The palliative care needs of adults with intellectual disabilities and their access to palliative care services: A systematic review

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Abstract

Background: There is evidence that people with intellectual disabilities experience healthcare inequalities, including access to specialist palliative care, but to date there has not been a systematic review of empirical evidence.

Aim: To identify the palliative care needs of adults with intellectual disabilities and the barriers and facilitators they face in accessing palliative care.

Design: Systematic review using a narrative synthesis approach (PROSPERO registration number: CRD42019138974).

Data sources: Five databases were searched in June 2019 (MEDLINE, Embase, PsycINFO, the Cochrane library and CINAHL) along with hand searches and a search of the grey literature. All study designs were included.

Results: 52 Studies were identified, all of which were conducted in high income countries, the majority in the UK (n=28). From a total of 2970 participants across all studies, only 1% were people with intellectual disabilities and 1.3% family members; the majority (97%) were health/social care professionals. Identified needs included physical needs, psychosocial and spiritual needs, and information and communication needs. Barriers and facilitators were associated with education (e.g. staff knowledge, training and experience); communication (e.g. staff skill in assessing and addressing needs of people with communication difficulties); collaboration (e.g. importance of sustained multidisciplinary approach) and health and social care delivery (e.g. staffing levels, funding and management support).

Conclusions: This review highlights the specific problems in providing equitable palliative care for adults with intellectual disabilities, but there is a lack of research into strategies to
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improve practice. This should be prioritised, using methods that include people with intellectual disabilities and families.

Keywords: Palliative Care, Health Services Needs and Demand, Intellectual Disability, Health Services Accessibility.

**Key statements**

**What is already known about this topic?**

- Adults with intellectual disabilities experience health care inequalities and are less likely to have access to palliative care.
- The specific palliative care needs of people with intellectual disabilities are poorly understood.
- Guidance and policy in this field is mostly built upon theoretical and anecdotal evidence with a very limited empirical knowledge base.

**What this paper adds**

- While the palliative care needs of adults with intellectual disabilities mirror those of the general population, there are specific and complex challenges associated with these individuals.
- Adults with intellectual disabilities face multiple barriers to accessing palliative care.
- There is a paucity of high quality research in this field and people with intellectual disabilities themselves, and their families, are not represented through the currently available evidence.

**Implications for practice, theory or policy**

- Addressing the inequalities in accessing palliative care for people with intellectual disabilities should be an urgent international priority.
- Examples of good practice and promising initiatives identified in this review need to be supported by good quality research, embedded in national policy and adequately funded.
- Future research should focus on developing measurable outcomes specifically related to people with intellectual disabilities to allow for large scale interventional studies that demonstrate these initiatives are effective and worthwhile.
The global prevalence of intellectual disability is estimated at approximately 1%\(^1\). Improved health and social care means this population is expanding\(^5\). While there are various definitions of intellectual disability, there is international consensus that it is present when the following three criteria are met: a significantly reduced ability to understand new or complex information and to learn and apply new skills (impaired intelligence); a reduced ability to cope independently (impaired social functioning); and beginning before adulthood, with a lasting effect on development\(^2-4\). In the UK, people with intellectual disabilities make up around 1.5 million people, and this is expected to rise by 1.1% annually, with those aged over 60 years set to increase substantially\(^5\). With an ageing population comes a rising prevalence of chronic and age-related illness, and subsequently an increased need for palliative and end of life care\(^5-8\).

Despite this trend, people with intellectual disabilities die approximately 25 years sooner than the general population\(^9\). Evidence from the World Health Organisation (WHO) atlas of global resources for people with intellectual disabilities reveals that people with intellectual disabilities are often denied their right to health care, even in countries with a relatively high standard of living\(^10, 11\). Worldwide, there is a paucity of documentation, information or epidemiological data about this population\(^10\). As such it is challenging to fully appreciate the prevalence and impact of intellectual disabilities on health care needs, and the associated barriers and facilitators experienced. Investigations into the deaths of people with learning disabilities in the UK\(^9, 12, 13\) have identified institutional discrimination and considerable evidence of health care inequalities contributing to avoidable excess mortality. In addition, people with intellectual disabilities were less likely to have access to specialist palliative care services and received less opioid analgesia in their final illness than people without intellectual disabilities\(^13\). The independent regulator of health and social care in England (the Care Quality Commission: CQC) found that exclusion of people with intellectual disabilities from wider health services was a significant barrier to care at end of life\(^14\).

Existing guidance to improve palliative and end of life care for people with learning disabilities is mostly based on theoretical or anecdotal evidence, expert opinion and case reports\(^6, 14, 15\). Guidance from the National Institute for Health and Care Excellence (NICE) in the UK regarding older people with intellectual disabilities\(^16\) was based on a systematic review which identified only two studies reflecting the views of people with intellectual disabilities and their families. In order to understand more about this gap in knowledge, and about the best direction for further research in this area, it is essential to examine in detail what is known about the inequalities faced by this population. The aim of this systematic review was to identify the palliative care needs of adults with intellectual disabilities and the barriers and facilitators this population face in accessing palliative care.
Methods

The protocol for this systematic review was registered in the PROSPERO database\textsuperscript{17} (CRD42019138974) and is reported according to the PRISMA guidelines\textsuperscript{18}.

Review questions
What are the palliative care needs of adults with intellectual disabilities?
What are the barriers and facilitators this population face in accessing palliative care?

Search Strategy

MEDLINE, Embase, PsycINFO, the Cochrane library and CINAHL were searched in June 2019. MeSH terms and key words, guided by previously published systematic reviews\textsuperscript{19-21}, were combined with a pre-defined palliative care filter\textsuperscript{22} to form the search strategy (Appendix 1). Hand searching included two intellectual disability specific journals: The Journal of Applied Research in Intellectual Disabilities (JARID) and The Journal of Intellectual Disability Research (JIDR). These were considered to be the most relevant journals in the field if intellectual disability. They were searched online, without any date restrictions, to check for articles that may not have been captured in the database search. Reference lists of key publications\textsuperscript{6, 9, 13-15, 23, 24} and of identified studies were also reviewed. The grey literature was searched online using OpenGrey\textsuperscript{25} and CareSearch\textsuperscript{26}.

Inclusion and exclusion criteria

An inclusive approach was adopted given the paucity of research in this field. Multiple study designs and all health care settings were included and no geographical or date limitations applied. Table 1 details inclusion and exclusion criteria.

Table 1: Eligibility criteria

| Inclusion criteria |  |
|-------------------|---|
| **Population**    | • Studies including adult participants aged 18 and over with intellectual disabilities and a ‘life-threatening illness’ as per the WHO definition of palliative care\textsuperscript{27}.  
• Studies including participants who are carers/relatives or health/social care staff caring for this population. |
| **Setting**       | • Home, hospital, hospice, nursing/residential home, outpatient, primary care/community.  
• Worldwide. |
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Outcomes

- Any outcomes relating to palliative care needs of adults with intellectual disabilities.
- Any outcomes describing barriers and/or facilitators to accessing generalist or specialist palliative care.

Study designs

- Qualitative and quantitative research methods.
- Experimental study designs: randomised controlled trials (RCTs), quasi-experimental.
- Observational study designs: cross-sectional, cohort, case-control.
- Prospective and retrospective designs.
- Literature reviews and systematic reviews.
- Case series and case reports.

Exclusion criteria

- Discussion and opinion papers, conference abstracts, editorials, letters, comments, guidelines.
- Non-English articles where translation cannot be achieved.

Study Selection

The electronic searches identified 6632 articles. Following removal of duplicates, titles and abstracts were reviewed to assess if they merited full text analysis. A second researcher (ITW) reviewed a sub-set of 100 randomly selected titles and abstracts to check for inter-rater agreement with the primary researcher (EA). Any disagreement was resolved with a third researcher (KES). A kappa value of 0.67 demonstrated ‘substantial’ agreement.

Analysis of the full text was then undertaken by EA. Endnote and Rayyan were used to manage the selection process. A total of 52 articles were included for final analysis. Figure 1 details the selection process.

Data extraction

Data were extracted using a Microsoft Excel template (Appendix 2) developed with guidance from the Cochrane ‘Checklist of items to consider in data collection or data extraction’.

Grading of quality

In order to evaluate the quality of the evidence included in the review, the articles were assessed using Hawker et al’s critical appraisal tool (Appendix 3). This was designed specifically to assess research conducted using different paradigms. It consists of 9 questions, each of which are scored on a four-point scale from very poor (1 point) to good (4 points). Based on a scoring system adapted by Voss et al, total scores of 18 or less were defined as poor, 19-27 as moderate and above 27 as good (Appendix 4).
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**Data analysis**

Information was collected for any outcomes relating to the palliative care needs of adults with intellectual disabilities and/or facilitators and/or barriers to accessing palliative care. Due to the heterogeneity of the studies, narrative methods were employed to synthesise the data. This encompassed the generation of themes, which was supported by the qualitative data analysis software NVivo. This narrative synthesis approach involved collating study findings into a textual narrative, along with tables and graphs. It was considered an appropriate choice of method, being suited to both quantitative and qualitative data and where statistical synthesis is not possible.
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Figure 1: Screening process based on the PRISMA flow diagram

Articles identified through database searching (n=6632)
- MEDLINE (n=894)
- Embase (n=3452)
- PsycINFO (n=915)
- Cochrane (n=495)
- CINAHL (n=876)

Duplicates removed (n=1486)

Titles and abstracts screened (n=5146)
- Wrong population (n=4871)
- Wrong publication type* (n=119)
- Wrong outcome** (n=2)

Full text articles assessed for eligibility (n=154)
- Wrong population (n=67)
- Wrong publication type* (n=40)
- Wrong outcome** (n=2)
- Duplicate (n=2)

Articles identified through hand search and grey literature (n=9)

Articles included in final analysis (n=52)
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Figure 1 legend

*Wrong publication: fell into one of the following categories in the exclusion criteria: ‘discussion and opinion papers, conference abstracts, editorials, letters, comments, guidelines.’

**Wrong outcome: did not include ‘Any outcomes relating to palliative care needs of adults with intellectual disabilities’ or ‘Any outcomes describing barriers and/or facilitators to accessing generalist or specialist palliative care’ as listed in the inclusion criteria.

Results

Study Characteristics

All studies were conducted in high income countries, with more than 50% conducted in the UK (Table 2).

Table 2: Geographical location of research

| Country                      | Number of articles |
|------------------------------|--------------------|
| UK                           | 28                 |
| Netherlands                  | 6                  |
| USA                          | 6                  |
| Ireland                      | 4                  |
| Australia                    | 3                  |
| Multiple European countries  | 3                  |
| Canada                       | 1                  |
| New Zealand                  | 1                  |
| Total                        | 52                 |

Qualitative methods were the most commonly used (n=17). There were 12 mixed method studies, 12 literature reviews, 10 studies with quantitative methods and 1 systematic review. There were no randomised controlled trials (RCTs). According to Hawker et al’s checklist\textsuperscript{34}, four articles were graded ‘poor’, 25 were ‘moderate’ and 23 were ‘good’ (Appendix 4).

