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Taylor, B. orcid.org/0000-0003-4090-3047, Tod, A. orcid.org/0000-0001-6336-3747, Gardiner, C. orcid.org/0000-0003-1785-7054 et al. (8 more authors) (2023) Mesothelioma patient and carer experience research: A research prioritisation exercise. *European Journal of Oncology Nursing*, 63. 102281. ISSN 1462-3889

<https://doi.org/10.1016/j.ejon.2023.102281>

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Title: Mesothelioma patient and carer experience research: A research prioritisation exercise

Abstract: 230/250

Objectives

Incidence of mesothelioma worldwide is growing and the UK reports the highest global incidence. Mesothelioma is an incurable cancer with a high symptom burden. However, it is under researched when compared to other cancers. The aim of this exercise was to identify unanswered questions about the mesothelioma patient and carer experience in the UK and to prioritise research areas of most importance through consultation with patients, carers and professionals.

Materials and methods

A virtual Research Prioritisation Exercise was conducted. This involved a review of mesothelioma patient and carer experience literature to identify research gaps and a national online survey to identify and rank research gaps. Following this, a modified consensus method with mesothelioma experts (patients, carers and professionals from healthcare, legal, academic and volunteer organisations) was undertaken to reach a consensus regarding mesothelioma patient and carer experience research priorities.

Results

Survey responses were received from 150 patients, carers and professionals and 29 research priorities were identified. During consensus meetings, 16 experts refined these into a list of 11 key priorities. The five most urgent priorities were symptom management, receiving a mesothelioma diagnosis, palliative and end of life care, treatment experiences, barriers and facilitators to joined up service provision.

Conclusion

This novel priority setting exercise will shape the national research agenda, contribute knowledge to inform nursing and wider clinical practice and ultimately improve the experiences of mesothelioma patients and carers.

Keywords: caregivers, clinical nurse specialist, clinical practice, consensus, grey literature, mesothelioma, nursing, patient experience, research prioritisation, United Kingdom

Introduction

Malignant mesothelioma is a rare, life-limiting and aggressive cancer with a high symptom burden (Odgerel et al, 2017). It is predominantly a pleural disease, affecting the lining of the lungs (89%) but can occur in the peritoneum (3%) or testes and other unspecified sites (8%) (Cancer Research UK, 2020). Mesothelioma is treatable but not curable and is associated with a range of debilitating symptoms including breathlessness, pain, cough, lethargy, weight loss and sweating (British Thoracic Society, 2007). This symptom burden creates challenges to nursing care. Often, patients enter into lengthy, complex clinical trials resulting in a range of side effects with no guarantee of benefit. With or without treatment, survival is usually measured in months with approximately 60% of patients not surviving beyond a year (Royal College of Physicians, 2020). While there are currently no curative treatments, recent innovations in surgery, radiotherapy and immunotherapy offer promise in terms of length of life and palliation of symptoms (Bibby et al, 2016). Integral to the experiences of mesothelioma patients and their families is the role of clinical nurse specialists (CNSs), providing invaluable support and care across the mesothelioma journey (Taylor et al, 2019; Gardiner et al, 2022). A growing network of Mesothelioma CNSs, funded by the charity Mesothelioma UK are based in NHS hospitals throughout the UK.

The only known cause of mesothelioma is exposure to asbestos (Mesothelioma UK, 2020). The latency period between exposure and disease presentation is usually between 15-45 years (MUK, 2020b). The UK now reports around 2700 mesothelioma diagnoses annually, the highest global incidence of mesothelioma (CRUK, 2020). Incidence is higher in certain occupational groups, including asbestos mining and disposal and construction industries (Rake et al, 2009). There is an increasing awareness of mesothelioma in other occupational groups including health care professionals and teachers (Howie, 2018; Allmark et al, 2020; Alpert et al, 2020). There is rising concern about the, as yet unquantified, risk of mesothelioma from asbestos exposure in living, educational and working environments. A recent parliamentary review (House of Commons Work and Pensions Committee, 2022) highlighted these concerns and warned of the urgent need for investment in a robust and systematic approach to measure asbestos exposure and reduce the associated catastrophic risks.

The growing incidence of mesothelioma cases in the UK and the historical and ubiquitous use of asbestos mean that it is increasingly important to recognise mesothelioma as a national priority. In 2013, the National Institute for Health Research (NIHR) funded a James Lind Alliance Priority Setting Partnership to agree mesothelioma research priorities, specifically identifying those unanswered questions that involved an intervention. This made a significant contribution to understanding

research priorities and improving care for mesothelioma patients (Stephens, Whiting and Cowan, 2015). However, the focus was on interventional research. This meant that all research priorities regarding mesothelioma patient and carer experience, and any other aspects of treatment or care not involving an intervention, were deemed out of scope and excluded.

Without research which focuses explicitly on patient and carer experience and delivery and organisation of services, there is a risk that new interventions, treatments and innovations in service delivery will not meet the needs of the people receiving them. Although the last decade has witnessed an increase in mesothelioma research, including some experience-based studies, many unanswered questions remain. If answered, these have the potential to inform clinical practice, address challenges of nursing care provision in rare cancers (Charalambous and Biagioli, 2022) and improve the mesothelioma patient and carer experience.

