. Homecare workers have a difficult balancing act of managing the intricate care needs of the patient while respecting the privacy of the family.(227) Discordance between the patient, family and homecare workers has been reported by Heggested et al. For example, homecare workers may have differing views to the patient and or family with respect to viewing safety and risk minimisation as a priority; a patient and their family may be willing to accept higher levels of risk in the manner in which they want to live their daily life at home.(230) The ethical considerations of such circumstances are discussed in Section 7.3. Similarly, Dale et al. describe potential sources of conflict between family carers and homecare workers. Family carers may have specific knowledge of the various care requirements of their family member and feel accountable for their care in the home. Tensions may arise as above in instances where choices and actions taken by family carers in the interest of the patient are called into question by homecare workers.(231) Furthermore, homecare workers with inexperience around domiciliary ventilation were reported to negatively influence care provision in instances where the input of family carers was minimised or disregarded.(231) Family carers may find it difficult to hand over care responsibility to homecare workers.(227) Dale et al. reported that family carers would feel a sense of duty to train and supervise inexperienced homecare workers, increasing the burden on these family carers.(231) Furthermore, if there is a high turnover of homecare workers, then the patient-homecare worker relationship may need to be repeatedly re-established to form trust, which may be very challenging.(218)

Safeguarding is defined as putting measures in place in services to reduce the risk of harm, to promote people’s human rights and their health and wellbeing, and to empower people to protect themselves. Safeguarding is fundamental to high quality health and social care. Given the unregulated nature of the home environment and the vulnerability of the population, it is critical that safeguarding measures are put in place to protect the patient.(232) A qualitative study found that patients availing of domiciliary ventilation needed complete trust in their nurse in order to feel safe.(233) The study found that the interpersonal nurse–patient relationship was key in promoting patients’ feeling safe.

Communication needs

In addition, it is essential that homecare staff are knowledgeable of the issues experienced by these patients beyond that of their medical and daily care needs, such as issues relating to ventilator-supported communication.(222) It has been reported that there is a lack of knowledge around the communication experience of these patients, particularly difficulties they may have with speech or voice production.(222) These limits in communication capacity require patience from those providing care, so that they listen and pay attention to verbal and non-verbal signals from the patient and ensure they feel understood. This is necessary to provide comforting care to these patients.(117) Therefore, it is important that hospital and community services work together with the family in the appropriate planning of home adaptations prior to discharge, ensuring that homecare staff are trained and prepared for the complexities of working in private home spaces.(226) Collaboration between all parties at this stage prior to discharge is important to ensure appropriate boundaries are established to help balance the provision of complex care in the home with respect for the patient and other family members.(227)

It is important to note that these are examples or potential sources of conflict that could occur in a given context that have been reported on in the literature. It is not to say that conflicts such as these are unavoidable elements of domiciliary ventilation for these patients. The purpose of presenting these examples is to raise awareness and shed light on such issues with a view to pre-empting potential conflicts.

7.2.3.3. Access to healthcare

Reliable and timely access to health advice and support is very important to patients with SCI receiving domiciliary ventilation.(117, 224) Trust between the patient, homecare workers and family has been reported to be central to ensuring a positive relationship and achieving comforting care that makes the patient feel safe.(117, 218) This can be facilitated by having known and trusted advisers (that is, professionals who can provide advice or assistance to the patient and with whom the patient has a trusting relationship), receiving support from an interprofessional team, and access to 24 hour emergency service.(117)

Delays in accessing or receiving healthcare can negatively impact on the patient and family. Being cared for in the home environment, outside of a hospital setting, may elevate feelings of concern when health problems are encountered. For example, patients on long term ventilation are particularly prone to infections,(234) with respiratory infections being the leading cause of death for patients with both acute and chronic SCI.(235) In Ireland, the Community Intervention Team (CIT) can deliver intravenous (IV) antibiotics to patients at home to treat infections. However, SCI patients availing of domiciliary ventilation services have reported that the initiation of these IV antibiotics can take several days as these antibiotics are not routinely available from local hospitals and must be obtained by the CIT from the HSE central pharmacy. These delays can cause anxiety for the patients and their family given the uncertain health impact that this may have on the patient.(172) This also emphasises the importance of trusted relationships between patients and those providing care so that they feel safe during such instances.

Given the vulnerability of these patients, and the complexity of care involved, it is important that patients and family know who to contact, and when, during different types of situations.(138) Family carers can often feel anxious and stressed when emergencies arise (for example, accidental decannulation, obstructed tracheostomy tube), due to their lack of familiarity with managing such scenarios.(236) Different medical issues may also arise as the patient gets older and so continuous training is important for the patient and their family.(138, 236) The ability to access expert advice and information on an ongoing basis is key to providing assurances to the patient and family.(237)

The quality and consistency of care provided to patients will directly impact their quality of life. However, there is currently a staffing issue with regards to homecare services in Ireland, which is making it challenging to ensure continuity of care.(183) HIQA has previously found that a "poor quality or unfulfilled service" was the greatest risk to people receiving homecare services. Continuity issues were commonly reported in homecare services, such as frequent homecare staff changes or absence, missed homecare calls and an inability to provide care at the right time.(183)(238) According to SCI patients availing of domiciliary ventilation in Ireland, high homecare staff turnover and shortages of trained staff is a significant issue.(172) Competency and continuity of homecare staff are important for the patient and their family.(237)

Such factors may negatively impact important considerations such as trust and feeling safe at home, and the extent to which the patient feels that their individual needs are sensitively being considered.(218) The published literature emphasises the importance of person-centred care by involving the patient in decision-making and tailoring this care to an individual's needs.(117, 218, 224) Issues such as high staff turnover and training insufficiencies represent serious risks to achieving sustained high quality care for these patients. To overcome the various challenges associated with their life-changing situations, patients need ongoing support from competent homecare workers with an understanding and knowledge of the issues related to living with a spinal cord injury and requiring ventilator care.(222)

Advance healthcare directives

An advance healthcare directive is a document which sets out a person's instructions in relation to the healthcare treatments they wish to refuse, or would like to request, in the future when they no longer have capacity to do so.(239) The availability of advance healthcare directives is an important consideration for patients, as detailed in Chapter 4. Of note, since 26 April 2023, under the Assisted Decision-Making (Capacity) Act 2015, there is now a statutory provision for the making and recognition of Advance Healthcare Directives.(240) Compassionate counselling and support services should be made available so that patients and care professionals can establish healthcare directives to prepare for future potential outcomes, such as crisis events or end-of-life care.(139) Such discussions should not be a one-off event, but rather an ongoing aspect of care, so that the directives can be updated to reflect the wishes and opinions of the patient over time.

7.2.3.4. Integration into society

The provision of domiciliary ventilation care to patients permits a more normal everyday life compared to that experienced in an institutionalised or hospital setting.(218) This cohort of patients are completely dependent on others to manage their considerable medical and daily care needs. As a consequence, communicating needs and desires is paramount for these patients and provides a way to gain control and feel seen.(222, 228) The combination of optimising the technology to support the patient's ability to communicate and the willingness of the carers to support and listen will determine communicative performance of the patient.(222) From the perspective of those providing care, having established routines and a positive relationship with the patient can help to deliver care that meets the expressed needs and preferences of the patient.(228) Verbalising their needs can be exhausting for the patient, depending on the health and respiratory state. As such, the responsiveness of the helper and being familiar with the patient can reduce the burden on the patient in communicating their needs.(228)

With the correct supports in place, these patients can be enabled to participate to the greatest possible extent in social and community life.(172, 241) The loss of mobility and high care needs of these patients present significant barriers to daily life. In addition to modifications to the home, transportation barriers exist, as these patients will require wheelchair accessible vehicles. Typically, trips must be planned in advance, due to the requirement to be accompanied by a homecare worker at all times.(172, 219) Poor quality infrastructure in the community, such as poorly maintained roads and pathways, and reduced accessibility, are additional environmental barriers for patients to manage. Poor accessibility can be experienced as a restriction by society toward the patient.(228) Similarly, it may not be possible or practical for patients to visit friends, even those living nearby, because of the practical challenges. In contrast, technology and the internet can enable patients to participate in social life, since patients can experience an active and outgoing life on the internet.(228) Community social support groups may also be beneficial in providing help and support to patients to overcome social isolation and reduce stigma associated with various aspects of their care.(221) It should be noted that while a patient may experience some mobility barriers in the community, this should be contrasted with the significant barriers faced by long-term residence in a hospital setting.

7.2.3.5. Carer burden

The substantial and sudden change to the life situation of the patient with a high cervical SCI requiring ventilation support impacts the entire family. Family members often take on the responsibility of being a carer to the patient, while the provision of domiciliary ventilation care for patients at home can considerably change the home environment.(117, 227) Carer burden refers to the physical, emotional, mental, practical and financial challenges these family members experience while providing care.(117, 227, 229, 242, 243) The HSE integrated care pathway for the management of spinal cord injury in Ireland highlights the significant requirement for the training and education of families and carers.(2) However, it has been noted in the international literature that healthcare providers often fail to consider carer burden as the focus is instead on providing the carer with specific knowledge and practical skills to manage the patient.(242) As outlined in Chapter 3, there is strong evidence of carer burden among family carers of patients with SCI.(106) For example, in a 2018 Italian study, the authors found that increased carer burden and the level of SCI dependency was significantly correlated ($p < 0.01$) with greater need of psychological support and lower perceived health and quality of life.(242) This is particularly relevant for this HTA given the patients under consideration are fully dependent on others for all aspects of their physical care. For family carers, their caring responsibility determines their daily life and is associated with high workload, bureaucracy, lack of flexibility, change in social life, financial burden, and restrictive housing conditions.(117, 172, 229, 242, 243) Despite its challenges, research indicates that family carers still report a reasonably high level of satisfaction (~80%) with their decision to care for their loved ones availing of domiciliary ventilation.(106) Parental carers in particular appear to have a strong sense of satisfaction with their decision, possibly reflecting their desire to protect their child at all costs.(106)

The international literature outlines approaches to lessen the carer burden experienced by family members. Greater provision of paid care, professional support, and respite care have been recommended to alleviate carer burden and make care provision more sustainable.(229) Paid care can help to reduce the burden of various tasks for the carer and represents an important support to family carers.(242) It is important to note that paid care should not be seen as a replacement to family care. Many daily care tasks, such as respiratory care and bladder management, require collaboration or alternation between homecare workers and family carers. Moreover, family carers often provide complex care for long time spans as these patients require life-long care. As such, homecare workers should acknowledge the expertise of family carers, such as their knowledge of the health needs and relationship with the patient.(242) Respectful care and collaboration can empower family carers; the absence of such collaboration and support can however contribute further to the physical and emotional strain experienced by carers.(117, 172)

7.3. Ethical considerations

7.3.1. Introduction

Ethics refers to the understanding and study of morality. The term 'morality' encompasses beliefs, standards of conduct, principles and rules which may guide personal, professional and institutional behaviour. Morals are defined as standards that are widely shared and that form some degree of social consensus.(190) An ethical analysis as part of a HTA involves exploring the possible consequences of implementing or *not* implementing the health technology under consideration. The consequences are considered in relation to both the prevailing societal values and also in relation to the norms and values that the technology itself constructs when it is implemented. The moral value that societies attribute to the consequences of implementing a technology is affected by socio-political, cultural, legal, religious and economic differences.(190)

An ethical analysis was undertaken to consider prevalent social and moral norms and values relevant to the national delivery of care for adults (including young adults ≥ 16 years old) with SCI who require permanent invasive mechanical ventilation and in whom discharge home is deemed clinically appropriate. It is important to remember that the HSE decision that the HTA is intended to inform is *not* whether domiciliary ventilation should be provided to this population, but rather to advise on *how* such a service is made available. While provision of domiciliary ventilation services for patients with SCI is already included in the HSE's integrated care pathway and is currently organised at a local level on an individual patient basis, it is currently ad hoc and there are many challenges in enabling individuals to be discharged home, as discussed in Chapter 6. Many of these individuals will remain in hospital for a prolonged period of time, often several years, even though they are considered *clinically* suitable for home discharge. To illustrate the excessive length of time that patients can wait before going home, a case study is presented. This particular patient spent almost a full year in the National Spinal Injuries Unit (NSIU) in the

Mater Hospital, Dublin before being transferred to Stoke Mandeville spinal unit in the UK to receive ventilation in a rehabilitation hospital (as this service was not available in Ireland at the time). After 304 days in Stoke Mandeville, the patient was transferred back to the Mater Hospital, where they spent another 165 days before being transferred to an ICU in a regional acute hospital. The patient spent over a year in the regional hospital before they eventually got home. In total the patient spent over three years in various hospitals before returning home.(244)

While the status quo in Ireland is that these individuals may eventually avail of domiciliary invasive ventilation, for the purpose of this ethical analysis, the scenario whereby these individuals remain in hospital for an indefinite period of time was considered as the appropriate comparator. It was considered important to explore the ethical implications of delivering this complex care to this vulnerable population in a home setting compared with a hospital setting, given the inherently different nature of these settings and the potential consequences of shifting care from hospitals to the home setting.

An ethics workshop was undertaken whereby members of HIQA's HTA Directorate discussed these issues. The findings of this workshop were supplemented by a review of relevant literature and discussions with various EAG members. The analysis of this information was guided by the ethical domain of the EUnetHTA HTA Core Model Version 3.0, which considers six topics covering 20 issues.(190) The four principles approach to biomedical ethics (otherwise known as principlism) was used as a framework for this ethical analysis.(245) Principlism comprises four key moral principles:

1. Respect for autonomy (that is, respecting the decision-making capacities of an autonomous person)
2. Non-maleficence (that is, avoidance of harm)
3. Beneficence (that is, providing benefits and balancing benefits against risks and costs)
4. Justice (that is, fairness in the distribution of benefits, risks and costs).