The studies covered a range of health and social care settings including home, hospice, hospital, community, residential and nursing homes. Many studies did not describe the
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setting (n=24). For those that did (n=28), most involved participants based in community or residential settings (n=16).

There were 2970 participants included across the 52 studies. The majority of participants were health or social care professionals (n=2875, 96.8%). There were more intellectual disability professionals (n=1167, 39.3%) than there were specialist palliative care professionals (n=783, 26.4%) and more nurses (n=256, 8.6%) than physicians (n=129, 4.3%). There were 38 family members (1.3%) and 31 people with intellectual disabilities and a life-threatening illness (1%) acting as participants themselves (Figure 2).

Figure 2: Participants across studies

| Type of Participant                                      | n     |
|----------------------------------------------------------|-------|
| Person with intellectual disability as case report       | 26    |
| Person with intellectual disability as participant       | 31    |
| Family member                                            | 38    |
| Physician                                                | 129   |
| Nurse                                                    | 256   |
| Other health/social care professional                    | 540   |
| Specialist palliative care professional                  | 783   |
| Intellectual disability care professional                | 1167  |

*Qualitative synthesis*

The data were synthesised according to needs, barriers and facilitators.

**Needs**

Information relating to the palliative care needs of people with intellectual disabilities was identified in 32 studies. With the exception of one paper\(^{38}\), all of these studies reflected the perspectives of health care professionals or relatives. Three themes were identified: 1) Physical needs, 2) Psychosocial and spiritual needs, 3) Information and communication needs.
1) Physical needs
The most common physical need identified by staff was pain management. Many health care professionals described the challenge of symptoms being expressed as objectively observable signs or behaviours such as irritability, inactivity, loss of appetite and sleep disturbance, rather than spontaneous complaints. Other symptoms requiring attention included; nausea, vomiting, fatigue, shortness of breath, constipation, delirium, urinary incontinence and oral thrush. Monitoring of hydration and nutrition was also important. Many patients experienced declining mobility and needed physiotherapy. Physical needs also involved wound care, pressure area care and personal care such as washing.

2) Psychosocial and spiritual needs
The importance of family and the need for a social network was expressed widely. A study involving interviews with health professionals and carers highlighted that people with intellectual disabilities at the end of life need to be surrounded by people that know them well and can advocate for them. Socialising, friendships and human contact are important. People with intellectual disabilities who are dying require continuation of safe routines, treasured activities and important relationships. One study also highlighted the need to be occupied. Many studies recognised the importance of creating a familiar and predictable environment. For many this involved provision of care in the person’s home for as long as possible. A change in environment or unfamiliar faces may cause distress and hinder communication.

People with intellectual disabilities and a life-limiting illness experience fear and anxiety particularly when receiving bad news or having difficulty understanding medical information. Some studies spoke of grief and loss, non-verbal expression of grief and the recognition of complicated grief, and endorsed the need for appropriately tailored counselling services. There is also evidence that many people with intellectual disabilities have additional mental health problems which require increased support measures. Spiritual needs were identified, but not explored in detail. The need for culturally appropriate care was also highlighted.

3) Information and communication needs
People with intellectual disabilities are often not provided with information in an accessible format. This is important in enabling them to understand their diagnosis, prognosis and the symptomatic course of their illness. It is the responsibility of health care professionals to consider communication differences and the difficulty people with intellectual disabilities may have in understanding abstract concepts. People with intellectual disabilities often need help to express their views and participate in decision making. They need honest communication and opportunities to make choices. This
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is also important for advance care planning, with several papers placing emphasis on the need to discuss, document and respect preferred place of care and death, which tended to be home\(^1, 43, 46, 62, 63\).

**Barriers and facilitators**

Many factors acted as barriers as well as facilitators to providing palliative care to people with intellectual disabilities, depending on whether they were present or absent. Four themes were identified: 1) Education, 2) Communication, 3) Collaboration, 4) Health and social care delivery. A conceptual model of barriers and facilitators was developed from these themes (Figure 3).

1) **Education**

*Education – barriers*

Inadequate education was the most widely reported barrier. Particularly evident was the lack of experience, preparation and training in delivering end of life care amongst staff looking after people with intellectual disabilities in community or residential settings\(^46, 48, 50, 62, 64-70\). Lack of preparedness for seeing residents entering the dying phase of their lives hindered engagement with palliative care services\(^68\). A survey exploring the educational needs of intellectual disability care practitioners revealed poor understanding of diagnosis and causation of death, which raised concerns about their ability to recognise a dying patient\(^48\). These issues are compounded by carers working in isolated community settings without access to guidelines\(^42\). It was often standard practice for support workers to be on their own with residents when they were dying\(^68\) and staff that spent the most time with patients were often the least trained\(^54\).

The data also reflected a need for specialist palliative care professionals to improve their knowledge and experience in managing patients with intellectual disabilities\(^8, 49, 53, 54, 65, 71-73\). Additionally, several studies highlighted the need to educate registered nurses in both hospital and community settings in caring for patients with intellectual disabilities at the end of life\(^8, 44, 61, 74-76\).

Inadequate training can heighten communication fears\(^67\), lead to lack of confidence amongst staff\(^51\) and elevated levels of stress\(^45\). An intellectual disability support worker commented in a focus group discussion that there was a tendency of staff in hospital settings to put patients with intellectual disabilities and palliative care needs into the "too-hard basket"\(^8\).
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Contributing to this knowledge gap is the absence of formal protocols, policies and guidelines regarding end of life care for people with intellectual disabilities. A study examining guidelines in 11 European countries suggested that European national palliative care guidelines do not meet the needs of people with intellectual disabilities. Lack of organisational policy leads to inconsistent practice across settings and a postcode lottery whereby end of life care outcomes are often determined by individual staff.

Education – facilitators

Proposed educational incentives included a hospice resource folder containing information on local intellectual disability services, an intellectual disability toolkit designed to support hospital professionals and recruitment of a hospice intellectual disability link nurse to encourage integrated learning. Some studies suggest specific areas to focus on such as breaking bad news training for staff in intellectual disability settings, advanced training on recognition of non-verbal signs of pain and leadership skills. There is evidence that education in the form of a study day for paid carers can lead to improved knowledge and increased awareness about end of life care. A multi-layered approach to learning, delivered at both the individual and organisational level, contributed to successful implementation of the ‘Steps to Success Palliative Care Programme’ for people with intellectual disabilities living in residential care homes.

Professionals working within intellectual disability and palliative care services expressed a desire to learn from each other through joint education, exchange placements and liaison working. Small group discussion between peers and hearing the perspectives of people with intellectual disabilities themselves were also effective educational techniques. Wider approaches are the inclusion of palliative care for people with intellectual disabilities as a core component in medical and nursing curriculums and examinations.

2) Communication

Communication – barriers

Many studies reported inadequate communication skills amongst staff working with people with intellectual disabilities. Fear of initiating conversations about death and lack of experience in breaking bad news were widely reported issues, with a tendency in intellectual disability settings to keep things positive. Staff were also concerned that they may cause distress to the person with intellectual disabilities. Health care professionals may use complex language and fail to recognise the difficulty people with intellectual disabilities can have in understanding abstract concepts relating to death. An assumption that the person with intellectual disabilities lacks capacity and cannot provide informed consent leads to an over-reliance on carers or relatives as communication proxies, facilitates information gatekeeping and prevents open
Communication barriers also relate to the impaired ability of a person with intellectual disabilities to express themselves and understand information\(^{51, 53, 55, 69, 80}\). People with intellectual disabilities who are non-verbal present a particular challenge for staff and carers\(^{55, 60, 74, 79, 81}\). Symptom assessment is difficult, and there may be a diversity in signals that lead to recognition of the dying phase\(^{82}\). Acquiescence also presents a challenge for health care professionals, who may not recognise that people with intellectual disabilities are unlikely to question treatment decisions\(^{38}\). Difficulties confirming understanding, ascertaining information requirements and establishing wishes\(^{45}\) can result in failure to involve people with intellectual disabilities in the decision making process\(^{1, 51, 81}\). This can lead to conflict and uncertainty when the person’s health deteriorates\(^{83}\). A UK survey describing end of life care outcomes for adults with intellectual disabilities found that few individuals had their end of life preferences recorded and the majority were not aware they were going to die\(^{63}\).

**Communication – facilitators**

A person-centred approach incorporating compassion and empathy are important when caring for people with intellectual disabilities who have difficulty communicating\(^{7, 67, 69}\). Taking time to build relationships, gain trust and confirm understanding can facilitate effective communication\(^{1, 43, 49, 51, 60, 73}\). As people with intellectual disabilities are likely to take longer to express themselves and may rely on communication tools, health care professionals should allocate more time for consultations\(^{39, 60}\). This is particularly important when talking about death and dying\(^{61}\). Continuity of care\(^{43, 45, 73}\) and involvement of a family member can also aid communication\(^{3, 39, 41, 56, 58}\). Helping people with intellectual disabilities to understand and cope with bad news requires building of knowledge gradually over time and support from the person’s family and professional network\(^{57}\). Translating medical terminology into simpler concepts maximises involvement in decision making\(^{43}\). Involving speech and language therapists is another approach\(^{46, 55}\). Pictorial books designed to help people with intellectual disabilities understand and discuss terminal illness can be helpful\(^{56, 60}\). Structured models for breaking bad news to people with intellectual disabilities have also been developed\(^{56, 57}\).

Tools to aid assessment of physical symptoms include the ‘DisDAT’ (Distress Assessment Tool)\(^{39, 56, 58, 67}\), developed for people with severe communication problems including those with intellectual disabilities, the ‘REPOS’ (Rotterdam Elderly Pain Observation Scale)\(^{41}\) and the Abbey scale\(^{56}\). The ‘PALLI’ (PALliative care: Learning to Identify in people with intellectual disabilities) is a tool for use by proxies\(^{84}\).
Collaboration - barriers

Lack of collaboration between services was a widely reported barrier. Most often this was between intellectual disability and specialist palliative care services\textsuperscript{44, 53, 62, 65, 71, 72}. Evidence suggests poor referral rates for patients with intellectual disabilities to specialist palliative care services\textsuperscript{54, 65, 71}, with few patients receiving dual hospice and intellectual disability care\textsuperscript{72}. There is a deficiency of established relationships between intellectual disability and palliative care services\textsuperscript{72} encompassing a poor understanding of each other’s role, what the service is providing and how it is run\textsuperscript{54, 65}. For example, one paper described an intellectual disability care home manager who did not know how to access the palliative care team\textsuperscript{42}. Another issue is the medical versus social model of care\textsuperscript{55, 71}. Carers familiar with the social care model may neglect the physical aspects of care for the dying\textsuperscript{48}. Interviews with intellectual disability and specialist palliative care professionals revealed mistrust between services or conflict regarding ownership of the patient\textsuperscript{71}. Limited sharing of information between services and poor referrals can lead to inadequate knowledge of the patient\textsuperscript{42, 44, 61}. A view that patients require specialist intellectual disability services leads to exclusion from general palliative care services\textsuperscript{65} and a reluctance of intellectual disability services to acknowledge death can mean patients who are dying often remain hidden\textsuperscript{46}.