Furthermore, the recent Covid-19 pandemic has had a profound and devastating impact on people with mesothelioma and their families (Taylor et al, 2021; Gardiner et al, 2022). The legacy of the Covid-19 pandemic persists as evidence shows sustained delays in cancer diagnoses and treatments (Morris et al, 2021), generating a dangerous backlog of cancer patients (Macmillan, 2020) and adding further strategic pressure on the UK health care system. Mesothelioma patient and carer experience research will help to understand the long term implications of the pandemic, for example how to maximise the benefits of remote communication to ensure efficient and effective care delivery by nurses and other clinicians.

Aim and objectives

The aim of this Research Prioritisation Exercise (RPE) was to identify unanswered questions about the mesothelioma patient and carer experience and then to prioritise these research topics through consultation with patients, carers and professionals. This RPE focused on mesothelioma patient and carer experience research across the care pathway, from diagnosis to end of life care and bereavement. This included experiences of the disease and its management as well as care received, services provided and support given.

Objectives:

- Review existing literature to identify research gaps
- Seek the views of patients, carers and professionals as to how the experiences of people living with mesothelioma could be improved
- Consult with patients, carers and professionals to agree a prioritised list of research areas.

Ethical approval was obtained from the University of Sheffield Research Ethics Committee (reference number 038688).

Material and methods

The design for this study was an adapted consensus method, informed by pre-existing recommendations for consensus research (Jones and Hunter, 1995; Waggoner, Carline and Durning, 2016). Due to the social distancing restrictions imposed because of the Covid-19 pandemic, we adopted novel online and virtual methods of undertaking the RPE. The principles of good practice in priority setting, as laid out by Viergever et al. (2010) were followed throughout, demanding an inclusive, transparent and thoughtful approach. Careful consideration of the exercise focus, who to involve, plans for implementation and evaluation was important.

This section will describe each of the key stages of this RPE: establishing the steering group, evidence synthesis, a national online survey and a series of online meetings and ranking exercises.

Establishing the steering group

A diverse steering group (SG) was established at study outset. This comprised 16 people with expertise in mesothelioma, including patients (n=4), carers (n=2), clinical professionals (n=6), legal professionals (n=2) and professionals who provide advice and lead support groups (n=2).

Evidence synthesis

This RPE was informed by a synthesis of existing evidence to identify gaps in knowledge. This comprised three parts: rapid review of peer-reviewed published literature (Ejegi-Memeh et al. 2022), a review of grey literature and the James Lind Alliance Priority Setting Partnership for mesothelioma (JLA PSP) (James Lind Alliance, 2014).

Part 1: The Mesothelioma UK Research Centre conducted a rapid review of patient and carer experience research. The review asked the question ‘What do we know about the experience of living with mesothelioma, from the perspective of patients and their family carers?’ The process of reviewing existing literature enabled the identification of research gaps and these provided an evidence-based foundation for this RPE. The review methods and findings are reported separately (Ejegi-Memeh et al. 2022).

Part 2: A review of grey literature identified documents that had not been published in peer reviewed journals but still provided insights into the experiences of mesothelioma patients and carers and therefore potential gaps in knowledge. The incorporation of grey literature sources is essential to providing a comprehensive and nuanced overview of evidence. This is particularly

important when making decisions in areas of health policy and practice lacking in peer-reviewed published evidence (Benzies et al, 2006), such as mesothelioma patient and carer experience. Ethos (an e-theses online service) and Open Grey (a grey literature database) were searched using the search terms derived for the rapid review. Following this, the research team and SG members identified further items of grey literature relevant to the mesothelioma patient and carer experience. For a list of included documents please see supplementary file A.

Part three: The JLA PSP for mesothelioma, completed in 2013, was restricted to interventional questions (e.g. could be written as a PICO format). Many of the 'out of scope' topics identified (JLA, 2014) can be considered under the umbrella of 'mesothelioma patient experience' research and were therefore incorporated into this evidence synthesis.

The results from the rapid review were used as a framework when reviewing grey literature and the findings from both reviews were cross referenced with the JLA out of scope issues. This evidence synthesis identified 29 research gaps (see supplementary file B).

National online survey

An online survey was designed using Google Forms (the survey can be viewed in supplementary file C). The aim of the survey was to learn from the experience of those living and working with mesothelioma and to provide patients and family carers with the opportunity to inform the direction of future research.

The survey asked participants to score 29 identified research gaps on a scale of 1-10 (1 = not at all important, 10 = extremely important). In order to increase accessibility and communicate complex topics we summarised the 29 topics under 8 thematic groupings. Eight short video recordings were embedded in the survey, each one summarising research gaps identified from the evidence synthesis. Participants were also asked to provide free-text responses, detailing additional research priorities not already identified.