7.3.2. Benefit-harm balance

Before investing (or disinvesting) in a health technology, it is important to consider the balance of benefits and harms. In an ideal scenario, the patient, family, health and social care professionals, healthcare system and wider society would all obtain benefit from a technology without any harms. However, in reality there is always a risk of harm with any technology and this needs to be carefully balanced against the potential for benefit.(190) In essence, the risks of harm are only ethically acceptable if they are outweighed by potential benefits.(245) However, while there are clear distinctions between the moral duty to refrain from knowingly causing harm and the duty to provide benefit, these are not always easily separated. In other words, sometimes the benefit can only be realised when accompanied by the risk of harm.(246)

7.3.2.1. Burden of disease and epidemiology

The burden of disease and epidemiology for this condition (that is, adults with high cervical SCI (C1-3) who require permanent mechanical invasive ventilation) is described in detail in Chapter 2. In brief, high cervical SCI can be catastrophic for an individual. These injuries can result in permanent tetraplegia and respiratory failure among a range of other life-changing consequences, rendering these individuals completely dependent on others for many aspects of their physical care.(15, 89) Permanent mechanical invasive ventilation is required in a minority (approximately 3% to 5%) of patients with tetraplegia.(28) This patient cohort requiring permanent mechanical invasive ventilation predominantly comprises young males. Though the international literature reports trends of increasing age among patients with SCI overall,(58) the provision of domiciliary invasive ventilation services may be less likely in older adults given the complexity of care requirements.(143) Life expectancy is substantially reduced in comparison with the general population, however patients with SCI who are ventilator-dependent can still live for 10-20 years post injury, depending on the age of injury onset.(6, 46)

Of note, this is a rare event. While there is no national registry, expert opinion is that there are currently between five and ten of these patients receiving domiciliary ventilation in Ireland.(143) It was previously estimated, based on data extrapolated from other jurisdictions, that an additional one to two patients per year will fall into this category,(2) however the experience to-date has been that this number may be smaller (approximately one new patient every year or every other year).(143) Measuring quality of life in SCI patients presents challenges. Studies have had variable results, but a common finding is that patients are glad to be alive and that they often experience meaningful and fulfilling lives post-injury.(247) These findings challenge a commonly held view among the general public that death may be preferable to life in this condition.(247)

7.3.2.2. Potential benefits of implementing the technology

There are many potential benefits both for the patient and others (for example, family members, healthcare system and the wider community) associated with the implementation of a national provision of care for this population.

One of the key benefits is that it would respect the patient’s preference to return home, hence allowing them to be closer to family and friends, and enabling the patient to participate in society.(248) By providing the patient with all the appropriate supports, this may enable them to return to education or work, and to undertake every day social and leisure activities (for example, attend the cinema or meet friends). There is also some evidence, though limited, that provision of domiciliary ventilation to these patients may improve their health related quality of life (HRQL), particularly within the area of mental health.(106) Intensive care units (ICU) (or high dependency units (HDU)), where these patients would most likely be cared for within a hospital setting, are known to be loud, highly stimulating, busy clinical areas which also present infection risks. ICU patients and family members often report negative experiences of these settings.(249) Residential care may not present an acceptable alternative for many of these patients given that those normally resident in these settings are typically much older. Therefore the needs and preferences of these younger patients with a disability may not be met in a residential care setting either.(216) Long term stay in hospital or residential care is far from ideal and so the safe provision of care with all the necessary supports in a familiar home environment may be the most ethically appropriate and acceptable for these patients and their families.

There are also many benefits to family members for having their loved one cared for in the home. The family would get to spend more time with their loved one, allowing the patient to see their children grow up, for example, hence supporting family cohesion.(250) It would also reduce the time and money spent on travelling to and from the hospital to see their loved one. This may be a particular issue if families are reliant on public transport and or do not live close to the hospital.(251) While substantial changes are necessary to accommodate the safe care of the patient in the home, it is possible that a reasonably normal and active life can resume.(237)

Shifting of care out of hospitals and into the community also potentially frees up scarce ICU/HDU beds, allowing critical care to be provided to other patients.(215) This may also result in cost savings for the public acute hospital system as the substantial long term costs of providing care for these patients no longer falls on the acute hospital budget, however there may be a net budget impact on the HSE overall as the burden now falls predominantly on community budgets supplemented by out-of-pocket expenditure by the patient and the family (see Chapter 5).(252) There may also be benefits to healthcare staff and managers in terms of improved efficiencies if a centralised budget and streamlined system of organising home care packages is implemented.(2) It is also important to consider that there may also be financial benefits to private home care providers and companies providing and servicing equipment, should more patients be able to avail of domiciliary invasive ventilation services.

7.3.2.3. Potential harms of implementing the technology

The potential benefits must be balanced against the potential harms. There are potentially significant harms associated with providing complex care in an unregulated home environment. As described in Chapter 3, provision of care to patients with high cervical SCI who are invasively ventilated is complex, and requires round-the-clock care. By providing this care outside of a hospital setting this means that access to specialist medical teams and equipment, as might be required in various emergencies (for example, cardiac arrest), is limited, which may result in delays to care and subsequent negative outcomes for these patients.(253) This is particularly problematic as the specialised transport needed to transfer critically ill ventilated patients may not be available 24/7.(254)

There is a reliance on a 24/7 service which may be challenging in times of severe weather; gaps in service provision could cause patient harm. While technical issues associated with domiciliary invasive ventilation are rare (for example, ventilator failure), they can have fatal outcomes if not rectified immediately. There is also the potential for medical emergencies to occur in these individuals (for example, autonomic dysreflexia, etc.). It is critical that homecare staff are present and competent to deal with such eventualities.(137) If not, then there is a significant risk of harm, including death.(80) There is evidence from Switzerland of patients with SCI and their families relocating to reside closer to specialised SCI centres, in order to maintain good access to the appropriate healthcare services.(255) The patient is entirely dependent on the skill and knowledge of the homecare staff to survive and these may be influenced by the level of training and support provided by the homecare provider and others. Given the unregulated nature of the home environment and the vulnerability of the population, safeguarding issues (for example, physical abuse, emotional abuse, sexual abuse, neglect of the person and financial abuse) may also present a risk of harm to the patient.(232) It is also possible that infection prevention and control (IPC) measures may be more lax in a home environment putting the patient at risk of acquiring infections.(256)

It is also important to consider that sometimes home placements are unsuccessful for a variety of factors, necessitating a readmission to hospital or to a residential care facility with potentially negative ramifications on the patient's mental health. In other words, the provision of this service may increase expectations that cannot realistically be met for some individuals.(257) Conversely if there was an expectation to avail of this service once clinically suitable, it is plausible that there might be a pressure on patients to vacate their hospital bed, even if they felt safer in a hospital environment.(258)

There are also risks of harm towards the family given the substantial additional strain (financial, psychosocial and physical) that caregiving for an individual with complex care needs presents.(198) While a suitably funded package by the HSE should cover round-the-clock care by at least one homecare staff member, there is still an expectation that family members would play a pivotal role in the day-to-day care of the patient. The 24/7 reliance on trained homecare staff presents significant challenges to the family in that unavailability of staff can lead to pressure on families to provide the required care. The worry that there may not be continuous care provision may be a particular source of stress and anxiety for families.(250) Provision of domiciliary invasive ventilation requires substantial upfront costs, for example home modifications and acquisition of an adapted vehicle. While grants are available in Ireland to support families in this regard,(195) there are limits to these supports, and so substantial out-of-pocket expenditure by the family is often required to plug this gap.(252) Caregiving is also associated with negative impacts on physical and mental health.(198) The Irish Health Survey 2019 found that 19% of carers reported experiencing depression compared with 13% of non-carers.(259) There is substantial emotional burden associated with this intensity of caregiving, with many personal sacrifices having to be made by family members so that their loved ones can live a good life.(248) As the home becomes like a hospital, the family members may perceive it as an invasion of their personal space.(227) Family members may not be comfortable with the constant presence of unfamiliar homecare staff 24/7.

Caregiving is generally considered a gendered role; the Irish Health Survey 2019 found that more females (14%) than males (11%) are carers.(259) Given that carers often have to leave full time employment to assume unpaid caring roles, with a knock-on negative impact on income and career development, this may potentially further exacerbate gender inequality.(260) In light of the full time nature of the caring in the home, there may be no or limited possibility of respite for the family. It is important to question in whose best interest it is to provide domiciliary invasive ventilation.(248) While it may be of benefit to the patient to move home, it is possible that this may be to the detriment of the family.

There could also be harms to the homecare staff in terms of perceptions of increased isolation, stress and burn-out associated with providing high intensity care in someone else's home.(261) There may also be a risk of harm to homecare staff given that there is evidence from the literature that they are commonly subjected to aggression and violence when working in peoples' homes.(262) Finally, there may be harms for the healthcare system, specifically in relation to the opportunity cost associated with providing this resource-intense service. Given the substantial funding and resources required to deliver a package of care to these individuals, estimated at €3.4 million per patient over five years (Chapter 5), this may deplete local budgets meaning that other patients in the community may be deprived of an important service. There may also be a litigation risk should an adverse event occurs in the community;(263) however, the risk may be mitigated if a clearer governance structure is implemented as part of a new provision of care.

7.3.2.4. Potential benefits and harms of the status quo

It is important to consider the potential benefits and harms of the status quo, that is patients remaining in hospital for a prolonged period of time before eventually being discharged home. While patients eventually get home, the process is ad-hoc and there are differences in the availability and quality of service provision depending on where the patient lives.(2) The main benefit of the current ad-hoc approach is that there is onsite access to a suite of healthcare professionals who are experts in managing SCI (for example, physiotherapy, occupational therapy, rehabilitation medicine, nursing etc.). However, there are many potential harms with the current approach, such as the psychological distress for the patient and the family due to the uncertainty of the entire process and the difference in service availability depending on the area.(189) Given the length of time it can take to get the right supports in place, and the strain this can put on families, there can be deterioration in personal relationships.(189) Patients often spend years in ICU beds, and so these beds are unavailable for other patients.(2)

7.3.3. Respect for autonomy

Respect for autonomy implies *"acknowledging that autonomous agents are entitled to hold their own viewpoints, are free to make choices, and act voluntarily according to their values, beliefs and preferences"*.(245) By respecting autonomy a clinician should acknowledge the agency of the patient to exercise the right over all clinical decisions relating to their care without undue interference or influence, thus enabling self-determination. An important ethical dilemma arises regarding the limits to which a clinician can accept "non-interference" in a patient's decision-making, particularly when there is a known risk of harm associated with that decision.(264)

7.3.3.1. Patient autonomy

Patients with long term care needs often (but not always) express a strong desire to receive care in the comfort of their own home surrounded by their loved ones.(265) This is also the case for patients with high cervical SCI who are ventilated.(172) Provision of a national delivery of care for these patients in whom discharge home is deemed clinically appropriate should therefore be viewed as supportive of patient autonomy where a clear preference to move home is expressed. Patient autonomy in this regard is supported by the Assisted Decision-Making (Capacity) Act 2015. Some of these patients may also sustain brain trauma and so there may be a degree of cognitive impairment that needs to be taken into consideration.(25) At its core, this Act provides for the patient's right of autonomy and self-determination to be respected, and is used when the patient has decision-making challenges.(240) This Act aims to assist in complying with human-rights obligations contained in the Constitution of Ireland, the European Convention on Human Rights, and the United Nations Convention on the Rights of Persons with Disabilities.(240)

It is important that the patient and their family have been adequately appraised of the benefits and risks of receiving homecare, and that comprehensive education has been provided to alert them to the daily realities of the complexity and intensity of homecare required, so that true informed consent can be given by the patient.(79) It is possible that more flexible discharge options (for example, shared care between the home and hospital), rather than a binary option of remaining in hospital versus being discharged home, may offer choices that are more appropriate for the needs of the patient. While the Assisted Decision-Making (Capacity) Act works to support the patient's right to make decisions about where they receive care, it is important to consider that this may conflict with family members' rights, who may have safety concerns and reservations regarding the psychosocial and financial impact of taking on the burden of home care.(229) Conversely, if a patient feels safer in a hospital setting and expresses a desire to remain there despite being deemed clinically suitable for home discharge while the hospital bed is needed for another patient, this might result in another conflict of rights.(266) Denson et al. refer to the balance of safety and autonomy with regards to discharge planning and acknowledge that "despite good intentions and a shared focus on the best interests of the [patient], stakeholders may hold very different values about good outcomes and how to decide them."(267)

7.3.3.2. Professional autonomy

Respect for autonomy also extends to healthcare professionals. In a systematic review, Lamiani et al. found that low levels of autonomy in clinical decision-making was associated with moral distress, that is, the "negative experience when one knows the right thing to do, but institutional constraints make it nearly impossible to pursue the right course of action."(268)