Family members are often expected to make complex ethical decisions\textsuperscript{83} and lack of understanding regarding focus of care in advanced illness\textsuperscript{44, 56} can lead to limited cooperation with palliative care services\textsuperscript{44}. Conflicts between staff and surrogate decision makers have been cited as a common barrier to hospice care\textsuperscript{80}. One study identified several nurses who were barred from visiting a client by family members who feared they would disclose a poor prognosis\textsuperscript{57}.

Collaboration - facilitators

Collaborative working between palliative care services, intellectual disability services and carers was an effective way to deliver care to people with intellectual disabilities\textsuperscript{43, 44, 49, 51, 65, 68, 79}. Earlier involvement of palliative care builds familiarity and trust between staff and services\textsuperscript{56}. Incentives such as regional meetings, joint working or shadowing in both clinical areas can be effective\textsuperscript{71}. A mixed methods study described the UK’s first specialist palliative care home for older people with intellectual disabilities, demonstrating positive results for quality of life\textsuperscript{7}.

Key to effective collaboration is a multidisciplinary approach allowing shared expertise between intellectual disability, specialist palliative care, hospital services, community teams and GPs\textsuperscript{40, 46, 52}. Specialist palliative care professionals found that liaising with intellectual disability professionals who knew the patient well was helpful around issues of mental
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capacity and consent. Link-workers acting as conduits between palliative care and intellectual disability services are also helpful. Working collaboratively with the family should encompass recognition that they know the person best, building trust, sharing information, involving them in decision making and supporting them emotionally.

4) Health and social care delivery

Health and social care delivery - barriers

Diagnostic overshadowing is a prevalent issue resulting in late diagnosis of terminal illness and delayed recognition of dying. Compliance with care, examination or prescribed medication may be a challenge among people with intellectual disabilities which can also compromise identification and management of symptoms. Some doctors declined to take on patients with intellectual disabilities at the end of life because they lacked the time to manage their complex medical issues. People with intellectual disabilities often have unpredictable clinical trajectories making it difficult to prognosticate and an ageing intellectual disability population brings changing health care needs and disease profiles. The authors also observed that time constraints, inadequate staffing levels and underfunding were barriers to supporting the additional health care needs of people with intellectual disabilities at the end of life.

An ageing intellectual disability population means parent caregivers are increasingly elderly and for people with intellectual disabilities living at home, lack of home care options means they require transfer to long-term facilities when their health deteriorates and their parents can no longer cope. Given the short-term nature of hospices, many people with intellectual disabilities are misplaced in nursing homes for people much older than themselves, which lack the expertise to meet their needs. There are few nursing homes that have this expertise. Intellectual disability staff resistance to provide end of life care at home may also prompt transfer to hospital or a nursing home when their client’s health deteriorates.

Health and social care delivery - facilitators

Several studies have highlighted the role of the GP as a key player in identification of need and coordinating referrals for people with intellectual disabilities. Developing community based services with input from GPs and district nurses will support people with intellectual disabilities living at home and allow them to die there.

Delivery of palliative care to this population is often dependent on committed staff who are willing to work beyond their call of duty. Delivery of highly individualised care requires teamwork, empathy and enthusiasm. Many studies highlighted the value of good management and support for staff within their own organisation. This should
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encompass emotional and bereavement support\textsuperscript{38, 46, 61, 67, 68, 71}. Building resilience and empowering the workforce enables them to deal with grief and in turn support the patient\textsuperscript{57, 62, 70, 71}. In organisations where managers provided positive role modelling by talking about death and dying, junior staff were more likely to feel comfortable discussing these topics\textsuperscript{70}. Adequate staffing and minimal staff turnover were also important to deliver effective and sustainable palliative care to this population\textsuperscript{7, 68, 78}.

Simple practical adjustments that help people with intellectual disabilities engage with palliative care services include allowing the presence of a family member or keyworker during hospital admissions, consultations and investigations\textsuperscript{42, 43, 46} and visits to hospital or treatment units beforehand\textsuperscript{40}. Good practice also includes the integration of the views of service users into models of care\textsuperscript{45}. 
Figure 3: Conceptual model of barriers and facilitators to accessing palliative care for adults with intellectual disabilities

**Barriers**
- Inadequate knowledge and training, lack of preparedness, learning on the job, fear of death.
- Lack of palliative care knowledge amongst intellectual disability staff.
- Lack of experience of intellectual disabilities amongst specialist palliative care staff.
- Absence of protocols, policies and guidelines.
- Inadequate communication skills amongst staff, avoidance of death related communication.
- Use of complex language by staff.
- Assumption that person lacks capacity.
- Fear of causing distress, gatekeeping, paternalism, collusion.
- Person with intellectual disabilities: difficulty expressing themselves and understanding information, non-verbal, acquiescence.
- Lack of collaboration between intellectual disability and palliative care services, lack of awareness of each others role, lack of referrals to palliative care.
- Absence of confident formal leadership.
- Medical versus social model of care, conflict between services, limited sharing of information, poor communication between staff.
- Lack of family involvement in care, conflict with family.
- Diagnostic overshadowing, late diagnosis, delayed recognition of dying.
- An ageing population with complex medical needs.
- Time constraints, poor staffing, underfunding.
- Lack of community and home care options = inappropriate transfer to nursing home or hospital.
- Lower levels of support in hospital.

**Facilitators**
- Hospice resource folders, intellectual disability toolkits.
- Intellectual disability link nurse.
- Staff study days, breaking bad news training, non-verbal communication skills, leadership skills.
- Multi-layered approach: delivered at organisational and individual level.
- Joint education, exchange placements, liaison working.
- Inclusion in medical and nursing curriculums and examinations.
- Person-centred approach, compassion, empathy, familiarity, trust, time.
- Skilled observation, assessment tools e.g. DiSDAT, PALLI.
- Translating medical terminology into simpler concepts.
- Total communication techniques, involving speech and language therapist, tools such as books beyond words.
- Structured models for breaking bad news.
- Early involvement of palliative care.
- Regional meetings, joint working.
- Multidisciplinary approach, shared expertise.
- Link-workers, palliative care outreach.
- Working collaboratively with family.
- Educational resources for family.
- GP as key player, developing community services.
- Public health approach.
- Highly individualised care, committed staff, compassion.
- Teamwork, good management, emotional support for staff.
- Adequate staffing, minimal turnover.
- Family member presence during admissions and consultations.
- Integration of views of service users into models of care.
Discussion

Main Findings
This systematic review identified 52 studies providing information on the palliative care needs of adults with intellectual disabilities and the barriers and facilitators this population face in accessing palliative care. All of these studies were conducted in high income countries, the majority in the UK. Qualitative methods were most commonly used. The studies mainly reflected experiences from community or residential settings and there was a strong bias towards the experiences of health care professionals. From a total of 2970 participants, just 31 people with intellectual disabilities were included. Identified needs included physical needs, psychosocial and spiritual needs, and information and communication needs. Barriers and facilitators were associated with education, communication, collaboration, and health and social care delivery.

What this study adds
This systematic review provides the first synthesis of the palliative care needs of adults with intellectual disabilities and the barriers and facilitators this population face in accessing palliative care. The current available evidence is almost exclusively reflective of the perspectives of health and social care staff, and the voices of people with intellectual disabilities and their families are lacking.

The palliative care needs identified in this review mirror those of the general population. The European Association for Palliative Care has also made this observation. However, meeting these needs is complicated by the challenges associated with this population. Understanding and recognition of these challenges for each individual, and anticipation of the problems they will face, are key to providing reasonable adjustments, a legal duty of all health and social care services that people with intellectual disabilities so deserve. Addressing education and communication barriers has potential to improve palliative care for this population. Closer attention is needed to how these can be addressed on a wider scale, with accompanying policies and guidelines to standardise practice. This needs to target staff at all levels, across both palliative care and intellectual disability services. Most studies in this review that evaluated interventions were small, and only provided information on the benefits to staff. Large scale interventional studies exploring the effectiveness of interventions in improving palliative care for people with intellectual disabilities are needed. Building links between palliative care and intellectual disability services is crucial. Involving family members and carers is also important in delivering individualised care, as this provides an advocate that knows the person well and can facilitate communication with health care professionals. However, they must act in collaboration with the individual with intellectual disability, involving them in decision making as much as possible, and facilitating their right to autonomy.
The WHO acknowledges the need to improve access to end of life care for hard to reach groups. Indeed, it has been stated that “how we care for the dying is an indicator of how we care for all sick and vulnerable people”. Many of the identified barriers and facilitators to accessing palliative care are likely to be encountered by other socially disadvantaged groups. The conceptual model presented in this paper may therefore be of use to service developers and policy makers in other areas. Thus policy makers have much to learn from acknowledging the barriers faced by this population.

**Limitations**

Only five electronic databases were searched. The hand search included only two intellectual disability specific journals and search of the grey literature was limited to two online databases. However, no date or geographical limitations were applied to the database search, which widened its scope. Although multiple researchers were involved in the screening process, EA conducted data extraction and grading of quality independently, presenting a source of bias. Yet the use of a predefined data extraction template and a structured critical appraisal tool acted to reduce subjectivity as much as possible. It was a challenge judging the quality of such a heterogenous group of studies with one tool, however, Hawker et al’s checklist is appropriate to appraise multiple study designs. Loosely defined populations meant in some cases it was difficult to separate participants that were eligible for inclusion alongside those that were not. As far as possible data was only cited where the source of the information was clear, and Figure 2 only includes participants that were clearly defined. The narrative approach used to synthesise the evidence may be seen as subjective and therefore open to bias. However, the use of NVIVO enabled a more systematic approach to this. The data predominantly reflect the experiences of health care professionals in high income countries so may not be generalisable to other settings.