Sampling and recruitment

To achieve input across a range of relevant fields of mesothelioma expertise, eligible participants included anyone with lived experience of mesothelioma or who worked/ volunteered to support, treat or care for mesothelioma patients or carers. A variety of sampling strategies were employed. Firstly, SG members extended invitations within their networks, purposively sampling organisations and individuals with relevant experience (e.g. mesothelioma/cancer charities, asbestos support groups, law firms etc.). The Mesothelioma UK Research Centre team drafted invitations and worked with the SG to gather survey responses via the identified organisations. Secondly, identified

organisations/individuals were asked to share invitations within their own networks, both formal and informal, thus extending sampling via snowball sampling. Thirdly, a convenience sampling strategy was employed, inviting participants to complete the survey via adverts in newsletters and social media posts shared on behalf of Mesothelioma UK and the Mesothelioma UK Research Centre team. The survey was open for eight weeks (26th April to 25th June 2021).

Analysis

Closed question responses were analysed using descriptive statistics in Excel. Scores were combined and an average score for each of the 29 identified research gaps was calculated. A list of research topics was then generated in order of average scores and further analysis was undertaken, comparing results within and across sub-groups of the sample (for example patients, carers, professionals, those with experience of peritoneal or pleural mesothelioma). Responses to the open free-text questions were then analysed thematically using Quirkos data analysis software, grouped into categories and cross-referenced with existing identified research gaps.

Priority setting

In order to reach a consensus regarding how the identified research gaps should be prioritised, SG members completed an online ranking exercise and attended two online meetings during which a series of activities and discussions aided deliberation.

A nominal group technique was adopted for this stage of the RPE. This combined consensus-based (group discussion) and metrics-based approach (pooling individual rankings of research topics), a common combination when undertaking priority setting exercises (Viergever et al, 2010). Two virtual meetings were hosted to discuss all identified research priorities, and an online ranking exercise was completed by steering group members in between these two virtual meetings.

Priority setting meeting one

In meeting one, the research team presented the quantitative findings from the survey responses, and the ranking of each identified research gap. The SG discussed whether they broadly agreed with the findings, helped to refine general ideas into specific research topics and identify overarching themes.

Ranking Exercise

Informed by the discussion in meeting one, the research team re-categorised and merged the research topics. In doing so, a list of 11 research areas was generated. An online ranking activity was then circulated by email, inviting SG members to anonymously and individually rank these 11 topics

in order of importance and provide additional comments. SG members were asked to complete the exercise within two weeks of meeting one. The results were synthesised and guided the discussion and activities that took place in meeting two.

Priority setting meeting two

During meeting two, the research team presented the results of the ranking exercise and the findings from the qualitative analysis of open text survey responses. A facilitated discussion, and a series of live interactive polls were used to move the group towards a consensus regarding the final list of research priorities.

A number of issues were raised and discussed by the steering group. Examples include:

- Should peritoneal mesothelioma be recognised as a separate research priority?
- Should the research priorities be distinct or overlapping?
- Is there any added value to ranking the research priorities in order of importance?

Following constructive and respectful discussion, there was a high level of agreement on all issues raised. A draft research priority statement was then prepared. This was an iterative process, drawing on discussion with the SG, quantitative and qualitative survey responses and existing literature. The draft research priority statement was then shared with the SG for final comments.

Results

Demographic information

We received survey responses from 150 participants. Sample demographics are described in table 1.

	Participant characteristic	Number/ percentage of 150 respondents (to 1d.p)
Type of expertise/ experience (150 responses)	Person with a diagnosis of mesothelioma	56/ 37.3%
	Family member/ close friend of someone with a mesothelioma diagnosis	17/ 11.3%
	Bereaved family member or close friend	19/ 12.7%
	Health care or allied health care professional	25/ 16.7%
	Asbestos support group professional	5/ 3.3%
	Legal professional	15/ 10%
	Other professionals	13/ 8.7%
Participant age (150 responses)	<30 years	7/4.7%
	31-40 years	27/ 18%
	41-50 years	25/ 16.5%

	51-60 years	31/ 20.7%
	61-70 years	26/ 17.3%
	71-80 years	26/ 17.3%
	>80 years	7/ 4.7%
	Preferred not to say	1/0.7%
Gender (150 responses)	Female	104/ 69.3%
	Male	45/ 30%
	Preferred not to say	1/ 0.7%
Ethnicity (150 responses)	White	146/ 97.3%
	Asian	1/ 0.7%
	Caribbean or Black	1/ 0.7%
	Coptic	1/ 0.7%
	Preferred not to say	1/ 0.7%
Location (143 responses)	England	120/ 83.9%
	Scotland	11/ 7.7%
	Wales	9 (6.3%)
	Northern Ireland	3 (2.1%)

Table 1: A table describing survey sample demographic

Of the 92 patients and carers who participated, 80 responded to the question asking about type of mesothelioma. Of these, 37.5% (n=30) had lived experience of peritoneal mesothelioma and 57.5% (n=46) had lived experience of pleural mesothelioma while the remaining 5% (n=4) did not know. When asked about time since receiving their own or their loved one's mesothelioma diagnosis, 18.8% (n=15) had been diagnosed within the last 6 months, 20% (n=16) 7-11 months ago, 31.2% (n=25) 1-2 years ago and 30% (n=24) more than two years ago. The majority of professionals or volunteers (78.2%, n=43) reported that most of their patients had pleural mesothelioma, some said 'it varies' (10.9%, n=6), one person said peritoneal mesothelioma (1.8%) and those remaining did not know (9.1%, n=5).