Provision of homecare to these patients, with clear policies and procedures within a strong governance framework, can be considered as supportive of professional autonomy. In this situation, public health nurses can be enabled to lead on the care of these patients in the community, working closely with GPs and health and social care professional colleagues in primary care with links to secondary and tertiary care as appropriate. Registered nurses working within their Scope of Nursing and Midwifery Practice Framework can delegate care in the home setting to unregulated healthcare workers who are deemed competent to undertake these tasks.(184) The delegator is accountable for ensuring that the delegated role or activity is appropriate to the level of competence of the unregulated healthcare workers to perform. Some countries, such as the UK, New Zealand and the Netherlands, permit homecare workers, other than nurses, who are deemed competent, to provide care to ventilated patients with SCI at home, instead of 24/7 care from a nurse (Chapter 4).(126, 138) However, in the Irish context this could potentially infringe on the professional autonomy of the registered nurse with subsequent uncertainty regarding accountability should an adverse event occur.(184)

There may also be potential challenges to professional autonomy in light of the provision of complex care in a home setting. Heggestad et al. describe how homecare workers often have to deal with situations where patients' or families' views and wishes conflict with their own professional values and norms and what they think is the best care.(230) This can be particularly challenging as the homecare worker is both a professional and a guest in the patient's home, and so it may be perceived that there is an asymmetrical relationship whereby the patient has a greater say in their care than they normally would in a healthcare setting. The boundary between professional relationship and friendship could also become blurred over time which may present further challenges to the homecare worker when making independent clinical decisions.(230) Importantly, establishing trust is particularly important in balancing homecare workers' role between professionalism, privacy, and friendship, and this may be hampered in the context of substantial staff turnover.(230)

7.3.3.3. Respect for persons

Respect for persons relates to the concept of autonomy and comprises two separate moral requirements: the requirement to acknowledge autonomy and the requirement to protect those with diminished autonomy.(269) Careful consideration is required in relation to the potential impact of the implementation or use of the technology on human dignity; moral, religious or cultural integrity; and privacy.(190)

Implementation of a systematic provision of care would enable these patients to successfully leave the hospital and return home with an appropriately funded care package, in a timely manner. A nationally coordinated and well-implemented system would provide clarity to the patient and family and would also prevent the patient remaining in a hospital indefinitely, which may be undignified and unacceptable to them.(2) Patients may be particularly adversely affected by the lack of privacy in hospital settings, compounding the emotional turmoil associated with the life-changing SCI. Patients need time to come to terms with their injuries, with some drawing parallels to grieving the death of a loved one.(270) In a qualitative study, Samuel et al. reported that SCI patients often described feelings of "amplification of vulnerability" in hospital settings during rehabilitation.(271) In this study, patients reported experiencing significant violations of their privacy in these environments (for example, staff switching lights on and off and opening curtains without warning). This may particularly be the case for female SCI patients in the context of mixed sex wards.(271) Sometimes SCI patients end up in residential care settings. In these institutional settings, it may not be dignified for SCI patients, who are generally young and without cognitive impairment, to share living spaces with older adults, many of whom may have dementia.(216) Young patients with SCI may not have their unique needs (medical, social, emotional, sexual etc.) met in residential care settings and may suffer as a consequence.(272)

Delivery of high intensity care in a home environment by homecare staff might also be perceived as an invasion of privacy for the patient and the family. The home effectively becomes a hospital, with staff onsite at all times, inevitably disrupting the normal family routines.(229) The house needs to be modified to facilitate equipment and access and to provide breakrooms for staff. Medical emergencies may occur and may be upsetting for family members to witness. A qualitative study by Dickson et al. described how some SCI patients express the need for clear boundaries between them and the homecare team so as to preserve some sense of privacy and independence in their own home. The authors also reported how some family carers expressed how their home no longer felt like their home and that it sometimes felt “threatened” by the presence of the homecare staff.(227) This tension can sometimes lead to conflict between family members and homecare staff.(261) Moving to home, while still the preferred option, may also create dignity issues if family members are involved in the personal care (for example, bathing, bowel and bladder care) of the patient, who may be their spouse or partner, for example. Neurogenic bowel and bladder dysfunction, which may result in accidents, may be a particular source of embarrassment for many such patients.(273)

Ventilator-dependent patients with SCI have a reduced life expectancy compared with the general public(46) and so due consideration with regards to end of life care is important for this population.(139) Patients with SCI should be treated with dignity and respect at all stages throughout their care, but particularly towards the end of life. Decisions regarding “change of therapeutic goals” during palliative care should occur in conjunction with the patient, the doctor and the nurse, ideally in agreement with an advanced healthcare directive as per the Assisted Decision-Making (Capacity) Act 2015.(139, 240) However, ethical issues arise in the context of medical assistance in dying (MAID) (which includes both assisted suicide and euthanasia),(274) which is distinct from palliative care approaches and is in place in some countries, such as Canada, Switzerland, the Netherlands, Belgium, Luxembourg and Spain under certain strict circumstances.(139, 274) Provision of MAID may infringe on patient’s and professional’s moral, religious or cultural integrity(275) and is currently not legally permissible in Ireland.(276)

7.3.4. Justice and equity

The principle of justice refers to the requirement for people to be treated fairly and equitably both in terms of the distribution of benefits and burdens, as well as the recognition of people's rights and responsibilities.(245) In relation to population level healthcare decisions, opportunity costs are an essential factor to consider. Consideration needs to be given to the total size of these opportunity costs as well as understand who will bear the consequences of these costs.(277)

From an ethical perspective, cost-effectiveness accounts for opportunity costs to unknown patients in society relative to the benefits to the known patients who use the technology.(277) It was not possible to determine the cost-effectiveness of this technology given the challenges in quantifying the associated effectiveness with such few patients, and also due to the fact that domiciliary ventilation services are already provided to these patients, though in an ad-hoc manner. Importantly, the cost for a national provision of care is substantial with a total budget impact of €3.4 million per patient over five years (Chapter 5), though at a population level the budget impact may be low given the rarity of its occurrence. It would require a significant re-allocation of specialist human resources, funding and training to provide care for a very small number of patients with a potential knock-on impact on other patients who may not be able to avail of services in the community.(190) Therefore, at a population level, provision of this national service with a ring-fenced budget and access to the required resources is likely not equitable from a purely utilitarian perspective (that is, the greatest benefit for the greatest number of people).(278) However, given the current geographical disparities in access to this services (for example, local budget holder refusal, limited availability of homecare staff, rural area located too far from specialist services, limited availability of suitable housing etc.),(189) provision of a systematic nationwide service may improve equity of access at the patient level, despite the high costs. In addition, from a 'justice as fairness' perspective as described by the philosopher John Rawls, it could be argued that *"distribution should be based on a reasonable expectation of advantage for all, but also to the greatest benefit of the least advantaged in society."*(279) Furthermore, fundraising and the charitable support of the wider community have been noted to be an important source of financing for affected families in Ireland, thus it could be argued that families require a greater level of financial support than is currently provided.(172) On this ethical basis it may be justified to provide substantial funding and resources to these few patients given their significant levels of disability.(279) Dywbwik et al. discuss the ethical challenges when considering the financial implications in the decision-making process regarding funding of a domiciliary ventilation service.(248) The authors describe the difficulties clinicians face balancing patients' rights to accessing this service and the associated benefits for these patients, with the resource consequences for other patient groups.(248)

There are other patient groups that can avail of domiciliary ventilation services, such as patients with chronic obstructive pulmonary disease (COPD), amyotrophic lateral sclerosis (ALS) which is a sub-set of motor neurone disease, and obesity hypoventilation syndrome (OHS).(134) However, it is important to note that the care needs for all of these patient groups differ substantially from those of SCI patients. Firstly, for many of these other patient groups, the need for mechanical ventilation arises from progressive clinical deterioration over time and so the decision to commence a patient on invasive ventilation is elective and based on a risk-benefit discussion.(134) This is in contrast to traumatic, high cervical SCI, the main focus of this HTA, where the need is immediate and life sustaining. In addition, non-invasive ventilation (NIV) which is associated with fewer care needs and complications than invasive ventilation (for example, no tracheostomy required) is more commonly used in these other patient groups.(91) Of note, domiciliary NIV has been available in Ireland for these conditions since at least the 1980s.(104) Secondly, the tetraplegia that occurs with high cervical SCI presents additional care challenges in relation to mobility, bowel, bladder and skin issues etc., which are not experienced by these other patient groups, with the possible exception of patients with ALS(280) (though life expectancy is significantly reduced in ALS).(281) However, as outlined in Chapter 4, there are many commonalities in recommendations regarding the provision of care to patients availing of domiciliary ventilation, regardless of the underlying diagnosis. In addition, there are also other patients with lower or incomplete SCI who may only require NIV but may still have very high care needs, similar to those with higher complete SCI.(133) Therefore, it could be argued that provision of a dedicated nationwide service exclusively to invasively ventilated patients with C1-3 SCI may not be equitable to other patient groups who may have the potential to benefit from access to this service. Conversely, it could be argued that the needs of these particular patients with high cervical SCI are the most pressing and are not being met by the current system, and so it is fair to prioritise this cohort in the first instance.

From a distributive justice/equity of care perspective, there will be a subset of these patients who face greater practical challenges to returning home. For example, single parents, and those who live alone or in rented accommodation. To be equitable, the national provision of care should also aim to enable these patients to return home despite the obstacles.(282)

7.3.4.1. Legislation

According to HIQA’s Guidance on a Human Rights-based Approach in Health and Social Care services: *"Human rights are the basic rights and freedoms that all people should enjoy; including people using services and staff. Human rights are about people being treated with fairness, respect, equality and dignity; having a say over* *their lives and participating as fully as possible in decisions about their care and support. There is a duty on the State and on health and social care providers to uphold the human rights of people using services and staff.'*(283) The United Nations declare that human rights are universal, inalienable, interrelated, interdependent and indivisible.(284) Respect for human rights is both a professional obligation implicit within national standards and is also enshrined in law (for example, the Irish Constitution 1937, the European Convention on Human Rights Act 2003, the Charter of Fundamental Rights of the European Union 2000, the Equal Status Acts 2000-2015, the Irish Human Rights and Equality Commission Act 2014). Important changes in Irish law include the ratification of the United Nations Convention on the Rights of Persons with Disabilities (UNCRPD) 2006, and the introduction of the Assisted Decision-Making (Capacity) Act 2015.(283) Of note, under the Assisted Decision-Making (Capacity) Act 2015, there is statutory provision for the making and recognition of Advance Healthcare Directives.(240) This provision is particularly important for patients with SCI who require permanent mechanical invasive ventilation, given their reduced life expectancy.(46)

Implementation of a systematic provision of care to patients with high cervical SCI, that enables patients to return home with all of the required supports and equipment, in line with their wishes, should be viewed as respecting the human rights of the patient. This may also realise one of the key ambitions of Sláintecare reforms which is the shift of care out of acute hospitals into the community and closer to a person's home, where safe to do so.(8) However, as discussed above, the delivery of this high intensity care at home may impact on the privacy of family members and it could be argued that this might infringe on their right to privacy.

Homecare in Ireland is currently unregulated. This means that HIQA (or any independent authority) does not currently have a statutory basis for inspecting homecare services. For this reason, there are concerns regarding the quality and safety of homecare services in Ireland. Of note, homecare providers in Ireland have cited difficulties with staffing, timing of homecare, funding, availability of homecare and the assessment process as significant challenges at present.(183) In addition there is no statutory scheme in place for homecare which means that it is a discretionary service and so the State currently has no obligation to provide care at home (unlike residential care where there is a statutory scheme).(217) In its position paper, HIQA has stated that it *"firmly believes that homecare should be person-centred, focused on quality, integrated, needs led, and inclusive of complex care where age or disability status is not a barrier or gatekeeper to access homecare services. Furthermore, the fundamental principles of a human rights-based approach — in addition to safety and wellbeing, responsiveness and accountability — should underpin the legislation, standards and regulation of this sector.'(217)* In light of impending legislation, and the subsequent development of National Standards in this area, these may impact on the national provision of a domiciliary invasive ventilation service. Consideration should also be given to the potential impact that introduction of a statutory homecare scheme (akin to the Fair Deal Scheme) may have on the personal finances of patients who avail of this service.(285) Currently, funding for these homecare packages does not involve financial contribution from the patient; however, some out-of-pocket expenditure is inevitable (for example, in relation to housing modifications).(205)

Right to discontinue treatment

In the event that a patient decides to discontinue treatment, there may be a lack of clarity about the roles and responsibilities of the person's carers. Under current Irish law, *"if you are a mentally competent adult, you have the right to refuse or discontinue medical treatment even if the inevitable consequence is that you will die. This is different from taking positive measures to end your own life or another person's life."*(286) Given that patients in the context of domiciliary ventilation are likely to be fully dependent on family and homecare workers for their physical requirements, it is important to acknowledge that, while a patient may legally have the right to refuse ventilation, this may result in an ethical dilemma and potential conflict between patient autonomy and the desire to do no harm and, potentially, concerns regarding possible legal ramifications.(287-289) In Ireland, under Section 2 of the Criminal Law (Suicide) Act 1993, anyone who "aids, abets, counsels or procures the suicide of another [person]" can be convicted and imprisoned for up to 14 years.(290) This may be contrasted with the concept of euthanasia. Euthanasia is the *"intentional termination of the life of a person, by another person, in order to relieve the first person's suffering"*, which occurs expressly with the individual's consent. In Irish law, in the context of end of life, there is an important distinction between passive euthanasia (that is, hastening death by withholding a treatment), which is a normal medical practice particularly during end of life care, and active euthanasia (that is, hastening death through active means, for example, by administering a drug), which is currently illegal.(290) The definition of 'end of life' care and the concept of the patient's right to refuse treatment may give rise to ambiguity in the setting of domiciliary ventilation, particularly given that this occurs outside of the typical healthcare setting. As outlined in Chapter 4, withdrawal of home mechanical ventilation is discussed in the German guidance document, which considers the circumstances whereby termination of mechanical ventilation is legally and ethically permissible in Germany.(79, 139) Withdrawal of mechanical ventilation therapy is not specifically addressed in any of the other included guidance documents as part of Chapter 4. Development of ethical guidance, in the context of domiciliary ventilation that occurs outside of healthcare settings, may provide clarity to patients, families, homecare staff and other healthcare professionals.