**Recommendations for future research**

There is a need for high quality studies that not only describe the problems faced by people with intellectual disabilities near the end of life, but evaluate the benefits of specific interventions. The paucity of research in this area further disenfranchises an already marginalised group. Areas to focus on are education, communication and service development. Potential initiatives that could be evaluated in future studies include the effectiveness of toolkits, link nurses, and training days on patient care. Other approaches could be developing assessment tools such as the DisDAT and PALLI. However, this review demonstrates a lack of suitable and validated outcome measures for people with intellectual disabilities and palliative care needs. It is vital that these are developed in order to prove the effectiveness of proposed initiatives in improving care for this population.
Future research must involve people with intellectual disabilities and their family members as active participants. This will provide a deeper understanding of the inequalities experienced by this population and of their priorities and perspectives of what ‘good palliative care’ looks like. Research should also be encouraged on an international scale, and involve low and middle income countries. This would help in getting palliative care for people with intellectual disabilities on the global health agenda.

**Conclusion**

Addressing the inequalities in accessing palliative care for people with intellectual disabilities should be an urgent priority, particularly given the ageing population and concomitant co-morbidities. Currently, much of the empirical research in this area has focused on describing the problem. However, there are examples of good practice or pioneering initiatives that have potential to address inequalities in accessing palliative care. Such initiatives need to be evaluated through high quality, appropriately funded research that involves people with intellectual disabilities and their carers, as well as relevant health care professionals. Reliance on “committed individuals” or a “holistic philosophical approach” is not sustainable. Reasonable adjustments should be standard practice and not award-winning exceptions. Good practice in palliative care for this population needs to be standardised and implemented in all health care settings, across specialties and into mainstream services.

**Authorship**

All authors were involved in the development of the study question. EA registered the study with PROSPERO, performed the literature search, study selection, data extraction, quality assessment, data analysis and wrote the manuscript. ITW and KES participated in study selection. All authors reviewed earlier versions of the manuscript and read and approved the final manuscript.

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**Declaration of conflicting interests**

The Authors declare that there is no conflict of interest.
**Ethics**

As a systematic review, the study did not directly involve human participants and required no approval from an Ethics Committee or Institutional Review Board.

**Data management and sharing**

Further information is available at request from the first author.

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**Supplementary data: appendices**

| Appendix 1 | Example of search strategy: MEDLINE |
|------------|-------------------------------------|
| Appendix 2 | Data extraction table               |
| Appendix 3 | Hawker et al’s critical appraisal tool |
| Appendix 4 | Grading of quality                  |
Appendix 1: Example of search strategy: MEDLINE

Database: Ovid MEDLINE(R) and Epub Ahead of Print, In-Process & Other Non-Indexed Citations and Daily <1946 to June 11, 2019>

Search Strategy:

Palliative care:
1  exp Terminal Care/ or caregiver$.mp. or bereave$.mp. or inpatient.tw. or attitude to death.mp. or end of life.af. or hospice$.mp. or terminally ill.mp. or palliative$.mp. or Advance Care.af. or palliat.af. or advanced.af. or (morphine and cancer).af. or cancer pain.af. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, organism supplementary concept word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms] (841709)

Intellectual disability:
2  exp Intellectual Disability/ (92889)
3  exp Learning Disorders/ (21509)
4  (mental* or developmental* or intellectual* or cognitive or learning).ab,ti. (1093120)
5  (retard* or disab* or deficien* or delay*).ab,ti. (1185948)
6  4 and 5 (153599)
7  2 or 3 or 6 (227297)

Needs:
8  exp "Health Services Needs and Demand"/ (57615)
9  exp Needs Assessment/ (28555)
10  (needs assessment or assessment of healthcare needs or assessment of health care needs or unmet needs).ab,ti. (10359)

Access:
11  exp Health Services Accessibility/ (104713)
12  exp Healthcare Disparities/ (14484)
13  (access or access to care or barriers to care or obstacles or equity of care or inequity or inequality or allocation of health care resources or rationing or gate keeping or evaluation of care or quality of health care or assessment of need or unmet need or health care need or pattern of referral or referral).ab,ti. (420591)

Needs OR Access:
14  8 or 9 or 10 or 11 or 12 or 13 (570630)

Palliative care AND Intellectual disability AND [Needs OR Access]:
15  1 and 7 and 14 (894)

***************************
## Appendix 2: Data extraction table

| Author/date | Title | Published in | Country | Study design | Aim | Population | Setting | Needs | Facilitators | Barriers |
|-------------|-------|--------------|---------|--------------|-----|------------|---------|-------|-------------|----------|
| Bailey et al., 2016 | Surveying community nursing support for persons with an intellectual disability and palliative care needs | British Journal of Learning Disabilities | Ireland | Descriptive cross-sectional survey | To describe the provision of community nursing support for persons with an ID and palliative/end-of-life care needs. | Public Health Nurses (PHN), Community Nurses, Practice Nurses, Hospice at Home Nurses and Palliative Care Nurses (n=43) | 6 health service regions in Ireland | Information, advice, emotional support, symptom management, nursing care at home, nutrition management, pressure area care, referral to agency, hospice referral. | Family involvement/support, nursing experience of ID, teamwork/collaboration between care providers, support from ID services, personal attributes of professionals: compassion, communication, knowledge, honesty. Building a relationship with patient/family | Lack of cooperation from family, lack of experience/education/knowledge/communication skills, lack of co-ordination within/between services, delay in recognising end of life, delay in referral to PC, lack of ACP, poor referral information, lack of knowledge about patient. |
| Bekkema et al., 2014 | Respecting autonomy in the end-of-life care of people with intellectual disabilities: a qualitative multiple-case study | Journal of Intellectual Disability Research | Netherlands | Qualitative interviews | To describe how caregivers and relatives shape respect for autonomy in the end-of-life care for people with ID. | 16 relatives & 31 HCP involved in the care of 2 recently deceased patients with ID | 10 ID care provider organisations in different parts of the Netherlands | Information, autonomy, need to communicate wishes, involvement in decision making, physical care. | Knowledge/skills in end of life care: communication skills, recognising EOL care needs, ability to interpret symptoms. Education | Lack of HCP experience, knowledge and skills. |
| Bekkema et al., 2014 | Training needs of nurses and social workers in the end-of-life care for people with intellectual disabilities: A national survey | Nurse Education Today | Netherlands | Postal survey | To gain insight into the quality of end-of-life care, past vocational training, training needs and expert consultation opportunities of nurses and social workers working in ID care services. | Nursing staff (n=93), Social workers (n=37). | Working in care services for people with ID in the Netherlands. | Collaborative working between ID and SPC services. | Lack of training/education in EOL care. Lack of access to external experts i.e. SPC. |
| Birtford, 2004 | Status of End of Life Care in Organizations Providing Services for Older People with a Developmental Disability | American Journal on Mental Retardation | USA | Survey (paper & electronic questionnaire) | To establish a baseline of information on the current status of end-of-life care in organizations serving older people with an ID and their families. | 60 professionals providing services to older people with an intellectual disability and their families. | National organizations delivering a range of services to people with ID | Training staff, coordinated system of care in the community, availability of resources on EOL care for HCPs/family/carers/patients, policies & guidelines, ACP | Availability of direct care staff, Availability of nursing/medical staff funding, Staff untrained in EOL care, anxiety about responding to families, Concerns about liability. |
| Brown et al., 2013 | Please don't let It Happen on my Shift! Supporting Staff who are Caring for People with Learning Disabilties who are Dying | Tizard Learning Disability Review | UK | Qualitative: case series, interviews, observation of team meetings. | Not stated | Case series: 21 people with ID and terminal illness who had died. Interviews: 31 care staff, 16 service managers, 18 health care/ID professionals, 6 service users, 6 family members, 8 local community representatives, 1 external representative. Meetings: 5 staff meetings, 1 group of older people with ID. | 22 services that have cared for a person with ID during a terminal illness. Staffed housing (3), residential home (3), hospital (2), community (2), hospice (1), respite service (1). | Pain, care at home, information | Speech and language therapists to aid communication, relationships/knowing the patient, Diagnostic overshadowing, late diagnosis, difficulties communicating, reluctance to investigate, uncertain trajectory, lack of co-ordination between health & social services, absence of formal protocols, lack of clear legislation (e.g. consent, mental capacity, proxy decision makers), fear of disclosure/conflict, conflict between carers and family members | |
| Reference | Title | Country | Methodology | Summary |
|-----------|-------|---------|-------------|---------|
| Cartledge and Read, 2010 | Exploring the needs of hospice staff supporting people with an intellectual disability: a UK perspective | England | Qualitative: 2 focus group discussion | To identify what educational and preparation skills hospice staff felt were needed to enable them to feel confident in providing specialist palliative care to people with ID. |
| Cordingley et al., 2014 | End-of-life training for paid carers working with people with learning disabilities | England | Focus group discussion & evaluation forms | To implement and evaluate a study day for paid carers on end-of-life care for people with ID. |
| Cross et al., 2012 | Practical Approaches toward Improving End-of-Life Care for People with Intellectual Disabilities: Effectiveness and Sustainability | England | Staff training | To capture the learning from a 3 year project aimed at exploring ways of increasing access to palliative care services by people with ID within one geographical area. |
| Dunkley and Sules, 2014 | The challenges of providing palliative care for people with intellectual disabilities: a literature review | UK | Literature review | To explore the challenges of providing effective palliative care for people with ID. |
| Flynn et al., 2015 | Psychosocial experiences of chronic illness in individuals with an intellectual disability: A systematic review of the literature | UK | Systematic review | To identify, evaluate and synthesize literature exploring the psychosocial experiences of chronic illness in adults with ID. |
| Forrest-Jones et al., 2017 | People with intellectual disabilities at the end of their lives: The case for specialist care? | England | Mixed methods: observations, interviews, questionnaires, costs analysis | To explore the organizational context of a specialist palliative and end-of-life care home to see how it related to the quality of life of people with ID who were approaching the end of their lives. |

**Communication Needs:**
- Time: everything takes longer.
- Communication: patient, use of assessment tools.
- Communication needs: time, empathy, knowledge, experience.