Statement of research Priorities

We now present the research priorities for mesothelioma patient and carer experience research. The research priority statement can be downloaded from the following link <https://www.sheffield.ac.uk/media/29388/download?attachment>. Box 1 summarises the 11 research priority topics. Each research priority is now described and illustrated with an open text response from the survey to illustrate the underpinning experiences and opinions.

The first five research priorities relating to mesothelioma patient and carer experience were identified as the most urgent:

- Symptom management for mesothelioma and support for patients and carers to manage the complex range of symptoms in mesothelioma. This includes management of physical

symptoms e.g. cough, breathlessness and management of psychological symptoms e.g. stress, depression, anxiety.

“When diagnosed I had a number of questions relating to self-help and self-management - these range from questions re complementary medicine to practical everyday things like diet.” (person with a diagnosis of mesothelioma)

- Receiving a mesothelioma diagnosis. This includes the way in which the diagnosis was given and the support provided immediately afterwards as well as barriers and facilitators to early diagnosis and also the role of occupational history taking.

“Reflecting across all the topics mentioned here I think the one that most impacts me is the immediate post diagnosis period and the support offered at this point - what works/what is unhelpful.” (health care/ allied health professional)

- Palliative and end of life care. This includes supporting quality of life across the patient pathway, from diagnosis to the end of life and bereavement.

“I think more exploration of the palliative care needs of patients and carers is required in order to increase our understanding of how they interact with services (i.e., their willingness to engage with services) and how this can be improved.” (other professional or volunteer)

- Experience of receiving treatments, the physical impact of treatments and their side effects as well as factors that influence treatment decision making.

“The reasons why some patients choose best supportive care and not active treatment for their Mesothelioma.” (legal professional)

- Barriers and facilitators to joined-up service provision. This includes access to services and how different health, care and other services can be brought together to meet the needs, choices and aspirations of the individual. The impact of geographical variation across the UK on patient and carer experience may be considered.

“I think the geographical variation in the services patients receive is huge and there would be a lot of value in understanding the difference between the experiences of patients living in urban environments treated in large teaching hospitals often with a CNS on site vs patients who live in rural areas” (other professional or volunteer)

Another six research priorities relating to mesothelioma patient and carer experience were also identified as important, through the research prioritisation exercise (*in no particular order*):

- Care delivery. This recognises the role of different professionals in supporting patients and their families, for example in maintaining hope. Preferences for the delivery of information about mesothelioma and the role of support groups are included.

“I think the role of specialist support groups in providing practical advice, benefits information and holistic support is under-researched. The patients I know who have accessed support via a group are generally then more well informed about benefits, support services in their local community and in touch with peer support systems which greatly assist in the management of their condition.” (legal professional)

- Living with peritoneal mesothelioma is a severely under-researched area therefore all aspects of the peritoneal mesothelioma experience need further research. The experiences of peritoneal mesothelioma patients and carers may be integrated into the other research priorities.

“Sufferers of peritoneal feel themselves at the end of the queue for advice and action on trials and treatment” (person with a diagnosis of mesothelioma)

- Mental health and well-being (positive and negative) across the mesothelioma journey. This includes the link between physical and psychological well-being.

“the psychological impact of uncertainty about how symptoms are likely to progress and life expectancy” (person with a diagnosis of mesothelioma)

- Experience of clinical trials, including factors influencing clinical trial participation as well as the costs and benefits of taking part in a clinical trial.

“Research to inform the development of better participant information and consent processes in mesothelioma clinical trials.” (other professional or volunteer)

- Experience of caregivers from diagnosis to bereavement and how best to support and communicate with caregivers during this time. It is important to research the impact of mesothelioma on family and intimate relationships and to explore positive and negative impacts of informal caregiving.

“The patient should always be at the heart of treatment, but guidance for carers on how they can engage with the NHS and get the information they need is important.” (bereaved family member/ close friend of someone with a diagnosis of mesothelioma)

- The experience of seeking compensation, including barriers and facilitators.

“From a legal point of view, we want patients to understand why the compensation process is important - and can include access to better treatment options and care packages”. (legal professional)

These research priorities, rather than research questions, provide direction but also space, encouragement and opportunity for a vast range of research exploring the experiences of mesothelioma patients and carers.

Out of scope issues

Nearly all of the free text responses embellished the topics already identified, providing further understanding and insight regarding why research on each topic is important and has the potential to improve the patient and carer experience. This was particularly helpful when including sub-themes within each topic. Nevertheless, a small number of responses to the open-ended survey questions were later identified as out of scope by the SG. These out of scope responses were grouped into two topics; 1) clinical interventional research 2) awareness of asbestos exposure and mesothelioma risk.

1. Clinical interventional research

Clinical research is essential to enable the delivery of cutting edge diagnosis techniques, medicines and treatments in a safe and effective way and is therefore invaluable. However, this RPE maintained a focus on the experiences of mesothelioma patients and carers. For this reason, research topics that focused on interventional or clinical aspects of the disease were considered out of scope.