Furthermore, it is relevant to note that the Dying with Dignity Bill 2020, a Private Member's Bill, which aims to *"make provision for assistance in achieving a dignified and peaceful end of life to qualifying persons and related matters"* is currently at the third stage of Dáil Éireann (also known as Committee Stage).(291) At this stage of the process, the Bill is examined section by section and amendments may be made. There are 11 stages in total for a Bill to be signed into law by the President.(291) The potential impact, if any, of this Bill on the lives of patients with high cervical SCI who are ventilated remains to be seen.

7.4. Discussion

The purpose of this Chapter was to outline the potential patient, social and ethical considerations relating to the national delivery of care for adults with SCI who require permanent invasive mechanical ventilation and in whom discharge home is deemed clinically appropriate. The considerations outlined are framed both in the context of the experiences of the patient and their family, and also through an ethical lens.

Living at home on a ventilator with high cervical SCI is associated with substantial challenges for both the patient and their family. Patients have expressed how these life-changing injuries are catastrophic and can often require a prolonged period of adjustment.(219) The complications arising from the injuries are profound and can have a hugely negative impact on patients requiring round-the-clock care from homecare staff.(2) There are also challenges with the discharge process, living at home, accessing healthcare and integrating into society. Families have expressed their strong desire to provide the best possible care for their loved ones,(106) but often to their own detriment (physically, emotionally and financially).(198) Carer burden is particularly problematic among these family carers, given the high level of dependency.(242) However, these patients have also demonstrated resilience in adapting to their 'new normal' and a willingness to live a fulfilling life despite the obstacles.(223) Provision of the required supports and services for these patients and their families, on an ongoing basis, may alleviate some of the hardship and make the care provision more sustainable.(229)

In terms of respect for autonomy, provision of a national delivery of care, for these patients in whom discharge home is deemed clinically appropriate, should be viewed as supportive of patient autonomy where a clear preference to move home is expressed. However, it is crucial that patients and families are fully informed of the benefits and risks of domiciliary ventilation, and that a true choice of options is provided to the patient that respects the preference of the individual.(79) This would prevent the patient remaining in a hospital indefinitely which may be undignified and unacceptable to them.(2) Autonomy also extends to healthcare professionals working in homecare settings. Provision of homecare to these patients with clear policies and procedures within a strong governance framework can be supportive of professional autonomy, allowing care to be delegated as appropriate, while maintaining high standards of care.(184) In the event that a patient decides to discontinue treatment, there may be a lack of clarity about the roles and responsibilities of the person's carers. Development of ethical guidance with regards to the withdrawal of ventilation therapy, in the context of domiciliary ventilation that occurs outside of healthcare settings, may provide clarity to patients, families, homecare staff and other healthcare professionals.

In terms of the benefit-harm balance, consideration needs to be given to the potential for the benefits of this national provision of care to be outweighed by its associated harms. Some of the key benefits are that it would respect patients' preferences to receive care at home and live within their communities,(248) would likely free up scarce ICU/HDU beds(2) and may improve patients' HRQL, particularly in the mental health domains.(106) However, this needs to be weighed against the increased safety concerns associated with providing complex care at home,(80) the substantial burden on families,(242) and the opportunity cost to other patient groups given the significant budget impact.(248) Policymakers need to carefully weigh up the potential benefits and harms when considering the implementation of a national provision of care for this population.

From the perspective of justice and equity, provision of a systematic nationwide service may improve equity of access for these patients given the current geographical disparities in funding and service provision which are causing particular hardship for patients and families.(189) While there is a substantial budget impact associated with implementation of a national provision of care, the creation of a nationwide ring-fenced budget could prevent local budgets being depleted, allowing funding for other important community services to be preserved.(2) However, provision of complex care to this small number of patients is still associated with a substantial opportunity cost. In particular, given the finite number of specially trained staff available in the community in the context of ongoing staffing issues and waiting lists in the health service,(183) it is important to consider the potential knock-on impact on other patients who may not be able to avail of services in the community.(248) It could also be argued that provision of a dedicated nationwide service exclusively to invasively ventilated patients with C1-3 SCI may not be equitable to other patient groups who may have the potential to benefit from access to this service. Policymakers should consider whether certain aspects of a national provision of care (for example, funding mechanism, education and training, tendering of services) could be of benefit to other patient groups with similar complex care needs who require domiciliary ventilation (for example lower/incomplete SCI, ALS).

It should be noted that while the ethical, patient and social considerations outlined are important and require due diligence in decision making, they are not necessarily unique to Ireland, nor to domiciliary ventilation for SCI specifically. These findings should not preclude the implementation of a national provision of care for this population, but rather provide tangible factors that should be considered during decision-making and accounted for during subsequent implementation.

8. Discussion

8.1. Introduction

Health technology assessment (HTA) is a multidisciplinary process that summarises information about the medical, social, economic, and ethical issues related to the use of a health technology. A HTA is performed in a systematic, transparent, unbiased, and robust manner with the intention of supporting evidence-based decision-making regarding the optimal use of resources in health and social care services.(190)

At the request of the Clinical Lead for the National Clinical Programme for Rehabilitation Medicine in the HSE, and the Programme Manager for the Spinal Cord System of Care Programme in the NRH, HIQA agreed to undertake a HTA on domiciliary invasive ventilation for adults with spinal cord injuries. Work commenced on this HTA in August 2022. This HTA aimed to assess the organisational, budget impact and resource implications associated with, and the social and ethical issues arising from, the provision of care to ventilator-dependent adults with SCI within their own home.

The purpose of this discussion chapter is to summarise the key findings within the Irish context, discuss the implications for policy, and present the strengths and limitations of the assessment overall.

8.2. Summary of key findings

Damage to the spinal cord can result in catastrophic and life-threatening injuries, which can lead to the affected person suffering from significant long-term morbidity and requiring life-long care.(15) The higher up the spinal cord that the injury occurs, the more extensive the range of impairments will generally be. Patients with complete C1-C3 level cervical lesions will experience the most severe level of impairment and tend to require a ventilator and tracheostomy to breathe.(4) In general, permanent ventilator assistance is required for complete SCI at C1-C3 due to diaphragmatic paralysis.(31) Patients with C3-C4 injuries experience diaphragmatic dysfunction, but may retain partial ventilatory function. Weaning from a ventilator may be achieved in patients with C3 injuries, while injuries of C4 and below are usually associated with successful weaning, in the absence of other complications.(15) Given the consequences of complete C1-C3 injuries such as tetraplegia, chronic respiratory failure, neurogenic bowel and bladder dysfunction, pressure injuries etc., a comprehensive package of care involving 24/7 care is required for these patients.

High cervical traumatic SCI are rare in Ireland, and numbers fluctuate year to year. Based on data from the National Spinal Injuries Unit (NSIU) from 2008-2021, the incidence of traumatic C1-C4 SCI was observed to fluctuate from 2.7 to 3.6 cases per million population over the 14 year period, with males being disproportionately affected (82% of cases). There is currently no evidence that the incidence of these types of injuries are increasing, however there is a shift with proportionally more older people presenting with C1-C4 traumatic SCI, with international evidence indicating that this reflects demographic changes, such as people living longer, and associated changes in the aetiology of SCI, such as falls becoming the most common cause of SCI.(63, 64, 70) While older people with traumatic SCI have poorer prognosis(46) and may not be suitable for domiciliary ventilation, there needs to be consideration of short- and long-term management for this cohort of older SCI patients, given the recent trends in the aetiology of SCI in the context of an ageing population.

Not all individuals with high cervical SCI will require permanent mechanical invasive ventilation. Data are sparse but it is estimated that between 3% to 5% of patients with tetraplegia will require permanent mechanical ventilation, with higher likelihood among those with complete C1-C2 injuries.(28) Furthermore, not all ventilator-dependent SCI patients will be suitable for home discharge, particularly if their prognosis is poor or if their care requirements are such that it is not possible to provide the appropriate level of care in the community. It is estimated that there are currently between five and ten adults with SCI in Ireland receiving invasive mechanical ventilation at home, with up to two new patients expected every year, though in some years there are no new patients.(69) While these occurrences are rare, the number of SCI patients availing of domiciliary invasive ventilation services will likely accumulate over time, with subsequent impact on resources and budget. Therefore it is important that a national provision of service is established that is adequately resourced, evidence-based, equitable and patient-centred.

As part of the Irish integrated care pathway for patients with SCI which has been developed by the National Clinical Programme for Rehabilitation Medicine, it is currently standard practice to send ventilator-dependent patients with SCI home with an appropriately funded homecare package when safe to do so.(2) However, discharge planning for these patients remains a huge challenge for a range of reasons, including the burden on families, the current fragmented funding model for community services, and the lack of suitably trained homecare staff in the community. There is currently no standard process for organising these homecare packages, with regional variations observed leading to inequities in timely access/provision to services across the country.(143) As a result, patients with SCI who are ventilator dependent often remain hospitalised for a prolonged period of time, sometimes several years, before being discharged home.(2) These delayed discharges can have a detrimental impact on the individual, their family and carers, management of bed capacity in the NRH and other hospitals, and the wider healthcare system due to the significant cost of treating these patients in hospital.

Patients with high cervical cord injuries requiring invasive mechanical ventilation have complex health care needs, and require substantial support from formal and informal caregivers in a home setting. International guidance documents identified in Chapter 4 discuss the importance of: appropriate preparation and timing of the transition to the home setting; anticipation of present and future requirements in terms of environment and training of those providing care; and anticipation of disease progression.(79, 88, 90, 126-139, 286) A comprehensive, holistic and person-centred approach to care, delivered from a well-coordinated interdisciplinary team, which respects the preferences of the individual, is advocated in these guidance documents. Given the substantial amount of equipment and staff required to provide a 24/7 domiciliary invasive ventilation service, there is clear guidance that adequate funding and acquisition of these resources must be obtained prior to discharge. Clear communication and collaboration across disciplines and settings, with a clear understanding of roles and responsibilities and the overarching governance structure, is recommended throughout. Advanced care planning is also recommended so that the wishes of the individual are understood by all involved in an emergency or end of life situation. It is however important to acknowledge that each patient is unique and has different care needs, and so a 'standard' or 'core' homecare package may be difficult to define. These packages currently need to be developed and provided on an bespoke basis to each patient in areas where there are varying levels of service availability and resources, including the provision of the personal care component of the package by private providers. This creates considerable challenges in ensuring comprehensive, integrated, safe and sustainable care packages are being consistently provided. There is currently also no centralised system of funding for these homecare packages which makes auditing, budgeting and planning difficult.

There is uncertainty regarding the exact extent of formal care hours that are required for patients availing of domiciliary ventilation, and, as noted, individual patients are likely to have different care needs. The Australian icare guidance, upon which the HSE attendant care recommendations are based,(2) recommends a minimum of 196 hours of formal care per week,(133) that is, 24/7 cover by one homecare staff plus overlap of 28 hours during the week by a second staff member for certain tasks (for example, bathing and changing tracheostomy tubes). However, several patients in Ireland are reportedly currently receiving 336 hours of care per week (two homecare staff each providing 24/7 care).(166) Given the significant cost of staffing, the total number of weekly paid formal care hours is the key factor influencing the costs of provision of domiciliary ventilation. Increasing such hours from 196 to 336 has a substantial impact on the total cost of a homecare package such that the five-year total budget impact per patient would increase from €3.4 to €4.8 million. However, it may be clinically appropriate and justified to provide this intensive level of care for these particular patients based on their individual care needs.(292) It may also be possible that some patients may need less than 196 hours of paid formal care, depending on their personal circumstances (for example, a family member may be a trained HCA or nurse and may be in a position to contribute a certain level of care hours in this capacity). While 196 hours was used in the base case for the BIA in this HTA to reflect a 'typical' homecare package based on international guidance,(133) this should be considered to reflect the *minimum* number of formal care hours provided, with the resulting budget impact estimate viewed as indicative rather than definitive. When planning homecare staff support, it is important to take human resource and practical issues (for example staff breaktimes etc.) into consideration. Continuity of care 24/7 is essential for these patients as they must be under the direct supervision of a competent person at all times, and this must factor into staff scheduling. However, due consideration also needs to be given to the limits of care supports that can realistically be provided in the community in the context of a fixed health budget and a limited availability of healthcare staff. Geographic inequities in service provision may be a feature even if a decision is made by the HSE to move towards a national provision of care. Further work is required to understand what level of service is currently being provided to these patients and how best it can be delivered to ensure a quality and equitable service within the resources available. This should involve consultation with the patients and families as well as with the homecare staff, private providers and clinicians involved in their care, with ongoing evaluation.