**Diagnostic Overshadowing:**
- Delayed diagnosis, reluctance to investigate, use of complex language by health care professionals, placed in wards or homes for older people.
| Study (Year) | Title | Population | Methodology | Findings/Results |
|-------------|-------|------------|-------------|------------------|
| Friedman et al., 2012 | Unique and Universal Barriers: Hospice Care for Aging Adults with Intellectual Disability | USA | Literature review | To better understand the unique barriers to end-of-life care for aging adults with ID. |
| Gilbert et al., 2007 | Supporting people with intellectual disability in the cancer journey: The ‘Living with cancer’ communication pack | UK | Qualitative methods: telephone interview, focus groups, postal questionnaire | Use of a tool designed by experts, encompassing ‘total communication techniques’ to aid communication and discussion of sensitive issues. |
| Grindrod and Rumbold, 2017 | Providing end-of-life care in disability community living services: An organizational capacity building model using a public health approach | Australia | Mixed methods: online survey, focus groups, interviews | Staff attitudes: dying best managed by medically trained, not in my job role, death should be in hospital or nursing home. Lack of awareness of policy/protocol. Lack of training/ experience. Inadequate staffing levels. Patients not offered choices. Absence of coordination between ID staff & SPC. Juggling needs of multiple stakeholders. Staff stress/ feeling unsupported. Fear of impact on other residents; grief/ resources taken up by dying. Absence of confident formal leadership. Inconsistent practice/policy between settings. |
| Hunt et al., 2019 | End-of-life care in intellectual disability: a retrospective cross-sectional study | UK | Retrospective cross-sectional survey | Patient lack of awareness that they were dying. Preferred place of death not recorded. Lack of involvement in their own care. Hospital-based care inferior to community-based care with lower levels of support from external services. |
| Authors                        | Title                                                                 | Journal                                    | Country | Methodology | Participants | Data Collection | Findings                                                                                                                                  | Implications/ Limitations                                                                 |
|-------------------------------|----------------------------------------------------------------------|--------------------------------------------|---------|-------------|--------------|----------------|------------------------------------------------------------------------------------------------------------------------------------------|------------------------------------------------------------------------------------------|
| Kim and Gray, 2018            | Palliative Care Experiences and Needs of Direct Care Workers          | Journal of Palliative Medicine            | USA     | Focus groups | 54 DNs       | To explore direct care workers experience with PC and their perceived challenges. Direct care workers serving people with ID (n=54) | Non-profit community-based organisations providing residential and day services for people with ID in urban and suburban areas within or surrounding a large Midwestern city. | Empathy. Extra attention. Adaptation in care/ taking more time. Peer support/ teamwork. Difficulties in communication with patient. Anxiety in anticipating death. Limited organisational resources or support; understaffing; inadequate communication between staff of different grades/ specialities, insufficient training. |
| Kinley et al., 2018            | Development and implementation of the Steps to Success Palliative Care Programme in residential care homes for people with a learning disability in England | International Journal of Palliative Nursing | England | Literature review, meetings with ID, SPC & care home staff. | 12 DNs       | To develop and implement a palliative care programme that would meet the needs of people with a ID, their families and care home staff. | Care home, ID and SPC Staff. 30 care homes | Delivery of an evidenced based formal staff education programme: improved identification/ assessment/ symptom management/ ACP/ changed attitudes towards EOL. Collaboration between services. Learning at an individual, organisational and systems level. Ongoing facilitation/ training new staff to achieve sustainability. |
| Lindop and Read, 2000          | District nurses' needs: palliative care for people with learning disabilities | International Journal of Palliative Nursing | England | Focus groups, questionnaires | 12 DNs, 106 DNs | Identify national PC issues, and current PC services provided by DNs for people with ID in North Staffordshire, UK. | Focus groups:12 DNs undertaking a diploma in PC. Questionnaires: 106 DNs (54% had contact with ID patients with PC needs). | North Staffordshire, DNs in community | Pain, information, care in the community. Education and training of district nurses. Communication difficulties. Lack of experience/training. |
| Marriott et al., 2013          | Good practice in helping clients cope with terminal illness           | Learning Disability Practice               | England | Case study | Male in 50s with lung cancer and mild ID | To provide an example of good practice in end of life planning and palliative care for a person with ID. Make in SIs with lung cancer and mild ID. | England: hospital & community. Dietary & nutrition needs. Assistance with complicated medication regime. Pain. Wish to die at home. Thorough and timely investigation. Allowing enough time for appointments to ensure understanding. Translating medical terminology into simpler concepts to maximise involvement in decisions. Continuity of care with same hospital consultant. Allocation of CLOT nurse. Collaboration between services. Family involvement. Building relationships with patient. Anticipating future needs/ACP. Documenting wishes. Regular GP & DN visits. GP as key player. | |
| McKechnie, 2006               | What does the literature tell us about death, dying and palliative care for people with intellectual disabilities? | Progress in Palliative Care               | New Zealand | Literature review | Not stated | Not stated | Not stated. | Difficulties in detecting disease in people who are non-verbal, diagnostic overshadowing, delayed diagnosis. Lack of involvement of the individual in their care: decisions being made for them by family members or health practitioners. Inadequate education of formal and informal carers. Changing healthcare needs/ disease profiles as people with ID are living longer. Health professionals making assumptions about understanding/ ability. Legal barriers around informed consent, capacity and competence. | |
| Authors          | Title                                                                 | Country | Research Type | Setting                          | Participants                                                                    | Methods                                                                 | Findings                                                                 |
|------------------|------------------------------------------------------------------------|---------|---------------|----------------------------------|--------------------------------------------------------------------------------|----------------------------------------------------------------------|--------------------------------------------------------------------------|
| McLaughlin et al.| Developing a best practice model for partnership practice between specialist palliative care and intellectual disability services: A mixed methods study | UK      | Questionnaires & interviews | To develop a best practice model which would guide and promote partnership practice between specialist palliative care and ID services | Questionnaires: ID services (n=10), SPC services (n=22), ID nursing homes (n=15). Interviews: ID HCPS who had provided EOL care to adults with ID. Primary and secondary care in one region of the UK. Partnership working between ID and SPC: easy to contact, regional meetings, joint study days, joint working/shadowing in both clinical areas. Co-ordination and continuity of care, familiarity with the patient. Educating, supporting and empowering staff. The role of the GP: early identification of need and specialist referral. Medical versus ‘social’ model of care; time constraints, limited/late referrals to SPC, misalignment/conflict between services. |                                                                 |                                                                 |
| McNamara et al.  | Creating person-centred support for people with intellectual disabilities at the end of life: An Australian qualitative study of unmet needs and strategies | Australia | Interviews | Use the perspectives of health professionals and paid carers to document the range of needs adults with an ID experience during the last months of their lives | Nurses (9), social workers (4), occupational therapists (3), Disability residential accommodation managers (6), disability support workers (2), psychologists (2) Community PC organization, community nursing organization, Disability accommodation services, public hospital, country health service. Western Australia. | KC P. Enabled to die in familiar surroundings: dying at home. Full time (24/7) care as illness progresses. Pain. Symptom control. Physio & OT needs: repositioning/wheelchairs. Person-centred care: awareness of social/ethnic background, personal history, and modes of communication. Opportunities for choice, religious/spiritual needs. Nutrition. Open communication with the person and their family. Good staff support and bereavement care. Familiarity with patient. Collaboration between MDT. Continuity of HCP. Greater openness/awareness & strong advocacy. Identify case coordinator/manager, shared care between services, training ID staff in PC/training PC staff in ID. Available PC guides/policies, routine use of disability-oriented pain/symptom assessment tools, early identification & referral to PC. Use speech therapists to assist with interpretation. Communication aids and electronic devices. Accompanied by staff/family during hospital stays. Reduce transitions and create familiar environments. GP as key player. | Paternally protecting other residents from grief. Poorly trained staff. High staff turnover. Communication issue. Diagnostic overshadowing. Lack of communication between hospital & community services. Lack of ACP = unnecessary ED admissions. Lack of funding. People with ID ‘hidden’ by society. |   |
| Moro et al., 2017 | Agency, social and healthcare supports for adults with intellectual disability at the end of life in out-of-home, non-institutional community residences in Western Australia: A literature review | USA     | Literature review | To summarise the literature on the agency, social and healthcare supports that impact the care of adults with ID at the end of life | Adults with ID/end of life care needs and those caring for them | All health & social care settings | Transitioning needs: home/residential care/nursing home/hospital. Expressing & documenting wishes. Care at home. Information. Help to understand diagnosis & prognosis. Proximity of family accommodated. | Time to build relationships between staff, patient and family. Partnerships between ID and PC. Education. Lack of staff training. Collusion. Proxy decision making. Reluctance to investigate/late diagnosis. Under RND over treatment. Transportation barriers and lack of accessible medical equipment in hospitals: hindering admission for medical care. Poor communication/negative rapport with patient/careers. |   |
| Morton-Nace & Schaler, 2013 | End of life care for people with a learning disability | England  | Face to face interviews | To explore the experiences of ID and District nurses caring for people with a LD at the end of their lives | Community learning disability nurses (n=3), District nurses (n=3), all with experience of providing care for people with ID at EOL. | Community | Being empowered to make choices. Effective and dignified management of symptoms. Physical comfort. Physical and mental pain. Time. Collaborative working between teams/services. Person-centredness, planning, preparation, outreach and sharing of information with other HCPS. Training in EOL care and policy. Emotional support for staff and residents. | Staff attitudes: unwillingness to put in extra time, complacency, divide between services, protective/paternal leading to withholding of info. Specific choices/decisions not documented creating conflict/preventing collaborative working. Patient non-compliance with medication. Diagnostic overshadowing. Being reactive rather than proactive. Fragmented communication between HCPS/patient/careers. Limited sharing of info. Patients’ inability to communicate their needs. Inexperience, lack of understanding/skills training. Discomfort when |   |
| Reference | Year | Title | Journal | Country | Methodology | Findings |
|-----------|------|-------|---------|--------|-------------|---------|
| Ng and Li, 2003 | | A survey exploring the educational needs of care practitioners in learning disability (LD) settings in relation to death, dying and people with learning disabilities | European Journal of Cancer Care | England | Questionnaires | To explore whether care practitioners have the knowledge and skills to care adequately for dying people with LD in residential settings. 35 qualified care practitioners working in residential settings. 15 residential care homes, 5 NHS Trusts in England. Wound care. Cultural awareness. Care after death. Support for family. Verbal and non-verbal communication skills. Education in EOLC skills. Updating nursing curriculum. |
| Northway et al., 2019 | | Nursing care at end of life: a UK-based survey of the experiences of support workers caring for people with learning disabilities | Journal of Research in Nursing | UK | Questionnaire | What are the levels of support provided by nurses in the last months of life for people with ID? 88 ID care providers in the UK. Social care residential setting or nursing home for people with ID. Training and education targeted towards community and hospital-based nursing staff: they were shown to have significant involvement in the care of people with ID in last months of life. |
| O'Regan and Drummond, 2008 | | Cancer information needs of people with intellectual disability: A review of the literature | European Journal of Oncology Nursing | Ireland | Literature review | A review of the literature with a specific focus on the cancer information needs of people with an ID. Adults with ID and cancer, those caring for them. All health & social care settings. Cancer information materials that can be accessed and understood by people with an ID. Poor communication skills. Social absences assumption they will not comprehend resulting in withholding information regarding diagnosis and prognosis. |
| O'Sullivan and Harding, 2017 | | Transition: the experiences of support workers caring for people with learning disabilities towards the end of life | BMJ Supportive & Palliative Care | England | Focus group | To provide a better understanding of the experience of support workers, as paid carers, caring for adults with LD nearing the end of life in residential settings. 3 residential support workers with direct experience of caring for a person with LD towards the end of life. 6 independent service provider settings for people with ID in London. Collaboration with other services. Partnership between ID and SPC. Support from management within own organisation. Adequate staffing at night. Collaboration/relationship with family. Early involvement of SPC. Education and training. Emotional support for staff. Bereavement. Denial and a lack of preparedness for seeing residents entering the dying phase of their lives. Lack of access to extra training and support. Lack of organisational policy/guidelines. |
| Study | Title                                                                 | Journal/Media | Country | Design | Participants & Intervention | Findings/Implications                                                                 |
|-------|----------------------------------------------------------------------|---------------|---------|--------|-----------------------------|--------------------------------------------------------------------------------------|
| Reddall, 2010 | A palliative care resource for professional carers of people with learning disabilities | European Journal of Cancer Care | England | Case study | F with ID and brain tumour | Day hospice: social contact with others. IPU: symptom assessment or management, despite. Spiritual needs. Project worker acted as conduit between generic end-of-life care and ID services. Fulfilment of any last wishes to augment a more peaceful death. MDT approach, best interest meeting. |
| Reddall, 2010 | A palliative care resource for professional carers of people with learning disabilities | European Journal of Cancer Care | England | Case study | 2 M with ID & prostate Ca | Regular contact with GP. Medication needs. Loss of appetite. Declining mobility. Urinary incontinence. Pain. Oral thrush. Fear of strangers. A desire to stay at home. Provision of easy to understand & easily accessible booklet on palliative care for staff in a learning disability care home. Presence of key worker that knows patient for consultations & scans. Interdisciplinary working, shared expertise of ID and SPC. Community team involvement: DNs & Macmillan nurse. MDT reflective session after death. |
| Ronneberg et al., 2015 | Promoting Collaboration Between Hospice and Palliative Care Providers and Adult Day Services for Individuals with Intellectual and Developmental Disabilities | Journal of Death and Dying | USA | Online surveys & focus groups | Surveys: ID service providers (n=103), hospice/PC providers (n=87). Focus groups: adult day service providers (n=10), hospice/PC providers (n=7). | Education around recognition of nonverbal symptoms of pain. Well-coordinated services, knowledgeable about one another. Lack of established relationships between ID and hospice services. Few clients receiving dual (hospice and ID) services. Need for training and education for both hospice and ID services. Lack of clearly established and communicated DNR policies, lack of support/guidance from state government around DNR policies. Misconception that PC patients must always be transferred to hospice for care. Hospice providers limited experience in working with persons with ID. Confusion around funding and regulations. Difficulty having client with extensive medical needs. Staff/family attitudes about death and dying/resistance to hospice philosophy. |
| Ryan et al., 2016 | The nature and importance of quality of therapeutic relationships in the delivery of palliative care to people with intellectual disabilities | BMJ Supportive & Palliative Care | Ireland | Focus groups | One Health Service Executive area in Ireland | Familiarity, time allowed to build relationships with new HCP. Pain. Importance of patient-professional relationship: building a therapeutic relationship. Continuity of relationship, knowing the individual, building trust. ID staff familiarity with patient: getting to know them over time. Strengthening interface between PC & ID services. PC staff unfamiliarity with patients with ID. Over-reliance on carers for communication/ information/ 3 way relationship: prevented PC staff building proper therapeutic relationship. PC staff failure to form authentic relationships with people with ID = poorer quality of care, harder to overcome collusion from family. |
| Reference                          | Title                                                                 | Country | Study Design  | Methods                                                                 | Findings/Implications                                                                 |
|-----------------------------------|-----------------------------------------------------------------------|---------|---------------|--------------------------------------------------------------------------|---------------------------------------------------------------------------------------|
| Ryan and McQuillan, 2005          | Palliative care for disadvantaged groups: supplementary intellectual   | Ireland | Literature review | Review of the palliative care needs of people with ID and PC needs, those caring for them. | Palliative care guidelines do not universally consider the needs of people with ID. Lack of validated tools or those designed specifically for people with ID. |
| Sampson et al., 2015              | European palliative care guidelines: how well do they meet the needs  | Europe  | Questionnaires | To review whether national palliative care guidelines in Europe consider pain assessment and management for the increasing numbers of adults with ID | Palliative care guidelines do not universally consider the needs of people with ID. Lack of validated tools or those designed specifically for people with ID. |
| Nin et al., 2008                   | Providing Palliative Care to People with Intellectual Disabilities,  | USA     | Survey        | To document the degree to which hospice and palliative care services were provided to New Jersey residents with ID, and the challenges in providing this care | Palliative care guidelines do not universally consider the needs of people with ID. Lack of validated tools or those designed specifically for people with ID. |
| Sue et al., 2019                   | Palliative care for patients with communication and cognitive         | Canada  | Literature review | To explore recent best evidence to address PC needs of people with ID. To help increase the confidence of caregivers and clinicians in providing PC to adults with ID using an easy-to-follow framework. | Palliative care guidelines do not universally consider the needs of people with ID. Lack of validated tools or those designed specifically for people with ID. |
| Authors | Title | Country | Research Design | Aim | Conclusions |
|---------|-------|---------|----------------|-----|-------------|
| Tuffrey-Wijne, 2003 | The palliative care needs of people with intellectual disabilities: a literature review | England | literature review | To answer the question: What are the palliative care needs of people with ID? | People with ID and PC needs, those caring for them. |
| Tuffrey-Wijne et al., 2007a | End-of-Life and Palliative Care for People with Intellectual Disabilities Who Have Cancer or Other Life Limiting Illness: A Review of the Literature and Available Resources | UK & Netherlands | literature review | To review the literature around the need of people with ID for palliative care | Adults with ID & PC needs, NH homes & settings. |
| Tuffrey-Wijne et al., 2007b | Palliative care provision for people with intellectual disabilities: interviews with specialist palliative care professionals in London | England | interviews | To identify the issues affecting the delivery of services by specialist palliative care professionals to people with ID in London | PC professionals & hospices in Greater London, representing urban as well as rural community. Personal care needs: washing, etc. (independence inhibited by illness). Symptom management = difficult to interpret individual pain behaviours. Emotional support for family. Community care to avoid change of environment. Mental health problems. |