2. Awareness of asbestos exposure and mesothelioma risk

Improving awareness of asbestos exposure and risk of developing mesothelioma was an issue that was repeatedly raised. This related to patients and carers' shock on receiving the diagnosis, concern for others and a sense of injustice that they or others were not aware of the risks of asbestos exposure. While this undoubtedly shapes the experiences of patients and carers, the SG considered this a wider political and societal issue that was out of scope of this RPE. The SG did however hope that conducting research focusing on the experiences of patients and carers, would contribute to this broader issue by improving knowledge and awareness of mesothelioma.

Discussion

The long latency period and continued use of asbestos worldwide, (Frank and Joshi, 2014), means mesothelioma will continue to be a disease of concern throughout the 21st century (Robinson, 2012). Fortunately, there is growing understanding of the disease and of how best to treat it (Fear, Cook and Fisher, 2019). Therefore, it is essential that we gather evidence to inform how new developments in treatments and interventions can be most effectively applied to clinical practice, shape service delivery and nursing care and therefore improve the experiences of patients and carers. There is some evidence documenting the challenges of nursing patients living with a rare cancer, such as mesothelioma (Charalambous and Biagioli, 2022). This mesothelioma patient and carer experience RPE provides a platform that will support and encourage the national research

community in the UK to fulfil the vital need for more evidence on the impact of living with mesothelioma. It is essential that these research priorities reach the nursing workforce to enable nurses to keep abreast of the future research agenda, seek opportunities to contribute to the evidence base and continue to provide high quality nursing care to mesothelioma patients and their families.

Valuable patient and carer experience research has recently been conducted outside of the UK (see Prusak et al, 2021; Bonafede et al, 2020; Girgis et al, 2019; Guglielmucci, Franzoi et al, 2018; Guglielmucci, Bonafede et al, 2018; Kasai and Hino 2018; Nagamatsu et al, 2018; Nagamatsu et al, 2019; Padilha et al, 2019; Walker et al, 2019; Warby et al, 2019; Williams et al, 2018) and demonstrates the growing impetus regarding mesothelioma patient and carer experience research. Although this RPE focused on the experiences of people living with mesothelioma in the UK and current UK healthcare and support provision, some insights are transferable to worldwide contexts. Lessons learned from the RPE can contribute to the development of similar exercises in other countries across the world to strengthen the international body of research dedicated to improving clinical practice and the experiences of mesothelioma patients and carers.

Due to the Covid-19 pandemic, it was crucial that this exercise was undertaken virtually to protect the health of SG members and participants, all of whom were patients, carers or worked in close contact with vulnerable individuals. This did bring some advantages, including no travel and reduced time demands on SG members and increased accessibility for people across the UK to contribute to the RPE. Further, the online setting contributes to methodological debate and innovations that are required to continue effective research during the COVID-19 pandemic (Howlett, 2022).

Nevertheless, it is not possible to know whether or how the research priorities would have differed if the exercise was completed in a face-to-face environment. Mesothelioma is a rare cancer and therefore the survey sampling strategy aimed to reach as many people as possible. While the survey received responses from people with a wide range of expertise and experience from across the four UK nations, the sample was not representative of all those living with mesothelioma and we acknowledge that those without access to the internet were unable to respond. The SG was diverse in terms of experience, expertise, gender and sector and this enabled diverse and rich opinions and contributions which helped to ensure that the resulting priorities correspond to the needs of those who will implement and/or benefit from these. However, the SG only included representation from two of the four UK nations: England, and Wales. Furthermore, patients who participated in the SG were not representative of the typical patient population, having all survived the disease for a longer period of time than the average patient.

This RPE successfully achieved its aim to generate a list of research priorities for mesothelioma patient and carer experience research. This list encompasses a diverse portfolio of research topics that have potential to improve the experiences of people living with mesothelioma. This exercise authentically captured the voices of patients and carers living with mesothelioma and professionals working to support, treat and care for these people so that future research undertaken in this field reflects the gaps and issues most important to them and to ensure that research investments are maximised. In doing so, this exercise demonstrates and supports research activity that values civic involvement and deliberative approaches, increasingly recognised as essential components of research that aims to shape health care policy and service delivery (Street et al. 2014; South et al. 2011). As a research centre, we aim to update this research prioritisation exercise in five years' time.

Conclusions

This RPE is the first of its kind to focus on mesothelioma patient and carer experience research. Although the body of evidence in this field is growing, many gaps remain. This exercise enabled patients, carers, nurses and other health professionals to shape the future research agenda. These research priorities provide direction for researchers and research funders to ensure that future research aligns with the voices of mesothelioma patients and their families to develop an evidence base required to improve their experiences. This novel priority setting exercise will shape the national research agenda, contribute knowledge to shape future priority setting exercises worldwide, shape the national research agenda and therefore evidence based practice and ultimately improve the experiences of mesothelioma patients and carers.

Conflict of interest

No competing interests to declare

Acknowledgements

This project was funded by Mesothelioma UK as part of a portfolio of research conducted by the Mesothelioma UK Research Centre (MURC). The MURC is an alliance between Mesothelioma UK and the University of Sheffield. The views expressed are those of the author(s), and not necessarily those of Mesothelioma UK or University of Sheffield.