Ongoing training and support for patients, families and homecare staff was identified as a key gap in this assessment. Patients and families need support in the community on an ongoing basis as the patient's care needs may change (for example, as they develop secondary complications).(172) From a clinical governance point of view, it is not clear who is currently responsible for providing training to new homecare providers, particularly in the years and months after the patient has been discharged from the NRH.(144) A bespoke national training and support nurse role in the community, distinct from the current disability case manager role, could potentially fill this gap and provide a level of consistent training to those providing care to these patients in the community and provide ongoing support to patients and families. Such a role could ensure that homecare staff are competent to deliver care for these patients and provide quality assurance of domiciliary invasive ventilation services nationally. Collaboration with other National Clinical Programmes is critical for the establishment of such a role given that there are other patient groups (for example, motor neurone disease, amyotrophic lateral sclerosis (ALS) which is a subset of motor neurone disease, spinal muscular atrophy, Duchenne muscular dystrophy, post-polio syndrome and other muscular diseases)(97) who may avail of domiciliary invasive ventilation and may also benefit from access to a specialist nurse on a national footing. Development of an integrated care specialist role in the community may also bring other benefits, for example better interdisciplinary collaboration, oversight of phrenic nerve stimulator users in the community, and development of skill expertise in this area. As outlined in Chapter 5, the total budget impact for establishing and implementing such a new role was estimated at €0.45 million over a five-year period.

While there is a clear need for homecare staff to demonstrate competence prior to providing care to these patients with complex needs, there is inconsistent guidance internationally as to whether this care must be delivered directly by a nurse or with nursing oversight. Current practice in Ireland is that a nurse is required to provide this care 24/7, and this has budgetary and resource implications particularly in the context of ongoing healthcare staff shortages in the community.(183) In contrast, in the UK this care is led by HCAs who have demonstrated competence, with supervision by nurses.(143) It is crucial that any homecare staff involved in the care of these patients are trained and competent to provide the complex care required and know how to deal with urgent and emergency situations as they arise. There needs to be a clear clinical pathway for these patients in the community underpinned by a strong governance framework. However, before a decision is made by the HSE as to whether HCAs can take on more clinical work under the supervision of a nurse, consultation with the Nursing and Midwifery Board of Ireland (NMBI) should be undertaken to discuss the implications for nurses' scope of practice of delegating high risk tasks to unregulated professionals.(184) Greater delegation of roles to competent HCAs may also enable greater access to these services in the community given the current recruitment and retention issue in the homecare sector.(183) However, it is critical that any potential savings and increased access to homecare services do not compromise patient safety.

One of the key organisational barriers to the discharge of these patients with an adequate homecare package is the fragmented funding system. As discussed in Chapter 6, the current funding of homecare packages for these patients with substantial care needs is a challenge as these are funded from various different sources including local budgets; the funding of high dependency patients may overwhelm local budgets in particular. Often local budget holders have to make difficult decisions as to whether they should fund a package of care for a single ventilator-dependent SCI patient at the cost of other services.(248) Development of a centralised, national, systematic, dedicated and integrated budget with an associated national tender framework to cover personal care, nursing care and equipment, would likely alleviate the administrative burden and inequity of the system, and expedite the approval of a fully funded homecare package for these patients. While the 'typical' package could be tightly defined in terms of elements, it might not be defined in terms of intensity, because individual need will vary from patient to patient. Based on the BIA, the five-year total budget impact for a single patient is estimated to be €3.4 million, however this could be as high as €4.8 million depending on the patient's individually assessed care needs. If there is an expectation that one new patient will require this service every second year (that is a second and third patient avail of the service, on years three and five respectively), and accounting for the bespoke training and support role, then the resulting five-year total budget impact for the HSE overall is estimated to be between €6.7 million and €9.2 million, with the higher estimate reflecting the cost for three consecutive patients who each require 336 hours of paid care per week. In contrast, assuming a worst case scenario of two new patients every year, the total cost to the HSE overall is estimated to be between €21.2 million and €29.8 million over five years. Such a ring-fenced budget would also likely protect the funding for local services, would provide oversight across all patients availing of the service, and potentially ensure the same standards are applied across patients. Should a decision be made by the HSE to implement a centralised, ring-fenced budget for the provision of homecare to invasively ventilated patients with SCI, this will require substantial organisational cooperation across multiple agencies and programmes. Aspects of such a funding model may also be applicable to other patient groups with very high care needs, though extension of the model to other groups should be done on a case-by-case basis to avoid the centralised budget being depleted, thereby defeating the purpose of its establishment. Evaluation of any change in the funding model should be undertaken after a period of time to determine whether it should be retained and expanded to cover other types of homecare packages. For example, an evaluation of the HSE-funded Intensive Home Care Package (IHCP) initiative was published in 2018.(293) The aim of the IHCP initiative, which was launched in 2014, was to provide a greater range and level of services to older people and their families, to tailor and individualise home care delivery and to help address the pressures on acute hospitals. The evaluation found that investment in IHCPs can keep people living at home for longer, including people with significant levels of disability and cognitive impairment. Family support was found to be critical in the success of the initiative. The authors concluded that the provision of responsive, personalised support to older people, people with dementia, and their families was critical to ensuring home care into the future.(293)

Clinical pathways and governance processes for domiciliary ventilation services are clearly outlined in other jurisdictions such as the Netherlands(138) and Germany.(79, 139) For example, in the Netherlands there are four designated centres for chronic ventilation (called home ventilation centres) in the country, with each centre serving a different region (Groningen, Utrecht, Rotterdam and Maastricht). Each home ventilation centre is affiliated with a teaching hospital, and provides specialised care to individuals with chronic respiratory insufficiency regardless of the underlying disease (for example, ALS, which is a sub-set of motor neurone disease, obesity hypoventilation syndrome (OHS) and SCI).(11) Within the Dutch guidance the governance structure is clearly described and there is an understanding of who is responsible for different aspects of the patient's clinical care and technical requirements.(138) In the Irish system currently, the clinical pathway and governance process for these patients within the community is less well defined and there is some ambiguity surrounding roles and responsibilities, particularly with regard to tasks such as maintenance and repair of certain equipment,(155, 214) and the ongoing training of homecare providers.(144) Further work is needed to develop and implement this clinical pathway into the community, and to ensure that it aligns with the National Neuro-Rehabilitation Strategy,(294) the National Clinical Programme for Rehabilitation Medicine's model of care,(7) as well as the ambitions of Sláintecare.(8) It is important to consider that development of a clinical pathway with a national system of governance and funding may also indirectly help address the unmet needs of other patient groups (for example, younger people with disabilities residing in nursing homes who wish to transition home).(216)

There are many patient and social factors that need to be considered when implementing complex care in a home setting. Given the very high care needs of these patients, the home becomes hospital-like, and with that brings benefits (such as greater autonomy and freedom for the patient)(218) but also substantial challenges (such as carer burden and financial strain on families).(227) While it is standard practice to send these patients home with an appropriately funded care package when safe to do so,(2) which is supportive of patient autonomy and in line with the Assisted Decision-Making (Capacity) Act,(240) this may conflict with the views and preferences of family members who may have concerns about the consequences of providing this complex care at home for years potentially.(250) Young males are disproportionately represented in this patient cohort and this brings additional challenges for their partners, dependents, parents etc. Transitioning of this complex care into the home environment requires a sensitive approach that acknowledges the potential benefits for the patient in that it may improve their quality of life and validate their personhood, as well as the harms for the family as it may have a detrimental impact on their employment, finances and health. Greater provision of paid care, professional support, and respite care have been recommended in the literature to alleviate carer burden and make care provision more sustainable.(229) In Ireland, there is currently no nursing home or residential care facility that provides long-term or respite care for these ventilated SCI patients.(182) As this patient cohort and their family carers age, it is important that alternative living and respite arrangements are put in place which may require changes to existing funding models, capital investment in facilities and upskilling of nursing home staff. There is a risk that should no such arrangements be put in place that the patient could end up back in an acute hospital long-term. A 'shared care' model may also be considered which may involve, for example, the patient remaining in hospital Monday to Friday and returning home for the weekend. Particular funding and governance arrangements need to be put in place to allow this to happen, however provision of alternative living arrangements may allow a certain cohort of patients to go home some of the time instead of remaining in hospital all of the time.

Consideration should also be given to the National Housing Strategy for Disabled People 2022-2027.(295) The vision for this Strategy is: "*to facilitate disabled people to live independently with the appropriate choices and control over where, how and with whom they live, promoting their inclusion in the community, and to further enable equal access for disabled people to housing with integrated support services.*" The Strategy acknowledges that disabled people have equal rights to access housing and equal rights to accessing supports necessary to enable them to living independently within the community. However, it is important to acknowledge that there may be challenges in implementing this Strategy in the context of patients with complex round-the-clock care requirements. This Strategy aligns with other strategies and policies such as the Wasted Lives Report, which supports the movement of younger people with disabilities out of nursing homes,(216) and is in line with United Nations Convention of the Rights of Persons with Disabilities (UNCRPD).(296) However, in the context of extremely limited housing availability in Ireland currently, there may be added challenges in securing housing for these patients should a decision be made to purchase a house based on the individual's circumstance (for example, previously lived in an apartment or a rental property). Therefore it is important to take into account that there are other factors that may further delay the discharge process for these patients that are beyond the control of the HSE.

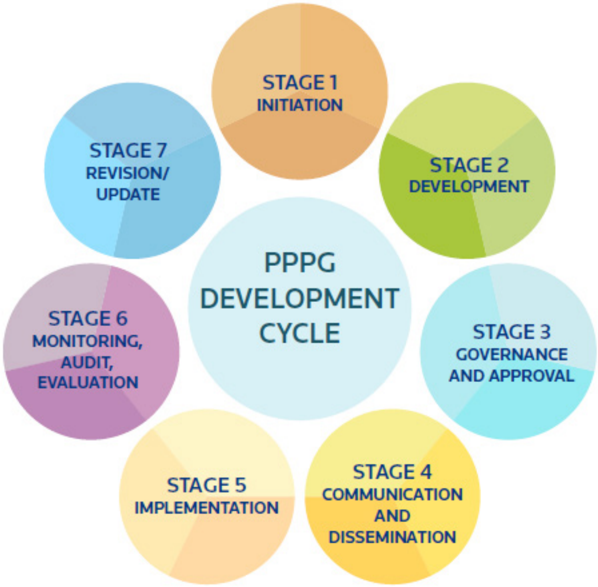
There are some important ethical issues with regards to the provision of domiciliary invasive ventilation services that need to be considered prior to the implementation of a national service. While provision of a national delivery of care for these patients should be viewed as supportive of patient autonomy, it is crucial that patients and families are fully informed of the benefits and risks of domiciliary ventilation, and that a true choice of options is provided to the patient that respects the preference of the individual. In terms of the benefit-harm balance, consideration needs to be given to the potential for the benefits of this national provision of care (for example, freeing up of scarce ICU/HDU beds and improvement of patients' quality of life) to be outweighed by its associated harms (for example, increased safety concerns associated with providing care at home, carer burden). From the perspective of justice and equity, provision of a systematic nationwide service may improve equity of access for these patients given the current geographical disparities in funding and service provision. Creation of a nationwide ring-fenced budget could prevent local budgets being depleted, allowing funding for other important community services to be preserved. However, it is associated with a significant opportunity cost given the substantial nursing requirements, and could be viewed as inequitable to other patient groups with complex care needs who could benefit from access to substantial nursing support. From a distributive justice/equity of care perspective, there will be a subset of these patients who face greater practical challenges to returning home. For example, single parents, and those who live alone or in rented accommodation. To be equitable, the national provision of care should also enable these patients to return home despite the obstacles.(282) It should be noted that while the ethical, patient and social considerations outlined are important and require due diligence in decision making, they are not necessarily unique to Ireland, nor to domiciliary ventilation for SCI specifically. These findings should not preclude the implementation of a national provision of care for this population, but rather provide tangible factors that should be considered during decision-making and accounted for during subsequent implementation should a positive recommendation be made.

It is important to acknowledge that while homecare is currently unregulated in Ireland, it is intended to introduce regulation in this area.(217) In light of the impending legislation, and the subsequent development of National Standards in this area, these may possibly impact on the national provision of a domiciliary invasive ventilation service. Consideration should also be given to the potential impact that introduction of a statutory homecare scheme (akin to the Fair Deal Scheme) may have on the personal finances of patients who avail of this service.(285) While regulation and national standards will aim to improve the quality of care delivered in the homecare sector, it is unclear at this time what impact, if any, these changes might have specifically on domiciliary invasive ventilation services in terms of staffing and qualification requirements, physical environment requirements, equipment and gas safety etc.

8.3. Policy implications

The work of the current HTA aligns with the HSE National Framework for developing Policies, Procedures, Protocols and Guidelines (PPPGs).(297) Specifically, conduct of the HTA fits into Stage 2 (Development) of the seven stage PPPG cycle (Figure 8.1) with the findings of the HTA relevant for informing a national clinical pathway for ventilator-dependent SCI patients that extends into the community.

Figure 8.1: PPPG Development Cycle



Key: PPPG - Policies, Procedures, Protocols and Guidelines.

Stage 2 of the PPPG cycle is the 'Development' phase and relates to *"the methodology by which the PPPG is developed and involves the integration of best research evidence with clinical/professional expertise, patient/client values and cost."* (297) The framework suggests that a HTA be undertaken in the case of more complex PPPGs, as was done in this instance. In the context of the national provision of services for ventilator-dependent adults with SCI, further work in the development stage is required by the National Clinical Programme for Rehabilitation Medicine in conjunction with the NRH Spinal Cord System of Care and HSE partners to describe the PPPG (that is, the clinical pathway) step by step using a clearly defined process, based on the advice and information provided in the HTA. Further consultation with relevant stakeholders including patients and family may be required. The next stage on the PPPG cycle is 'Governance and Approval'. This involves agreeing formal governance structures at local, regional and national level, based on the information provided in the HTA, with expert review of the draft PPPG as appropriate. The final PPPG document must be signed off by senior management/relevant governance process confirming that the PPPG meets the required standard. Stage 4 is 'Communication and Dissemination' and requires that a communication plan is developed to ensure effective communication and collaboration with all stakeholders throughout all stages. It is important that all affected stakeholders are aware of any imminent changes to the pathway. Stage 5 is 'Implementation' which refers to the process of putting to use or integrating new practices within a setting.(298) Five steps are outlined in the PPPG framework that need to be undertaken in order to meet the standards for Stage 5. The findings of the HTA will likely inform these steps. The five steps are:

- develop an implementation plan, including identification of responsible person(s), specifying the actions to implement the PPPG and timeframes for implementation.
- identify and record barriers and facilitators for implementation and use of the PPPG.
- align the implementation plan with the service plan and budgetary process.
- outline the supports required for education and training for staff on the implementation of the PPPG.
- establish good governance structures including strong leadership for the effective implementation of the PPPG being developed.