Concerns:
- Patient's life. Unclear trajectories, unexpected recovery after deterioration – difficult to prognosticate. Patient: Difficulty understanding abstract concepts relating to death.
- Patient's ability to consent. Difficulty obtaining consent, assuming patients lack ability to consent.
- Diagnostic overshadowing: difficulty identifying symptoms, resulting in advanced disease with severe symptoms. Lack of ID staff experience/training in caring for dying. Communication difficulties hindering recognition of pain: reliance on carers. \(n=32\). (Only 24 had seen patients with ID within PC). Patient: Difficulty recognizing illness. Patient: Unclear trajectories, unexpected recovery after deterioration – difficult to prognosticate. Patient: Difficulty understanding abstract concepts relating to death.
| Tuffrey-Wijne et al., 2008 | Palliative care provision for people with intellectual disabilities who have cancer: a questionnaire survey of specialist palliative care professionals | Palliative Medicine | England | Postal questionnaire | To gain insight into the current use of PC services by people with ID in London, to identify the prevalence and severity of problems faced by PC professionals, to identify strategies that will facilitate an improvement in access and delivery of PC services to people with ID. | PC professionals providing PC services for adults (n=543). Of these, 67% (n=364) had previously supported a patient with ID. | 2 SPC services in London: hospice, community & hospital setting. | Symptom control, Terminal care, Support for carers/ family, Psychological support, Hospice care, Day care. Fear, Pain, Emotional distress. Time, Familiarity. | Collaboration between services: Allocating link worker within each service. Formal training. Allowing time. Providing safe & predictable environment with familiar faces. Staff support. Family support. | End of life care. Diagnosis. Lack of referral to SPC. Fear of transfer to different setting if referred. Family protection/fear of PC. Lack of awareness by both SPC and ID services of each other’s existence and what each has to offer. Communication difficulties: understanding patient, symptom assessment, consent. Non-compliance with medication/care/ examination. Poor communication with family/carer. Patients’ difficulty with comprehending their illness and treatment. Those that spend most time with patient had least training e.g. HCAs. Lack of background information. Difficulty finding suitable long term accommodation when unable to cope at home. Financial concerns. |
| Tuffrey-Wijne and McElnay, 2008 | Communication difficulties and intellectual disability in end-of-life care | International Journal of Palliative Nursing | England | Literature review + drawing on 3 research studies conducted by the authors. | To map the communication difficulties experienced by people with ID within a PC setting. | People with intellectual disabilities, HCP & carers within a PC setting. | All health & social care settings. | Information, Honesty, Time. Difficulty understanding abstract concepts. | Use of communication aids: Objects of reference, sign languages, symbol-based systems. Use of simple, non-supernumerary language, avoidargon. Allowing extra time for consultations, addressing patient before carer. | Communication. Inability of patient to communicate verbally. Inability of HCP to confirm comprehension of diagnosis/treatment. Family collusion. |
| Tuffrey-Wijne et al., 2009 | People with learning disabilities who have cancer: an ethnographic study | British Journal of General Practice | England | Ethnographic: participant observation | To explore the experiences and needs of people with ID who have cancer. | Adults with ID and a cancer diagnosis (n=13) | Home, residential care home. | Lives defined by dependence on carers. Particularly vulnerable when ill. Anxiety precipitated by not understanding. Fear of cancer. Pain. Continuation of clear safe routines, treasured activities, relationships. Loneliness, isolation. | Preparation for appointments, family/carer present. Explanation of procedures/treatments in a way that is easy to understand. Familiarity with hospital/clinic. Maintaining routine. HCP becoming integrated into safe/trusted environment: a familiar face. Practical and emotional support for residential staff. | Diagnosis dependant on someone other than the patient noticing something is wrong or correctly interpreting behavioural changes: but often unnoticed or misinterpreted. Patient complaints ignored or disbelieved; attributing to ‘attention seeking’ behaviour. Medical staff guided by opinion of carers, without attempting to check views of patient. Not offered standard treatment options due to assumption that the person would not cope. People with ID unlikely to question treatment decisions. Information giving influenced by: personal opinion of the carer, desire to protect the person from distress and lack of skill and confidence in communicating with them. Family collusion. Acquiescence. HCP lack of experience: communication, fear of opioids, fear of death. Reluctance to ask busy doctors for more input. |
| Authors | Title | Country | Methodology | Phase 1 | Phase 2 | Phase 3 | Phase 4 |
|--------|-------|---------|-------------|--------|---------|---------|---------|
| Tuffrey-Wijne, 2013 | A new model for breaking bad news to people with intellectual disabilities | Palliative Medicine, England | 2 phase qualitative study: (1) focus group meetings, on-line focus groups, interviews; (2) structured feedback. | To develop a model for breaking bad news that meets the needs of people with ID. | To develop a model for breaking bad news that meets the needs of people with ID. | To develop a model for breaking bad news that meets the needs of people with ID. | Not stated. |
| Tuffrey-Wijne et al., 2016 | Defining consensus norms for palliative care of people with intellectual disabilities in Europe, using Delphi method: A White Paper from the European Association of Palliative Care | Palliative Medicine, Europe | Delphi study using MM. | To define consensus norms for palliative care of people with ID in Europe | Professionals with expertise in PC, ID or both (n=80). | 5 European countries, all health & social care settings. | Communication needs. Holistic approach: physical, emotional, social and spiritual needs. Counselling, support in maintaining social links with friends. Pain. Comorbidities. Right to live and a right to recognition of the value of their lives. Complicated grief. Process of disclosing bad news should involve collaboration between those who understand the content of the information (health care professionals) and those who best understand the patient’s communication needs (family). Use of a model for breaking bad news to people with ID. Emotional support for staff who may also be affected by bad news: in turn helps to support the patient. | |
| Tuffrey-Wijne et al., 2017 | Communicating about death and dying: Developing training for staff working in services for people with intellectual disabilities | Journal of Applied Research in Intellectual Disabilities, England | Stage 1: 3 semi-structured interviews. Stage 2: Development & evaluation of training course - feedback forms and questionnaires. | To assess staff training needs and to develop, trial and evaluate a training course on communicating about death and dying. | Stage 1: 20 staff working in ID residential or supported living services (13 had supported people with ID who had died in the past 6 months). Stage 2: 114 interprofessional delegates, 108 completed feedback forms and questionnaires + 24 follow-up questionnaires at 3 months. | 8 different ID residential or supported living services in London, UK. | Organisational culture: junior staff felt more comfortable talking about loss, death and dying in settings where managers talked with staff and clients about death, providing positive role modelling. Staff training: to improve the ability to talk about death and dying, not only with people with ID but also with each other. World Café approach: helping course delegates to talk about death and dying and “role-modelling” ways in which they could help their teams do the same. Involving people with ID by UK ID services: workforce, especially senior care staff, is largely unchanged. Staff often learn on the job. Staff frightened and unprepared for dealing with issues of death and dying, avoidance of death-related conversations, concerned that communicating about death might cause distress, partly due to their own lack of skill. Not knowing how to break bad news. Avoidance of death-related communication had roots in organisational culture. Tendency among staff to “keep things positive.” Lack of preparation and training for the job of care worker was striking. Lack of | Diagnostic overshadowing. |
Tuffrey-Wijne and Davidson (2018) Excellence in palliative and end-of-life care provision for people with intellectual disability.  International Journal of Palliative Nursing, 1(4), 194-200.  Thematic content analysis  To identify the key characteristics of practice initiatives and case reports that have won a UK award for the outstanding provision of end-of-life care for people with ID (2008–2018).  People, projects or teams who were award-winners or highly commended (n=25).  Individual and organisational commitment: Enthusiastic individuals/leaders going beyond the call of duty. Adequate funding and policies. Working together in collaboration: between LD and PC, with patients, carers, families. Regular meetings. Mutual learning. Availability of required professionals, 24/7 care, continuity of care. The person’s story is at the heart of care: Highly individualised care, creative & flexible, familiarity with patient, maintaining care in home environment, involving & supporting family. Developing tools and staff training: Interprofessional, regular, compulsory conferences, use of GSF, tools and pathways, early referrals, ACP, adapting from mainstream. Lack of national policies and adequate funding = postcode lottery, good initiatives not sustained when staff leave. “Good palliative care for people with ID should not be dependent on the fortunate presence of committed professionals with a passion for this area of care, or on being within the catchment area of one of these inspirational projects and services. Nor should it be up to a small charitable organisation like the PCPLD Network…”