Thank you to Mesothelioma UK and other organisations who supported the dissemination of the survey. Thank you to all survey respondents for taking the time to complete the survey. We are extremely grateful for the contributions, support and expertise of all steering group members throughout the RPE, named below.

- Hannah Ball, Mesothelioma UK Clinical Nurse Specialist

- Maria Guerin, Lung Cancer and Mesothelioma Nurse Specialist
- Lisa Clarke, Mesothelioma Clinical Nurse Specialist
- Mavis Nye, pleural mesothelioma patient
- Carrie Twist, peritoneal mesothelioma patient
- Julie Knights, bereaved mesothelioma carer
- Mary Sephton, bereaved mesothelioma carer
- Richard Green, partner, Hugh James legal firm
- Lucy Strong, partner, Hugh James legal firm
- Melissa Culligan, Registered Nurse, Director of Board of MARF, Director of Clinical Research University of Maryland
- Anna Bibby, Consultant Respiratory Physician & Honorary Senior Research Fellow
- Lorna Johns, Manager at Asbestos Awareness & Support Cymru (AASC)
- Donna Wakefield, Consultant in Palliative Medicine
- Gerry Courtney, pleural mesothelioma patient
- Sarah Thomas, mesothelioma welfare advisor
- Chris Willis, peritoneal mesothelioma patient

References

- Allmark, P., Tod, AM., Darlison, L. (2020) *MAGS: The healthcare staff mesothelioma asbestos guidance study*. Mesothelioma UK. Retrieved from <https://www.mesothelioma.uk.com/downloads/mags-report-2020/?wpdmdl=15390>
- Alpert, N., van Gerwen, M., & Taioli, E. (2020) 'Epidemiology of Mesothelioma in the 21st Century in Europe and the United States, 40 years After Restricted/ Banned Asbestos Use', *Translational Lung Cancer Research*, 9 (suppl 1), S28-S38. doi: <http://doi.org/10.21037/tlcr.2019.11.11>
- Benzies, KM., Premji, S., Hayden, KA., Serrett, K. (2006) 'State-of-the-evidence reviews: advantages and challenges of including grey literature' *Worldviews on Evidence Based Nursing*, 3(2), pp. 55-61. doi: <http://doi.org/10.1111/j.1741-6787.2006.00051.x>
- Bibby, A., Tsim, S., Kanellakis, N., Ball, H., Talbot, DC., Blyth, K., Maskell, N., Psallidas, I. (2016) 'Malignant pleural mesothelioma: an update on investigation, diagnosis and treatment', *European Respiratory Review*, 25, pp. 472-486. doi: <http://doi.org/10.1183/16000617.0063-2016>
- Bonafede, M., Ghelli, M., Corfiati, M., Rosa, V., Guglielmucci, F., Granieri, A., Branchi, C., Lavicoli, S., Marinaccio, A. (2018) 'The psychological distress and care needs of mesothelioma patients and asbestos-exposed subjects: A systematic review of published studies', *American Journal of Industrial Medicine*, 61(5), pp. 400–412. doi: <http://doi.org/10.1002/ajim.22831>
- Bonafede, M., Granieri, A., Binazzi, A., Mensi, C., Grosso, F., Santoro, G., Franzoi, I., Marinaccio, A., Guglielmucci, F. (2020) 'Psychological Distress After a Diagnosis of Malignant Mesothelioma in a Group of Patients and Caregivers at the National Priority Contaminated Site of Casale Monferrato', *International Journal of Environmental Research and Public Health*, 17(12), pp. 1–15. doi: <http://doi.org/10.3390/ijerph17124353>
- British Thoracic Society. (2007) *BTS statement on malignant mesothelioma in the UK, 2007 Thorax* 2007;62, ii1-ii19. doi:<http://doi.org/10.1136/thx.2007.087619>
- Cavone, D., Caputi, A., De Maria, L., Cannone, ES., Mansi, F., Birtolo, F., Delfino, MC., Vimercati, L. (2019) 'Epidemiology of Mesothelioma' *Environments*, 6(76). doi: <http://doi.org/10.3390/environments6070076>
- Charalambous, A., Biagioli, V., (2022) The Challenges of Rare Cancers in Nursing Care. *The European Journal of Oncology Nursing*, 60. doi: <http://doi.org/10.1016/j.ejon.2022.102195>
- CRUK. (2020) Mesothelioma Incidence Statistics. Retrieved from <https://www.cancerresearchuk.org/health-professional/cancer-statistics/statistics-by-cancer-type/mesothelioma#heading=Two>.
- Ejegi-Memeh, S., Sherborne, V., Harrison, M., Taylor, B., Senek, M., Tod, A., Gardiner, C. (2022) 'Patients' and Informal Carers' Experience of Living with Mesothelioma: a Systematic Rapid Review and Synthesis of the Literature', *European Journal of Oncology Nursing*, 58, 102122. doi: <http://doi.org/10.1016/e.jon.2022.102122>