Stage 6 is 'Monitoring, Audit and Evaluation'. It is crucial that a clinical monitoring, audit and evaluation plan for continuous improvement of the clinical pathway is developed. Specific relevant outcomes should be specified in advance (for example, patient outcomes, service improvement, expenditure etc.) and measured in a robust and timely manner that assists with quality improvement. Good governance structures should be in place to ensure there is continuous improvement in the development, implementation, monitoring, auditing and evaluation of PPPGs (such as a PPPG Development Group, project sponsors or appropriate governance group, quality and safety groups/committees etc.). The final stage of the PPPG cycle is 'Revision/Update', which refers to ensuring that the learnings from the PPPG process are used to amend and update or revise the original PPPG as new evidence emerges. The framework recommends that this update is carried out at least every three years, unless there is a need to make revisions based on information received in the interim (for example, based on audit or legislative changes etc.)(297)

While a body of work is still required by the HSE and the NRH in order to develop and implement a clinical pathway on a national basis for ventilator-dependent patients with SCI in Ireland, the current HTA provides a strong foundation for this work.

8.4. Strengths and limitations

The findings of this assessment should be considered in light of its overall strengths and limitations. An Expert Advisory Group (EAG) comprising a broad range of key stakeholders was established to support the assessment, and stakeholders outside of EAG members were consulted to ensure the perspectives of a broad range of individuals involved in the care of ventilator-dependent adults with SCI were appropriately represented in the assessment. This HTA followed a methodical and transparent approach and was carried out in line with best practice; a protocol was developed and reviewed by the EAG at the outset of the project, (13) a review of international literature was undertaken to provide important information and context, and the assessment overall was conducted in accordance with national and international HTA guidelines. (142, 190)

The patient cohort of interest in this assessment, that is, ventilator-dependent adults with complete C1-C3 SCI who are clinically suitable for discharge, represents a very limited number of individuals (estimated to be less than 10 nationally). As such, there are challenges in reliably estimating the demand for, and costs of, domiciliary invasive ventilation services for these patients, and to adequately account for the variation that may be seen year on year. Assumptions regarding the equipment and care requirements were therefore frequently guided by expert opinion and international guidance documents, with notable discrepancies in care requirements reported. However, these estimates were validated across sources wherever possible and extensively tested in sensitivity and scenario analyses to quantify the impact of uncertainty. Nonetheless, these limitations present a risk that decision-making in the Irish context may be relying on estimates which could under- or overstate the potential cost of providing this service to this patient cohort. As discussed, there is substantial variability across patients in terms of care needs. The estimates presented here are generally intended to reflect an 'on average' cost based on typical needs and requirements based on international guidance and expert opinion. Due to the small numbers of patients, there may be a number of consecutive patients with complex needs that may be at the upper end of costings. Equally there could be a number of patients with needs at the lower end. For this reason, the budget cannot be precisely estimated and will need to be adaptive to some extent.

While extensive stakeholder engagement took place for this assessment, a limitation is that patients and families currently availing of domiciliary invasive ventilation were not directly consulted by the evaluation team. A decision was made by the evaluation team that direct engagement may be inappropriate in this instance in the absence of an ethical approval process to provide assurances of appropriate safeguarding given the vulnerability of the patient population. Acquisition of ethical approval from a suitable institution was, however, not considered to be feasible within the strict timelines of the project. That being said, substantial engagement occurred with Spinal Injuries Ireland, who liaised closely with the affected patients and families to provide the patient and family voices for this assessment. This was supplemented by a comprehensive review of the literature, with Chapter 7 providing a detailed examination of the relevant patient, social and ethical considerations. In specifying the exact details of the service to be provided, there should be consultation with patients with direct experience of the service.

Finally, while listed as part of the terms of reference, this assessment did not estimate the cost of providing care for these patients in long-term care settings. During the course of the assessment, it became apparent that no nursing home or other residential care setting in Ireland currently provides care for ventilator-dependent adults with SCI.(182) Because of this, there were no data available to provide cost estimates for provision of care to these patients in these settings. However, a gap was identified with regards to the lack of alternative living and respite arrangements for an ageing patient cohort and their families. There is a risk that, should no such alternative arrangements be put in place, that the patient could end up returning to the acute hospital setting on a long-term basis.

8.5. Conclusion

High cervical SCI requiring permanent mechanical invasive ventilation are rare but have substantial consequences for patients, families and health services. The results of this assessment support the development of a national clinical pathway for this patient cohort that extends into the community. This pathway should have an overarching clinical governance framework, and would benefit from a national training and support role, and to be funded through a centralised budget. This new clinical pathway, would potentially alleviate some of the challenges experienced by patients and families, streamline the process for securing homecare packages, and increase efficiencies in the health system through freeing up scarce ICU/HDU beds. Development of a bespoke training and support role could aid quality assurance of domiciliary invasive ventilation services nationally through the ongoing provision of competency-based training. It is important that the clinical pathway considers the importance of appropriate preparation and timing of the transition to the home setting; anticipation of present and future requirements in terms of environment and training of those providing care; and anticipation of disease progression. A comprehensive, holistic and person-centred approach to care, delivered from a well-coordinated interdisciplinary team, which respects the preferences of the individual, is critical for its success. It is crucial that a clinical monitoring, audit and evaluation plan for continuous improvement of the clinical pathway is developed. Patient, social and ethical aspects should also be carefully considered during decision-making and accounted for during subsequent implementation.

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Appendix 1: Search strategy

Databases searched:

- Medline (EBSCO)
- Embase (OVID)
- The Cochrane Library
- ClinicalTrials.gov
- CINAHL (EBSCO)
- APA PsycInfo (EBSCO)

Registries searched:

- ClinicaiTrials.gov

Grey literature sources searched:

- TRIP (Turning Research into Practice)
- BMJ Best Practice
- UptoDate
- Guidelines International Network
- International HTA Database
- Core
- Google
- Google Scholar
- The Physiotherapy Evidence Database (PEDRO)
- Agency for Healthcare Research and Quality (AHRQ)

Websites of international/national societies and agencies searched

- WHO

- European Respiratory Society
- American Thoracic Society
- Canadian Thoracic Society
- American Association for Respiratory Care
- American Spinal Injury Association
- Paralyzed Veterans of America
- Multidisciplinary Association for Spinal Cord Injury Professions (MASCIP) UK
- Thoracic Society of Australia and New Zealand
- British Association of SCI Specialists (BASCIS)
- Respiratory Information for Spinal Cord Injury (RISCI)
- The International Spinal Cord Society (ISCOS)

Example of Medline (EBSCO) search strategy:

S42 S32 OR S41

S41 S16 AND S31 AND S40

CONCEPT 4: QUALITY OF LIFE SEARCH FILTER

S40 S33 OR S34 OR S35 OR S36 OR S37 OR S38 OR S39

S39 AB ("Quality of life" OR "Life quality" "personal satisfaction" OR "patient satisfaction" OR "Activities of Daily Living" OR "Quality adjusted life year*" OR "Personal autonomy" OR "Happiness" OR "Patient preference*" OR "fear of death" OR "family relation*" OR "Religion" OR "social support" OR "financial support" OR "positive experience") OR TI ("Quality of life" OR "Life quality" "personal satisfaction" OR "patient satisfaction" OR "Activities of Daily Living" OR "Quality adjusted life year*" OR "Personal autonomy" OR "Happiness" OR "Patient preference*" OR "fear of death" OR "family relation*" OR "Religion" OR "social support" OR "financial support" OR "positive experience")

S38 (MM "Personal Autonomy") OR (MM "Happiness") OR (MM "Self Concept") OR (MM "Family Relations") OR (MM "Religion") OR (MM "Social Support") OR (MM "Financial Support")

S37 (MM "Quality-Adjusted Life Years")

S36 (MM "Patient Satisfaction")

S35 (MM "Activities of Daily Living")

S34 (MM "Personal Satisfaction")

S33 (MM "Quality of Life")

S32 S7 AND S16 AND S31

CONCEPT 3: HOME CARE

S31 S17 OR S18 OR S19 OR S20 OR S21 OR S22 OR S23 OR S24 OR S25 OR S26 OR S27 OR S28 OR S29 OR S30

S30 AB home N3 discharge OR TI home N3 discharge

S29 AB (HMV OR HIMV) OR TI (HMV OR HIMV)

S28 AB domiciliary OR TI domiciliary

S27 AB ("assisted living" OR TI ("assisted living")

S26 AB (homecare OR ‘home based’ OR 'home-based') OR TI (homecare OR ‘home based' OR home-based')

S25 TI home

S24 AB ("primary health" OR "primary healthcare" OR "primary health care" OR "primary care") OR TI ("primary health" OR "primary healthcare" OR "primary health care" OR "primary care")

S23 (MH "Primary Health Care+")

S22 (MH "Skilled Nursing Facilities")

S21 (MH "Long-Term Care")

S20 (MH "Community Health Nursing+")

S19 (MH "Home Nursing+")

S18 (MH "Home Care Services, Hospital-Based")

S17 (MH "Home Care Services+")

CONCEPT 2: VENTILATION

S16 S8 OR S9 OR S10 OR S11 OR S12 OR S13 OR S14 OR S15

S15 AB (respiratory N5 (fail* OR insufficienc*)) OR TI (respiratory N5 (fail* OR insufficienc*))

S14 AB ventilator OR TI ventilator

S13 AB mechanical respiration OR TI mechanical respiration

S12 AB (artificial N2 (ventilat* OR respirat*)) OR TI (artificial N2 (ventilat* OR respirat*))

S11 AB Tracheostom* OR TI Tracheostom*

S10 AB mechanical ventilat* OR TI mechanical ventilat*

S9 (MH "Tracheostomy")

S8 (MH "Respiration, Artificial+") OR (MH "Respiratory Insufficiency+")

CONCEPT 1 SPINAL CORD INJURY

S7 S1 OR S2 OR S3 OR S4 OR S5 OR S6

S6 AB (myelopath* N2 (post-traumatic OR posttraumatic OR post traumatic)) OR TI (myelopath* N2 (post-traumatic OR posttraumatic OR post traumatic))

S5 AB (Paraplegi* OR Paralysis OR Quadriplegi* OR Tetraplegi*) OR TI (Paraplegi* OR Paralysis OR Quadriplegi* OR Tetraplegi*)

S4 TX (spine OR spinal) N3 (injur* OR trauma* OR posttrauma* OR 'post trauma*' OR contusion*OR lacerat* OR transect*)

S3 (MH "Spinal Cord+") AND (MH "Wounds and Injuries+")

S2 (MH "Paraplegia+") OR (MH "Quadriplegia+") OR (MH "Paralysis+")

S1 (MH "Spinal Cord Injuries+")

Appendix 2: Excluded studies after full text review

Title	Authors	Notes
Guidelines on respiratory failure	Regione Toscana,	Exclusion reason: Wrong intervention
SERVICE SCHEDULE FOR SPINAL CORD INJURY REHABILITATION SERVICE	Accident Compensation Corporation (ACC) New Zealand,	Exclusion reason: Wrong setting
[Recommendations for invasive home mechanical ventilation]	Randerath, W. J.; Kamps, N.; Brambring, J.; Gerhard, F.; Lorenz, J.; Rudolf, F.; Rosseau, S.; Scheumann, A.; Vollmer, V.; Windisch, W.	Exclusion reason: Document has since been updated
Rehabilitation after traumatic injury	National Institute for Health and Care Excellence (NICE),	Exclusion reason: Wrong intervention
MECHANICAL HOME VENTILATION GUIDELINES	Intensive Care at Home	Exclusion reason: Specific to one hospital/centre
From intensive care unit to home discharge in the 24 h ventilator- dependent patient	Simonds, A. K.	Exclusion reason: Wrong study design
Role of interdisciplinary team function in the successful discharge of a ventilator-dependent person with a spinal cord injury	Piskule, A. A.; Johnson, P. B.	Exclusion reason: Wrong study design
Statement on home care for patients with respiratory disorders	American Thoracic Society,	Exclusion reason: Wrong study design
QUEEN ELIZABETH NATIONAL SPINAL INJURIES UNIT	QUEEN ELIZABETH NATIONAL SPINAL INJURIES UNIT,	Exclusion reason: Specific to one hospital/centre
Home discharge from the intensive care unit in patients with permanent mechanical ventilatory support	Santos Bouza, A.; De La Torre Garcia, R.; Robelo Pardo, M.; Peraza Torres, A.; Martin Lopez, C.; Portela Leon, M.	Exclusion reason: Wrong study design
Preparing the patient and family for home mechanical ventilation	Glass, C.; Grap, M. J.; Battle, G.	Exclusion reason: Not evidence-based
Guide for Health Professionals on the Psychosocial Care of Adults with Spinal Cord Injury	Agency for Clinical Innovation	Exclusion reason: Secondary health conditions
Clinical Practice Guidelines for spinal cord injuries	Paralyzed Veterens of America	Exclusion reason: Secondary health conditions
Home mechanical ventilation: the Dutch approach	van den Biggelaar, R. J. M.; Hazenberg, A.; Cobben, N. A. M.; Gommers, Dampj; Gaytant, M. A.; Wijkstra, P. J.	Exclusion reason: Wrong study design
Collaborative support for caregivers of individuals beginning mechanical ventilation at home	Glass, C.; Boling, P. A.; Gammon, S.	Exclusion reason: Wrong patient population
Maintaining a ventilator-dependent tetraplegic in a rural home setting	Trosper, R. M.; Rogers, P. C.	Exclusion reason: Wrong study design
Long-Term Mechanical Ventilation: Management Strategies...49th Respiratory	White, Alexander C.	Exclusion reason: Wrong study design