Vrijmoeth et al., 2016a) Physician-Reported Symptoms and Interventions in People with Intellectual Disabilities Approaching End of Life.  Journal of Palliative Medicine, 15(11), 797-805.  Online retrospective survey  To determine the prevalence rates of physician-reported symptoms at the moment they recognise patient’s death in the foreseeable future.  0 physicians who had provided PC to a deceased patient with ID (n=81).  All health care settings, Netherlands  Physical/symptom needs: fatigue, drowsiness, decreased appetite, shortness of breath, pain, anxiety, depressed mood, nausea, constipation, delirium. Medical needs: sedation, wound care, antibiotics, artificial nutrition & hydration, surgery, chemo & radiotherapy, blood transfusion.  MDT approach to symptom control, use of proxies who know patient well, use of symptom assessment tools.

Vrijmoeth et al., 2016b) Physicians’ recognition of death in the foreseeable future in patients with intellectual disabilities.  Journal of Intellectual Disability Research, 61(13), 1419-1436.  Online retrospective survey  To explore when and based on which information sources and signals physicians recognised that a patient with ID would die in the foreseeable future.  0 physicians (n=41) & 3Ps (n=18) who had provided PC to a deceased patient with ID.  All health care settings, Netherlands  Close attention to changes in characteristic behaviour or mood. Gathering information collaborating with caregivers & family. Use of tools.  Physician delayed recognition of dying phase in people with ID & delayed discussion of PC until time when death is foreseeable. Diversity in signals that lead to recognition of death. Patient inability to communicate.
| Reference | Title | Country | Methodology | Purpose | Setting | Other Notes |
|-----------|-------|---------|-------------|---------|---------|-------------|
| (Welch et al., 2017) | Challenges in providing end-of-life care for people with intellectual disabilities: Health services access | Australia | Focus group interviews | To explore staff perceptions of the strengths and barriers associated with health service access for people with ID at the end of life. | Rural & 2 metropolitan locations, NSW & Queensland | Interviews; ID nurses (n=14), doctors (n=81), GPs (n=16), IDPCCs (n=185). |
| (Wagemans et al., 2013) | End-of-life decisions for people with intellectual disabilities, an interview study with patient representatives | Netherlands | Qualitative interviews | To clarify the process of end-of-life decision making for people with intellectual disabilities from the perspective of patient representatives. | All health care settings, Netherlands | Interviews: ID-physicians (n=81), ID-interns (n=10), ID-nurses (n=10), DCPs (n=14). Focus groups: ID-physicians (n=11), GPs (n=5), ID-nurses for people with ID (n=10), DCPs (n=14). Pilot: adults with ID, likely to be in last year of life (n=18). |
| (Vrijmoeth et al., 2018) | Development and applicability of a tool for identification of people with intellectual disabilities in need of palliative care (PALLI) | Netherlands | Mixed-methods: online retrospective survey, interviews, focus groups, prospective pilot survey | To describe the development of PALLI and to explore its applicability as reported by healthcare professionals. | All health care settings, Netherlands | Use of a tool to identify and assess people with ID who are in need of PC. Includes signals relevant and characteristic to people with ID. Can be completed by physician or carer proxy. |
| (Leguizamon et al., 2016) | Moreno | | | | | |
Integrating views of service users into models of care. Time/patience/flexibility. Evidence-based care.

Communication difficulties with patient: ascertaining information requirements, wishes, symptoms.

Abbreviations:
ACP=advance care planning, BBN=breaking bad news, Ca=cancer, CLDT=community learning disability team, CP=cerebral palsy, DCPs=daily care professionals, DNR=do not resuscitate, DNs=district nurses, ED=emergency department, EOL=end of life, F=female, GSF=gold standards framework, H@H=hospice at home, HCA=health care assistants, HCP=health care professionals, ID=intellectual disability, info=information, M=male, MDT=multidisciplinary team, MM=mixed methods, NH=nursing home, NHL=Non-Hodgkin Lymphoma, PC=palliative care, PPD=preferred place of death, Pts=patients, SPC=specialist palliative care
### Appendix 3: Hawker et al’s critical appraisal tool

1. **Abstract and title:** Did they provide a clear description of the study?
   - **Good** Structured abstract with full information and clear title.
   - **Fair** Abstract with most of the information.
   - **Poor** Inadequate abstract.
   - **Very Poor** No abstract.

2. **Introduction and aims:** Was there a good background and clear statement of the aims of the research?
   - **Good** Full but concise background to discussion/study containing up-to-date literature review and highlighting gaps in knowledge.
   - **Fair** Some background and literature review.
   - **Poor** Some background but no aim/objectives/questions OR Aims/objectives but inadequate background.
   - **Very Poor** No mention of aims/objectives.

3. **Method and data:** Is the method appropriate and clearly explained?
   - **Good** Method is appropriate and described clearly (e.g., questionnaires included).
   - **Fair** Method appropriate, description could be better.
   - **Poor** Questionable whether method is appropriate.
   - **Very Poor** No mention of method, AND/OR Method inappropriate, AND/OR No details of data.

4. **Sampling:** Was the sampling strategy appropriate to address the aims?
   - **Good** Details (age/gender/race/context) of who was studied and how they were recruited.
   - **Fair** Sample size justified.
   - **Poor** Sampling mentioned but few descriptive details.
   - **Very Poor** No details of sample.

5. **Data analysis:** Was the description of the data analysis sufficiently rigorous?
   - **Good** Clear description of how analysis was done.
     - Qualitative studies: Description of how themes derived/respondent validation or triangulation.
     - Quantitative studies: Reasons for tests selected hypothesis driven/numbers add up/statistical significance discussed.
   - **Fair** Qualitative: Descriptive discussion of analysis.
| Rating | Description |
|--------|-------------|
| Quantitative. Poor | Minimal details about analysis. |
| Quantitative. Very Poor | No discussion of analysis. |
| Ethics and bias: Have ethical issues been addressed, and what has necessary ethical approval gained? Has the relationship between researchers and participants been adequately considered? Good | Ethics: Where necessary issues of confidentiality, sensitivity, and consent were addressed. Bias: Researcher was reflexive and/or aware of own bias. |
| Ethics and bias: Have ethical issues been addressed, and what has necessary ethical approval gained? Has the relationship between researchers and participants been adequately considered? Fair | Lip service was paid to above (i.e., these issues were acknowledged). |
| Ethics and bias: Have ethical issues been addressed, and what has necessary ethical approval gained? Has the relationship between researchers and participants been adequately considered? Poor | Brief mention of issues. |
| Ethics and bias: Have ethical issues been addressed, and what has necessary ethical approval gained? Has the relationship between researchers and participants been adequately considered? Very Poor | No mention of issues. |
| Results: Is there a clear statement of the findings? Good | Findings explicit, easy to understand, and in logical progression. Tables, if present, are explained in text. Results relate directly to aims. Sufficient data are presented to support findings. |
| Results: Is there a clear statement of the findings? Fair | Findings mentioned but more explanation could be given. Data presented relate directly to results. |
| Results: Is there a clear statement of the findings? Poor | Findings presented haphazardly, not explained, and do not progress logically from results. |
| Results: Is there a clear statement of the findings? Very Poor | Findings not mentioned or do not relate to aims. |
| Transferability or generalizability: Are the findings of this study transferable (generalizable) to a wider population? Good | Context and setting of the study is described sufficiently to allow comparison with other contexts and settings, plus high score in Question 4 (sampling). |
| Transferability or generalizability: Are the findings of this study transferable (generalizable) to a wider population? Fair | Some context and setting described, but more needed to replicate or compare the study with others, PLUS fair score or higher in Question 4. |
| Transferability or generalizability: Are the findings of this study transferable (generalizable) to a wider population? Poor | Minimal description of context/setting. |
| Transferability or generalizability: Are the findings of this study transferable (generalizable) to a wider population? Very Poor | No description of context/setting. |
| Implications and usefulness: How important are these findings to policy and practice? Good | Contributes something new and/or different in terms of understanding/insight or perspective. Suggests ideas for further research. Suggests implications for policy and/or practice. |
| Implications and usefulness: How important are these findings to policy and practice? Fair | Two of the above (state what is missing in comments). |
| Implications and usefulness: How important are these findings to policy and practice? Poor | Only one of the above. |
| Implications and usefulness: How important are these findings to policy and practice? Very Poor | None of the above. |
## Appendix 4: Grading of quality