- Fear, V. S., Cook, A. M., & Fisher, S. A. (2019) 'The Future of Mesothelioma Research: Basic Science Research', *Caring for Patients with Mesothelioma: Principles and Guidelines*, pp. 203–227. doi: https://doi.org/10.1007/978-3-319-96244-3_18
- Frank, AL., & Joshi, TK., (2014) 'The Global Spread of Asbestos', *Annals of Global Health Volume 80*, 4, 257-262. doi: <https://doi.org/10.1016/j.aogh.2014.09.016>
- Gardiner, C., Harrison, M., Hargreaves, S., Taylor, B. (2022) 'Clinical Nurse Specialist Role in Providing Generalist and Specialist Palliative Care: a Qualitative Study of Mesothelioma Clinical Nurse Specialists', *Journal of Advanced Nursing*, 00, pp. 1-10. doi: <https://doi.org/10.1111/jan.15277>
- Girgis, S., Smith, A., Lambert, S., Waller, A., Girgis, A. (2019) "'It sort of hit me like a baseball bat between the eyes": a Qualitative Study of the Psychosocial Experiences of Mesothelioma Patients and Carers', *Supportive Care in Cancer*, 27, pp. 631–8. doi: <http://doi.org/10.1007/s00520-018-4357-0>
- Guglielmucci, F., Franzoi, I., Bonafede, M., Borgogno, FV., Grosso, F., Granieri, A. (2018) "The less I think about it, the better I feel": A Thematic Analysis of the Subjective Experience of Malignant Mesothelioma Patients and their Caregivers', *Frontiers in Psychology*, 9(FEB). doi: <http://doi.org/10.3389/fpsyg.2018.00205>
- Health and Safety Executive. (2019) Mesothelioma Statistics for Great Britain 2019. Retrieved from <https://www.hse.gov.uk/Statistics/causdis/mesothelioma/mesothelioma.pdf>.
- Howie, R. (2018) 'Mesothelioma Deaths in Teachers and Nurses in Great Britain'. *Environmental Health Scotland*, 29 (4), pp. 35-37.
- House of Commons Work and Pensions Committee. 'The Health and Safety Executive's approach to asbestos management'. HC 560, 2021-2022. Retrieved from <https://committees.parliament.uk/publications/21884/documents/162937/default/>
- Howlett, M. (2022) 'Looking at the 'field' through a Zoom lens: Methodological Reflections on Conducting Online Research During a Global Pandemic', *Qualitative Research*, 22(3), pp. 387-402. doi:<http://doi.org/10.1177/1468794120985691>
- James Lind Alliance. (2014) Mesothelioma Priority Setting Partnership. Retrieved from <https://www.jla.nihr.ac.uk/priority-setting-partnerships/mesothelioma/>
- Jones, J., & Hunter, D. (1995). 'Consensus methods for medical and health services research', *British Medical Journal*, 311, pp. 376–380.
- Kasai, Y., & Hino, O. (2018) 'Psychological Transition Characteristics of Patients Diagnosed with Asbestos-Related Mesothelioma', *Juntendo Medical Journal*, 64(2), pp. 114–121. doi: <http://doi.org/10.14789/jmj.2018.64.jmj17-0a09>
- Macmillan. (2020) The Forgotten 'C'? The Impact of Covid-19 on Cancer Care. Retrieved from <https://www.macmillan.org.uk/get-involved/campaigns/we-make-change-happen/we-shape-policy/covid-19-impact-cancer-report.html>
- Morris, EJA., Goldacre, R., Spata, E., Mafham, M., Finan, PJ., Shelton, J., Richards, M., Spencer, K., Emberson, J., Hollings, S., Curnow, P., Gair, D., Sebag-Montefiore, D., Cunningham, C., Rutter, MD., Nicholson, BD., Rashbass, J., Landray, M., Collins, R., Casadei, B., Baigent C. (2021) Impact of the COVID-19 Pandemic on the Detection and Management of Colorectal Cancer in England: a Population-Based Study. *Lancet Gastroenterology & Hepatology*, 6 (3), pp. 199-208. doi: [https://doi.org/10.1016/S2468-1253\(21\)00005-4](https://doi.org/10.1016/S2468-1253(21)00005-4)

Mesothelioma UK. (2020) About mesothelioma. Retrieved from www.mesothelioma.uk.com/information/about-mesothelioma/

Mesothelioma UK. (2020b) Symptoms and controlling them. Retrieved from www.mesothelioma.uk.com/information/symptoms-and-controlling-them/

Nagamatsu, Y., Oze, I., Aoe, K., Hotta, K., Kato, K., Nakagawa, J., Hara, K., Kishimoto, T., Fujimoto, N. (2018) 'Quality of life of survivors of malignant pleural mesothelioma in Japan: A cross sectional study', *BMC Cancer*, 18(1), pp. 1–7. doi: <http://doi.org/10.1186/s12885-018-4293-x>

Nagamatsu, Y., Oze, I., Aoe, K., Hotta, K., Kato, K., Nakagawa, J., Hara, K., Kishimoto, T., Fujimoto, N. (2019) 'Physician requests by patients with malignant pleural mesothelioma in Japan', *BMC Cancer*, 19(1), pp. 1–7. doi: <http://doi.org/10.1186/s12885-019-5591-7>