Care Journal Conference, â€œThe Chronically Critically Ill Patient,â€ September 2011, Florida		
Spinal cord injury rehabilitation practices	Spinal Cord Injury Research Evidence, SCIRE	Exclusion reason: Wrong study design
Treatment of the post-ICU patient in an outpatient setting	Volk, Bradford; Grassi, Frank	Exclusion reason: Wrong setting
[Home mechanical ventilation: Invasive and noninvasive ventilation therapy for chronic respiratory failure]	Huttmann, S. E.; Storre, J. H.; Windisch, W.	Exclusion reason: Wrong study design
Children requiring domiciliary/long term ventilation: Appendix to Standards of care for Spinal Cord Injured childrenand young people (0-19 yrs.)	MASCIP	Exclusion reason: Wrong setting
RISCI Respiratory Information for Spinal Cord Injury UK Weaning guidelines for Spinal Cord Injured patients in critical care units	Respiratory Information for Spinal Cord Injury	Exclusion reason: Wrong setting
Noninvasive Positive Pressure Ventilation in the Home	Wang, Zhen; Wilson, Michael; Dobler, Claudia C.; Morrow, Allison S.; Beuschel, Bradley; Alsawas, Mouaz; Benkhadra, Raed; Seisa, Mohamed; Mittal, Aniket; Sanchez, Manuel; Daraz, Lubna; Holets, Steven; Murad, M. Hassan	Exclusion reason: Wrong intervention
Noninvasive Positive Pressure Ventilation in the Home (with addendum)	Wilson, Michael; Wang, Zhen; Dobler, Claudia C.; Morrow, Allison S.; Beuschel, Bradley; Alsawas, Mouaz; Benkhadra, Raed; Seisa, Mohamed; Mittal, Aniket; Sanchez, Manuel; Daraz, Lubna; Holets, Steven; Murad, M. Hassan	Exclusion reason: Wrong intervention
Respiratory Information for Spinal Cord Injury	(RISCI), Respiratory Information for Spinal Cord Injury	Exclusion reason: Wrong study design
Evidence-based Practice Center Systematic Review Protocol Project Title: Home Mechanical Ventilators	Evidence-based Practice Center	Exclusion reason: Wrong study design
Development of the Canadian Spinal Cord Injury Best Practice (Can-SCIP) Guideline: Methods and overview	Eleni, M. Patsakos; Mark, Bayley; Ailene, Kua; Christiana, L. Cheng; Janice, J. Eng; Chester, Ho; Vanessa, K. Noonan; Matthew, QuerÃ©e; Craven, B. Catharine; Can, Scip Guideline Expert Panel	Exclusion reason: Wrong study design
Community Care for People with Complex Care Needs: Bridging the Gap between Health and Social Care	Kuluski, Kerry; Ho, Julia W.; Hans, Parminder Kaur; Nelson, Michelle La	Exclusion reason: Wrong study design
Living with a spinal cord injury	Queen Elizabeth National Spinal Injuries Unit	Exclusion reason: Wrong study design
[From case to case: nursing patients with a tracheostomy]	Messer, Melanie	Exclusion reason: Wrong study design
An interdisciplinary approach to transitioning ventilator-dependent patients to home	Warren, Mary Lou; Jarrett, Cheryl; Senegal, Rose; Parker, Anita; Kraus, Joy; Hartgraves, Debbie	Exclusion reason: Wrong study design
Home ventilation of adults with neuromuscular diseases a consensus paper	Winterholler, M.; Claus, D.; Bockelbrink, A.; Borasio, G. D.; Pongratz, D.; Schrank, B.; Toyka, K. V.; Neundorfer, B.	Exclusion reason: Wrong patient population

of the Bavarian Centers of Neuromuscular Disease		
A PARALYSED SYSTEM? An Inquiry into the provision of local health services forpeople being discharged from Spinal Cord Injury Centres	Spinal Injuries Association	Exclusion reason: Wrong study design
Home Mechanical Ventilation in Canada: A National Survey	Rose, Louise; McKim, Douglas A.; Katz, Sherri L.; Leasa, David; Nonoyama, Mika; Pedersen, Cheryl; Goldstein, Roger S.; Road, Jeremy D.	Exclusion reason: Wrong study design
[Non-invasive and invasive mechanical ventilation for treatment of chronic respiratory failure. S2-Guidelines published by the German Medical Association of Pneumology and Ventilatory Support]	Windisch, W.; Brambring, J.; Budweiser, S.; Dellweg, D.; Geiseler, J.; Gerhard, F.; KÃ¶hnlein, T.; Mellies, U.; SchÃ¶nhofer, B.; Schucher, B.; Siemon, K.; Walterspacher, S.; Winterholler, M.; Sitter, H.	Exclusion reason: Document has since been updated
Home mechanical ventilation for patients with Amyotrophic Lateral Sclerosis: A Canadian Thoracic Society clinical practice guideline	Rimmer, K. P.; Kaminska, M.; Nonoyama, M.; Giannouli, E.; Maltais, F.; Morrison, D. L.; O'Connell, C.; Petrof, B. J.; McKim, D. A.	Exclusion reason: Wrong patient population
SPINAL INJURIES BEST PRACTICE	North Wales Critical Care Unit	Exclusion reason: Wrong setting
An Official American Thoracic Society Clinical Practice Guideline: Pediatric Chronic Home Invasive Ventilation	Sterni, L. M.; Collaco, J. M.; Baker, C. D.; Carroll, J. L.; Sharma, G. D.; Brozek, J. L.; Finder, J. D.; Ackerman, V. L.; Arens, R.; Boroughs, D. S.; Carter, J.; Daigle, K. L.; Dougherty, J.; Gozal, D.; Kevill, K.; Kravitz, R. M.; Kriseman, T.; MacLusky, I.; Rivera-Spoljaric, K.; Tori, A. J.; Ferkol, T.; Halbower, A. C.	Exclusion reason: Wrong patient population
COMPLEX HOME VENTILATION (ADULT)	NHS England	Exclusion reason: Wrong patient population
Ventilatory support: preparing for discharge...The Howard H. Steel Conference on Pediatric Spinal Cord Injury, Rancho Mirage, California, December 3-5, 1999	Nelson, V. S.; Lewis, C. C.	Exclusion reason: Wrong patient population
[Support of a nursing system for patients requiring respiratory care: regional nursing and quality of life of patients with respiratory insufficiency]	Kinoshita, Y.	Exclusion reason: Wrong study design
Home care of the ventilator-dependent person	Gilmartin, M.; Make, B.	Exclusion reason: Wrong study design
Home rehabilitation and therapy	Garuti, G.	Exclusion reason: Wrong study design
LÃ©sions mÃ©dullaires traumatiques et non-traumatiques : analyse comparative des caractÃ©ristiques et de lâ€™organisation des soins et services de rÃ©adaptation au QuÃ©bec	Moutquin, J. M.	Exclusion reason: Wrong setting

[Mechanical home ventilation: its organization in Switzerland and perspectives]	de Haller, R.	Exclusion reason: Wrong study design
The ventilator-assisted children home program. Supporting families	Parra, M. M.; Goldsleger, F.; Chandler, S.	Exclusion reason: Wrong patient population
[An after-care treatment concept of artificially ventilated tetraplegic patients]	Meister, B.; Walther, W.	Exclusion reason: Wrong study design
[Health associated quality of life in home ventilation--1: In the labyrinth of concepts]	Fiedler, Sabine	Exclusion reason: Wrong study design
[Health related quality of life in home ventilation--2: Capable of more than breathing]	Fiedler, Sabine	Exclusion reason: Wrong study design
Home mechanical ventilation - A logistic and human medical challenge	Chevrolet, J. C.	Exclusion reason: Wrong study design
Chronic ventilation service	PATOUT, MAXIME; CUVELIER, ANTOINE; MUIR, JEAN-FRANÃ‡OIS; WIJKSTRA, PETER	Exclusion reason: Wrong intervention
Long-term ventilation: charting a pathway home	Graham, Robert J.	Exclusion reason: Wrong patient population
Home mechanical ventilation, the US perspective	Hill, N. S.	Exclusion reason: Wrong study design
Technology assessment and support of life-sustaining devices in home care. The home care physician perspective	Goldberg, A. I.	Exclusion reason: Wrong study design
Successful outcomes for the ventilator-dependent patient	Ecklund, M. M.	Exclusion reason: Wrong study design
Recommendations concerning the costing and management for patients requiring domiciliary ventilation	Krishnan, K. R.	Exclusion reason: Wrong study design
Tracheostomy Management; Clinical Guideline 2017.	Irish Association of Speech and Language Therapists	Exclusion reason: Not international
Home ventilator dependence after high cervical cord injury	Gower, D. J.; Davis, C. H., Jr.	Exclusion reason: Wrong study design
Invasive home mechanical ventilation, mainly focused on neuromuscular disorders	Geiseler, Jens; Karg, Ortrud; BÃ¶rger, Sandra; Becker, Kurt; Zimolong, Andreas	Exclusion reason: Wrong study design
Guidelines for the management of respiratory complications in patients with neuromuscular disease. Sociedad EspaÃ±ola de NeumologÃa y CirugÃa TorÃicica (SEPAR)	Farrero, Eva; AntÃ³n, Antonio; Egea, Carlos J.; Almaraz, M. JosÃ©; Masa, J. Fernando; Utrabo, Isabel; Calle, Miriam; Verea, HÃ©ctor; Servera, Emilio; Jara, Luis; Barrot, Emilia; CasolivÃ©, Vinyet	Exclusion reason: Wrong patient population
Long-term home mechanical ventilation in the United States	King, A. C.	Exclusion reason: Wrong study design
[Pediatric home ventilation--practical approach]	Rath-Wacenovsky, Regina	Exclusion reason: Wrong patient population
Best practice guidelines: spine Injury	American College of Surgeons	Exclusion reason: Wrong setting

Tracheostomy Care at Home	Total Community Care	Exclusion reason: Wrong study design website;
Home mechanical ventilation for children; the assessment	Jansen, N. J. G.; Westermann, E. J. A.; Van Der Voort, E.; Goorhuis, J. F.	Exclusion reason: Wrong patient population
Home mechanical ventilation	Stuban, S. L.	Exclusion reason: Wrong study design
A Discussion on Home Ventilation	King, Angela	Exclusion reason: Wrong study design
Preparing adults for home ventilator discharges	Buckley, T. W.	Exclusion reason: Wrong study design
Evaluation of the quality of published SCI clinical practice guidelines using the AGREE II instrument: Results from Can-SCIP expert panel	Patsakos, Eleni M.; Craven, B. Catharine; Kua, Ailene; Cheng, Christiana L.; Eng, Janice; Ho, Chester; Noonan, Vanessa K.; QuerÃ©e, Matthew; Bayley, Mark T.	Exclusion reason: Wrong study design
Integrated Care Pathway for the Management of Spinal Cord Injury	National Clinical Programme for Rehabilitation Medicine	Exclusion reason: Not international
GUIDELINES FOR THE PROVISION OF INTENSIVE CARE SERVICES	The Faculty of Intensive Care Medicine	Exclusion reason: Wrong setting
A Hospital-to-Home Program for Ventilator-dependent Children Sets the Standard of Care	Tamasitis, John; Shesser, Lynn	Exclusion reason: Wrong patient population
Home Mechanical Ventilation in Children	Preutthipan, Aroonwan	Exclusion reason: Wrong patient population
Home mechanical ventilation. A growing challenge in an aging society	Midgren, B.	Exclusion reason: Wrong study design
Best practice standards in home management of pediatric chronic invasive mechanical ventilation: A policy review for western Pennsylvania	Herrmann, Riley	Exclusion reason: Wrong patient population
Section 4: Home monitoring and follow-up of home-ventilated children	CÃ´tÃ©, Aurore	Exclusion reason: Wrong patient population
NIPH Systematic Reviews: Executive Summaries	Brurberg, K. G.; Landmark, B.; Kirkehei, I.; Reinar, L. M.	Exclusion reason: Wrong study design
Key constituents for integration of care for children assisted with long-term home ventilation: a European study	Brenner, Maria; O'Shea, Miriam P.; Larkin, Philip; Berry, Jay	Exclusion reason: Wrong patient population
Southport experience with domiciliary ventilation	Bingley, J. D.	Exclusion reason: Wrong study design
Pediatric home mechanical ventilation: A Canadian Thoracic Society clinical practice guideline executive summary	Amin, Reshma; Maclusky, Ian; Zielinski, David; Adderley, Robert; Carnevale, Franco; Chiang, Jackie; CÃ´tÃ©, Aurore; Daniels, Cathy; Daigneault, Patrick; Harrison, Christine; Katz, Sherri; Keilty, Krista; Majaesic, Carina; Moraes, Theo J.; Price, April; Radhakrishnan, Dhenuka; Rapoport, Adam; Spier, Sheldon; Thavagnanam, Surendran; Witmans, Manisha; Canadian Thoracic, Society	Exclusion reason: Wrong patient population
[Initiating patients on home mechanical ventilation]	de Mul, N.; van der Steen, M. S.; Gaytant, M. A.; Tjan, D. H. T.	Exclusion reason: Wrong study design