**Hawker's 9 item checklist:**

1. **Abstract and title:** Did they provide a clear description of the study?
2. **Introduction and aims:** Was there a good background and clear statement of the aims of the research?
3. **Method and data:** Is the method appropriate and clearly explained?
4. **Sampling:** Was the sampling strategy appropriate to address the aims?
5. **Data analysis:** Was the description of the data analysis sufficiently rigorous?
6. **Ethics and bias:** Have ethical issues been addressed, and what has necessary ethical approval gained? Has the relationship between researchers and participants been adequately considered?
7. **Results:** Is there a clear statement of the findings?
8. **Transferability or generalizability:** Are the findings of this study transferable (generalizable) to a wider population?
9. **Implications and usefulness:** How important are these findings to policy and practice?

**Total score:**

- 18 or less = poor
- 19 to 27 = moderate
- > 27 = good

| Study                          | Item 1 | Item 2 | Item 3 | Item 4 | Item 5 | Item 6 | Item 7 | Item 8 | Item 9 | Total | Overall grade |
|--------------------------------|--------|--------|--------|--------|--------|--------|--------|--------|--------|-------|---------------|
| Bailey et al. 2016             | 3      | 4      | 4      | 4      | 2      | 4      | 3      | 4      | 3      | 30    | Good          |
| Bekkema et al. 2014a           | 3      | 4      | 3      | 2      | 4      | 2      | 4      | 3      | 3      | 27    | Moderate       |
| Bekkema et al. 2014b           | 4      | 3      | 4      | 3      | 4      | 3      | 3      | 4      | 3      | 31    | Good          |
| Botsford 2004                  | 3      | 3      | 3      | 2      | 2      | 2      | 3      | 2      | 3      | 23    | Moderate       |
| Brown et al. 2003              | 2      | 2      | 2      | 3      | 2      | 2      | 3      | 3      | 2      | 21    | Moderate       |
| Cartlidge 2010                 | 4      | 4      | 4      | 3      | 3      | 4      | 3      | 3      | 3      | 31    | Good          |
| Codling et al. 2014            | 3      | 4      | 3      | 3      | 3      | 1      | 3      | 3      | 3      | 26    | Moderate       |
| Cross et al. 2012              | 3      | 3      | 2      | 2      | 1      | 3      | 3      | 3      | 3      | 22    | Moderate       |
| Dunkley & Sales 2014           | 3      | 3      | 4      | 4      | 3      | 3      | 4      | 4      | 4      | 32    | Good          |
| Flynn et al. 2015              | 3      | 3      | 4      | 3      | 3      | 3      | 3      | 2      | 2      | 26    | Moderate       |
| Forrester-Jones et al. 2017    | 4      | 3      | 4      | 3      | 4      | 4      | 3      | 3      | 3      | 31    | Good          |
| Friedman et al 2012            | 3      | 3      | 3      | 3      | 3      | 3      | 3      | 4      | 3      | 28    | Good          |
| Gilbert et al. 2007            | 1      | 1      | 3      | 2      | 2      | 3      | 2      | 2      | 2      | 18    | Poor          |
| Grindrod & Rumbold 2017        | 4      | 3      | 3      | 3      | 4      | 2      | 4      | 3      | 4      | 30    | Good          |
| Hunt et al 2019                | 4      | 4      | 4      | 4      | 4      | 1      | 4      | 3      | 3      | 31    | Good          |
| Kim & Gray 2018                | 4      | 3      | 3      | 4      | 4      | 1      | 4      | 3      | 3      | 31    | Good          |
| Kinley et al 2018              | 4      | 3      | 3      | 2      | 2      | 1      | 3      | 2      | 3      | 23    | Moderate       |
| Lindop & Read 2000             | 4      | 3      | 3      | 3      | 3      | 1      | 2      | 2      | 3      | 24    | Moderate       |
| Study                                      | Quality | MM | DI | CR | MR | SG | OC | SC | RL | SL | Rating |
|-------------------------------------------|---------|----|----|----|----|----|----|----|----|----|--------|
| Marriott et al 2013                       | 4       | 3  | 3  | 2  | 1  | 1  | 1  | 2  | 1  | 2  | Moderate |
| McKechnie 2006                            | 3       | 2  | 1  | 1  | 1  | 1  | 2  | 2  | 2  | 2  | Poor   |
| McLaughlin et al. 2014                    | 4       | 3  | 4  | 3  | 4  | 3  | 3  | 3  | 3  | 3  | Good   |
| McNamara et al. 2019                      | 4       | 3  | 3  | 3  | 3  | 3  | 4  | 3  | 3  | 3  | Good   |
| Moro et al. 2017                          | 4       | 4  | 3  | 4  | 3  | 2  | 3  | 3  | 3  | 3  | Good   |
| Morton-Nance & Schafer 2012               | 4       | 4  | 4  | 2  | 3  | 3  | 3  | 2  | 2  | 2  | Moderate |
| Ng & Li 2003                              | 3       | 3  | 3  | 3  | 3  | 3  | 3  | 2  | 2  | 3  | Moderate |
| Northway et al. 2019                      | 4       | 3  | 3  | 3  | 3  | 3  | 3  | 2  | 2  | 3  | Moderate |
| O'Regan & Drummond 2008                   | 3       | 2  | 2  | 2  | 2  | 2  | 3  | 3  | 3  | 3  | Moderate |
| O'Sullivan & Harding 2017                 | 4       | 3  | 3  | 2  | 4  | 3  | 3  | 2  | 3  | 2  | Moderate |
| Read & Cartlidge 2012                     | 4       | 3  | 3  | 2  | 2  | 2  | 4  | 3  | 2  | 2  | Moderate |
| Reddall 2010                              | 3       | 2  | 1  | 1  | 1  | 1  | 3  | 2  | 2  | 2  | Moderate |
| Ronneberg et al 2015                      | 2       | 1  | 2  | 1  | 2  | 1  | 3  | 2  | 2  | 2  | Poor   |
| Ryan et al 2016                           | 4       | 3  | 3  | 2  | 3  | 1  | 3  | 2  | 3  | 2  | Moderate |
| Ryan & McQuillan 2005                     | 3       | 2  | 1  | 1  | 1  | 1  | 3  | 2  | 2  | 2  | Poor   |
| Sampson et al. 2015                       | 4       | 3  | 3  | 3  | 3  | 1  | 3  | 3  | 3  | 3  | Moderate |
| Stein 2008                                | 3       | 2  | 3  | 2  | 2  | 1  | 3  | 2  | 2  | 2  | Moderate |
| Sue et al 2019                            | 3       | 2  | 2  | 3  | 2  | 1  | 3  | 2  | 3  | 3  | Moderate |
| Tuffrey-Wijne 2003                        | 4       | 3  | 3  | 3  | 3  | 2  | 3  | 3  | 3  | 3  | Moderate |
| Tuffrey-Wijne et al. 2007 (a)             | 4       | 3  | 4  | 3  | 3  | 3  | 3  | 3  | 3  | 3  | 29      Good |
| Tuffrey-Wijne et al. 2007 (b)             | 4       | 3  | 3  | 2  | 3  | 3  | 3  | 2  | 2  | 2  | 26      Moderate |
| Tuffrey-Wijne et al. 2008                 | 4       | 3  | 3  | 3  | 3  | 3  | 3  | 3  | 3  | 3  | 28      Good |
| Tuffrey-Wijne & McEnhill 2008             | 4       | 3  | 2  | 2  | 1  | 1  | 3  | 2  | 2  | 3  | 21      Moderate |
| Tuffrey-Wijne et al. 2009                 | 4       | 3  | 4  | 4  | 4  | 4  | 2  | 3  | 3  | 3  | 30      Good |
| Tuffrey-Wijne 2013                        | 4       | 4  | 3  | 3  | 4  | 4  | 4  | 3  | 3  | 3  | 31      Good |
| Study                                   | A | B | C | D | E | F | G | H | I | J | Score | Quality |
|----------------------------------------|---|---|---|---|---|---|---|---|---|---|-------|---------|
| Tuffrey-Wijne et al. 2016              | 4 | 4 | 4 | 4 | 3 | 3 | 4 | 3 | 3 | 3 | 32    | Good    |
| Tuffrey-Wijne et al. 2017              | 4 | 4 | 3 | 2 | 3 | 3 | 3 | 3 | 3 | 3 | 28    | Good    |
| Tuffrey-Wijne & Davidson 2018          | 4 | 3 | 4 | 4 | 3 | 3 | 4 | 2 | 3 | 3 | 30    | Good    |
| Vrijmoeth et al. 2016 (a)              | 4 | 3 | 4 | 3 | 3 | 3 | 4 | 3 | 3 | 3 | 29    | Good    |
| Vrijmoeth et al. 2016 (b)              | 4 | 3 | 3 | 3 | 3 | 3 | 3 | 3 | 3 | 3 | 27    | Moderate|
| Vrijmoeth et al. 2018                  | 4 | 3 | 3 | 3 | 3 | 3 | 3 | 3 | 2 | 3 | 28    | Good    |
| Wagemans et al. 2013                   | 4 | 3 | 4 | 3 | 4 | 2 | 3 | 2 | 3 | 3 | 28    | Good    |
| Mark et al. 2017                       | 4 | 2 | 3 | 2 | 3 | 3 | 3 | 3 | 2 | 3 | 25    | Moderate|
| Welch & Moreno-Leguizamon 2016         | 3 | 3 | 4 | 4 | 3 | 3 | 3 | 3 | 3 | 3 | 29    | Good    |