Odgerel, C., Takahashi, K., Sorahan, T., Driscoll, T., Fitzmaurice, C., Yoko-O, M., Sawanyawisuth, K., Furuya, S., Tanaka, F., Horie, S., Zandwijk, NV., Takala, J. (2017) 'Estimation of the global burden of mesothelioma deaths from incomplete national mortality data', *Occup Environ Med*, 74(12), pp. 851–858. doi: <http://doi.org/10.1136/oemed-2017-104298>

Padilha Baran, F., Mercês, NN., Sarquis, LM., Rosa, LM., Mensi, C., Brey, C. (2019) 'Therapeutic itinerary revealed by the family members of individuals with mesothelioma: Multiple case studies', *Texto E Contexto Enfermagem*, 28, pp. 1–15. doi: <http://doi.org/10.1590/1980-265X-TCE-2017-0571>

Prusak, A., van der Zwan JM, Aarts MJ, Arber A, Cornelissen R, Burgers S, Duijts SFA. (2021) 'The psychological impact of living with mesothelioma: experiences and needs of patients and their carers regarding supportive care', *European Journal of Cancer Care*, 30(6), e13498. doi: <http://doi.org/10.1111/ecc.13498>

Rake, C., Gilham, C., Hatch, J., Darnton, A., Hodgson, J., Peto, J. (2009) 'Occupational, domestic and environmental mesothelioma risks in the British population: a case-control study', *British Journal of Cancer*, 100(7), pp. 1175–83. doi: <http://doi.org/10.1038/sj.bjc.6604879>.

Robinson, B. (2012) Malignant pleural mesothelioma: an epidemiological perspective. *Annals of Cardiothoracic surgery*, 1(4), 491–496.

Royal College of Physicians. (2020) National Mesothelioma Audit Report 2020 (for the audit period 2016–2018). Retrieved from www.rcplondon.ac.uk/projects/outputs/national-mesothelioma-audit-report-2020-audit-period-2016-18

Stephens, R J., Whiting, C., & Cowan, K. (2015) Research priorities in mesothelioma: A James Lind Alliance Priority Setting Partnership. *Lung Cancer*, 89(2), 175–180. doi: <https://doi.org/10.1016/j.lungcan.2015.05.021>

South, J., Meah, A., & Branney, P. E. (2011) 'Think differently and be prepared to demonstrate trust': finding from public hearings, England, on supporting lay people in public health roles. *Health Promotion International*, 27, 284–294. doi: <http://doi.org/10.1093/heapro/dar022>

Street, J., Duszynski, K., Krawczyk, S., Braunack-Mayer, A. (2014) 'The use of citizens juries in health policy decision making: a systematic review', *Social Science & Medicine*, 109, pp. 1–9. doi: <http://doi.org/10.1016/j.socscimed.2014.03.005>

Taylor, B., Tod, A., Gardiner, C., Creech, L., Forde, F., Darlison, L. (2021) 'Effects of the COVID-19 pandemic on people with mesothelioma and their carers', *Cancer Nursing Practice*, 20(2), pp. 22–28. doi: <https://doi.org/10.7748/cnp.2021.e1773>

Viergever, R., Olifson, S., Ghaffar, A., Terry, RF. (2010) 'A checklist for health research priority setting: nine common themes of good practice', *Health Research Policy and Systems*, 8 (36). doi: <http://doi.org/10.1186/1478-4505-8-36>.

Waggoner, J., Carline, J. D., & Durning, S. J. (2016) 'Is there a consensus on consensus methodology? Descriptions and recommendations for future consensus research', *Academic Medicine*, 91(5), 663–668. doi: <http://doi.org/10.1097/ACM.0000000000001092>.

Walker, S., Crist, JD., Shea, K., Holland, S., Cacchione, PZ. (2019) 'The Lived Experience of Persons with Malignant Pleural Mesothelioma in the United States', *Cancer Nursing*, 44(2), pp. E90-E98. doi: <http://doi.org/10.1097/ncc.0000000000000770>

Warby, A., Dhillon, HM., Kao, S., Vardy, JL. (2019) 'A survey of patient and caregiver experience with malignant pleural mesothelioma', *Supportive Care in Cancer*, 27(12), pp. 4675–4686. doi: <http://doi.org/10.1007/s00520-019-04760-x>

Williams, L., Whisenant, MS., Mendoza, TR., Haq, S., Keating, KN., Cuffel, B., Cleeland, CS. (2018) 'Modification of existing patient-reported outcome measures: qualitative development of the MD Anderson Symptom Inventory for malignant pleural mesothelioma (MDASI-MPM)', *Quality of Life Research*, 27(12), pp. 3229–3241. doi: <http://doi.org/10.1007/s11136-018-1982-5>

Box 1: Summary of research priorities for mesothelioma patient and carer experience research

Five most urgent research priorities (in no particular order):

- Symptom management
- Receiving a mesothelioma diagnosis
- Palliative and end of life care
- Experience of treatments
- Barriers and facilitators to joined up service provision

Other research priorities (in no particular order):

- Care delivery
- Living with peritoneal mesothelioma
- Experience of clinical trials
- Experience of caregivers
- Experience of seeking compensation