Ventilatory support at home for children: A joint position paper from the Thoracic Society of Australia and New Zealand/Australasian Sleep Association	Chawla, Jasneek; Edwards, Elizabeth A.; Griffiths, Amanda L.; Nixon, Gillian M.; Suresh, Sadasivam; Twiss, Jacob; Vandeleur, Moya; Waters, Karen A.; Wilson, Andrew C.; Wilson, Susan; Tai, Andrew	Exclusion reason: Wrong patient population
Respiratory Management Following Spinal Cord Injury: What You Should Know A Guide for People with Spinal Cord Injury	Paralyzed Veterans of America	Exclusion reason: Wrong study design;
A primary care provider's guide to managing respiratory health in subacute and chronic spinal cord injury	Reyes, M. R. L.; Elmo, M. J.; Menachem, B.; Granda, S. M.	Exclusion reason: Wrong intervention
Integrated Care Pathway for Patients with Spinal Cord Injury in Ireland	O'Driscoll, Edina; McElligott, Jacinta	Exclusion reason: Not international

Appendix 3: Guidance documents - completed data extraction forms

Appendix 3 is located separately

Appendix 4: Key assumptions underlying the budget impact analysis

Key assumptions	Justification
Hospital care is costed using four different methods: A) assuming a fixed ICU bed day cost, B) applying the ABF 2022 Price List for patients coded as 'TRACHEOSTMY/VENT>=96HRS, MAJC', C) an average NRH bed day cost of ~€1,400, and D) an assumption of the need for 6.5 WTE of nurses (50:50 mix of Enhanced Nurse and Enhanced Nurse Senior grades) to provide 24/7/365 care to one patient.	The NRH is not currently funded through the ABF model and there are no per patient cost estimates available.(157) Use of four different methods provides four crude estimates of cost for the hospital care of these patients. The Fixed ICU bed day cost was obtained confidentially from the HPO based on 2018 cost estimates (and inflated to 2022 prices) and assumes 100% ICU capacity and the need to purchase additional ICU beds.(158) Clinical coders in NRH confirmed that they code the patients as 'tracheostomy',(154) and the evaluation team made the reasonable assumption that these patients would be classed as having major complications given their complex healthcare needs. The Finance Department in the NRH confirmed that the average bed day cost in 2023 is approximately €1,400, and that approximately 6.5 WTE are required to provide 24/7/365 care to these patients. It was confirmed that a mix of Enhanced Nurse and Enhanced Nurse Senior grades currently provide care to these patients.(159) While there is no way of confirming these cost estimates for the NRH, the Finance Office indicated that the fixed ICU bed day cost was more likely to be closer to the value/ actual annual cost of providing the increased/enhanced nursing care required for these very complex patients, particularly in light of recent national wage increases.(299)
All equipment require maintenance at 10% of purchase price per year commencing year two, with the exception of those that are rented	Previous HIQA HTAs assumed an annual maintenance cost of 10% of the original purchase price applying from year two.(162) Discussion with a clinical engineer provided some confidence that this assumptions is a reasonable ballpark for the manufacturer's service contract.(155) It was confirmed by the spinal liaison nurse that for rented equipment, maintenance costs are included in the rental price under a service level agreement.(150)

All equipment are new and for the duration of the five-year period, do not need to be replaced	With regular maintenance is it assumed that the equipment will last at least five years.(155)
Depreciation value not a cost to the HSE	Equipment and ICT (updates, printers etc.) costs are recorded as upfront investments. As the equipment is an upfront cost and will not be sold after use - there is an assumption that depreciation is not an additional cost to the HSE.
The equipment, consumables and medications listed are those that are typically used by ventilated SCI patients	Based on discussions with clinicians involved in the care of these patients, it was advised that the items and medications listed are those that are typically required for this patient cohort.(150, 151)
A minimum of 196 hours of paid formal care is required per week in the base case. It is assumed that family will assist during periods when staff are on lunch/tea break etc. to ensure 24/7 cover. In a scenario analysis, 336 hours of paid care per week were assumed (comprising one nurse and one HCA each providing 24/7 care)	Irish and Australian guidelines specify that a minimum of 196 hours of care per week (28 hours per day) is recommended for ventilator-dependent patients with complete C1-C3 SCI.(2, 133) Based on discussions with some EAG members, some patients receive 336 hours of paid formal care.(166)
In the base case it is assumed that one nurse is providing care at all times, supplemented by a HCA/HCSA	This is current practice in Ireland as confirmed by members of the EAG.(143) In scenario analyses the ratio of care provided by nurses and HCAs is altered to examine the impact on the total cost of varying the staffmix ratio. Given that HCAs and HCSAs have the exact same salary scale even though their roles may differ slightly, for the purpose of costing, differentiation between the role is not important and so the term HCA is used for consistency.(146)

Given the 24/7 care required by nurses and HCAs, including at weekends and bank holidays, premium pay at 30% of total salary cost and travel costs at 10% of total salary cost are factored in. For the national support and training role, it is assumed that normal working hours are worked, but travel is expected so travel costs at 10% of total salary cost are factored in.	This is current HSE budgeting practice according to various HSE managers and the Finance Department in NRH.(152, 153, 159)
In year 1: Patient receives eight visits from an OT, six visits from a physiotherapist and four visits from a clinical psychologist. In year 2: Patient receives six visits from an OT, four visits from a physiotherapist and four visits from a clinical psychologist. In years 3-5: Patient receives no visit from an OT, four visits from a physiotherapist and four visits from a clinical psychologist.	Based on EAG discussions and follow up conversations with physiotherapy, occupational therapy and clinical psychology, estimates of health and social care professional input in the community were provided.(169, 170) Importantly the number of visits will vary depending on the individual patient's needs as well as service availability locally.
In preparation for discharge:	Based on EAG discussions, follow-up conversations(169, 170) as well as other conservative assumptions when no estimates were available.

A physiotherapist will deliver four hours of in-house training and will deliver 90 minutes of in-house training (5.5 hours in total). An OT will deliver three hours of in-house training and will deliver two hours of in-house training (5 hours in total). A clinical psychologist will deliver three hours of in-house training (3 hours in total). A nurse will deliver a full week of training and in-hourse support (37.5 hours in total)	
In the base case it was assumed that family or friends will drive the patient. In a scenario analysis it was assumed that the patient needs a driver (1 WTE)	Based on discussions with Spinal Injuries Ireland the importance of a driver was emphasised to enable the patient to engage in society to the fullest possible extent,(172) however it was assumed that in the context of availing of a tax relief scheme for an adapted vehicle,(179) family or friends would be able to drive the patient as and when required.
A new training and support nurse in the community is established (1 WTE) and associated training equipment and consumables. The equipment does not require maintenance as it is for training purposes only	The importance of ongoing training and support in the community was emphasised by various EAG members(143, 144, 172) and in guidelines.(90) Cost assumptions are based from another HSE national training programme.(153)
When estimating the total budget impact for the HSE over a five-period, it was assumed that one new SCI patient will require domiciliary invasive ventilation every second year	Based on epidemiological data and expert clinical opinion from EAG members, a conservative estimate of demand was assumed (that is, one new patient every second year).(69)
Basic and senior staff grades, where these exist for roles, are averaged	For these roles, it is often unknown whether a senior or basic grade staff would deliver these services and may depend on local availability, so an average hourly rate

	based on the HSE Consolidated Salary Scales is assumed.(146) For nurses in the base case, a 50:50 split of Staff Nurse and Clinical Nurse Specialist grades was assumed.
The patient has a PNS installed but still requires tracheostomy and ventilator supplies	Based on discussions with clinicians involved in the care of these patients, PNS is a viable alternative to permanent mechanical invasive ventilation, however these patients still require tracheostomy and back up ventilator in case of device failure.(208)
The average yearly cost of the PNS maintenance programme is used	A comprehensive PNS maintenance programme is undertaken by an external body over the course of eight years, with the costs varying substantially from year one to year eight. For the purpose of this BIA, an average annual cost of the eight year maintenance programme was included for each year.(150)
There is no additional cost to the HSE for each GP visit	It is assumed that the patient has a medical card and that GPs are reimbursed on a capitation basis for all eligible patients.(148) However it is noted that these visits represent an opportunity cost.
Two outpatient visits occur per year	Confirmed by a clinician involved in the care of ventilated SCI patients.(151)
The patient does not require enteral feeding	The requirement for enteral feeding varies by patient depending on whether their swallow is affected, however many ventilated SCI patients are on normal diet and fluid as confirmed by the liaison nurse.(300)
An ambulance is not needed for outpatient and GP visits	A suitably adapted vehicle is sufficient to bring patients to appointments as confirmed by a clinician involved in the care of of ventilated SCI patients.(301)
Treatment effects are similar between acute and hospital care	The evidence regarding the clinical benefits and harms of domiciliary ventilation is relatively scarce and is based on heterogeneous populations, study designs and methodological approaches, as well as small sample sizes.(80, 106, 109, 113) Very limited data pertain specifically to individuals with SCI availing of domiciliary invasive ventilation, given the rare occurrence of this condition and the ethical challenges of conducting comparative research in this population. Therefore it is unclear whether

	domiciliary ventilation leads to better (or worse) outcomes for patients with SCI compared with acute hospital care.

Key: ABF – activity based funding; GP – general practitioner; HCA - healthcare assistant; HCSA - healthcare support assistant; PNS – phrenic nerve stimulator; OT – occupational therapist; WTE – whole time equivalent.

Appendix 5: Costs falling on other government departments

Scheme	Details	Source
Housing adaptation grant for people with a disability	The Housing Adaptation Grant for People with a Disability scheme provides grant aid to applicants to assist in the carrying out of works that are reasonably necessary for the purposes of rendering a house more suitable for the accommodation needs of a person with a disability. The scheme is administered by local authorities. The effective maximum grant under the scheme is €30,000, which may cover up to 95% of the approved cost of works.	Government of Ireland(178)
Adapted vehicles. Drivers and passengers with disabilities: tax relief scheme	The scheme is open to persons who meet the specified medical criteria and have obtained a Primary Medical Certificate to that effect. They can apply for relief either as a driver with a disability or a passenger with a disability. Tax relief is available for the following applicant types, depending on the level of vehicle adaptation and is subject to a maximum amount of relief. • For passengers with a disability/family member of a passenger with a disability: ○ €16,000 for a passenger with a disability / family member of a passenger with a disability whose vehicle has adaptations ○ €22,000 for a passenger with a disability / family member of a passenger with a disability whose vehicle has more extensive adaptations.	Revenue(179)

Disability allowance	Disability Allowance (DA) is a weekly allowance paid to people with a disability. There may also be extra social welfare benefits and other supplementary welfare payments To qualify for Disability Allowance you must: • Have an injury, disease or physical or mental disability that has continued for at least one year or is expected to continue for at least one year • Be substantially restricted because of your disability from doing work that would be suitable for a person of your age, experience and qualifications • Be aged between 16 and 66. • Pass a means test • Live in Ireland and meet the habitual residence condition. You can get Disability Allowance, if you are in hospital or residential care. If you are already getting DA and go into hospital or residential care, you will continue to get your payment, as long as you continue to meet the qualifying conditions above. The weekly maximum rate of disability allowance in 2023: • Personal rate (claimant): €220 • Adult dependent: €146 • Child dependent under 12: €42 (full rate) and €21 (half rate) • Child dependent aged 12 years or over: €50 (full rate) and €25 (half rate)	Citizen’s Information(180)

Carer's Allowance	Carer's Allowance is a weekly social welfare payment to people who are caring for a person who needs support because of their age, disability or illness (including mental illness). To be entitled to Carer's Allowance you must: • Be age 18 or over • Pass a means test • Provide full-time care to a person who is not living in a hospital, convalescent home or other similar institution • Not live in a hospital, convalescent home or other similar institution. • Not be employed, self-employed, do voluntary work, training or any education courses for more than 18.5 hours a week. • Be habitually resident in the State The person you are caring for must be: • Age 16 or over and so incapacitated as to require full-time care and attention or • Under 16 and getting a Domiciliary Care Allowance. You may continue to get your Carer's Allowance in the following situations: • The person being cared for is attending non-residential rehabilitation training or a day care centre • You are caring for a child who is admitted to hospital for not longer than 6 months • You or the person being cared for (an adult) is having medical or other treatment in a hospital or institution for not longer than 13 weeks	Citizen's Information(181)

	• The person you are caring for goes into a nursing home on a full-time basis, your Carer’s Allowance can continue for 12 weeks. The Carer’s Allowance rates for 2023 are as follows: • Carer under 66, caring for 1 person – maximum weekly rate of €236 • Carer under 66, caring for 2 or more - maximum weekly rate of €354 • Carer aged 66 or over and caring for 1 person - maximum weekly rate of €274 • Carer aged 66 or over, caring for 2 people – maximum weekly rate of €411 • There is an increase of €42 (full rate) and €21 (half rate) for child dependents under 12, and €50 (full rate) and €25 (half rate) for child dependents 12 and over.	

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