

# Perceptions and experiences of adults with neuromuscular disorders and their carers in relation to airway clearance and respiratory physiotherapy in the community

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## Abstract

### Background

Multidisciplinary care, including respiratory physiotherapy, is advocated for people with Neuromuscular disorders (NMD) to manage respiratory muscle weakness, maximise cough efficiency and support airway clearance. The experience and perceptions of adults with NMD regarding airway clearance and respiratory physiotherapy are not known.

### Methods

People with NMD and their carers participated in online semi-structured interviews. They were recruited via convenience sampling from a Neuromuscular Centre and the Motor Neurone Disease Association. Data was analysed using content analysis.

### Results

Five people with NMD and three carers were successfully interviewed. Results suggested barriers to specialised services for adults with NMD. People with NMD were either unclear about, or unable to identify what was meant by respiratory physiotherapy or airway clearance. They also reported having no access to a respiratory physiotherapist at clinics or charity settings. Conversely, all needed to clear their secretions daily and suggested that there was value in education and reassurance about their respiratory management. Fears over the potential lack of services, including physiotherapy, were expressed.

## Conclusion

People with NMD may have unmet needs in relation to airway clearance and respiratory physiotherapy in the community. Further research is needed to explore perceptions of the role of airway clearance, and respiratory physiotherapy among other clinicians, so that barriers to accessing respiratory physiotherapy services can be understood and addressed.

## Introduction

Neuromuscular disorders (NMD) are a heterogeneous group of progressive neurodegenerative disorders with varying prevalence. For example, reported prevalence for motor neurone disease and muscular dystrophies are five and 16 people per 100,000 respectively (1, 2). People with NMD have progressive muscle wasting, often affecting either one or all the major respiratory muscle groups (3). Respiratory muscle weakness causes hypoventilation and reductions in functional vital capacity and total lung capacity (4). Airway clearance (AC) is impaired as distal lung regions are under ventilated, compromising collateral ventilation, interdependence, secretion mobilisation and cough (4). Acute respiratory infections and chronic insufficiencies can follow, causing significant morbidity and mortality (3).

A multi-disciplinary approach to the care of people with NMD is widely advocated to improve quality of life, reduce medical complications and improve survival (5, 6). Respiratory physiotherapists are integral to the multi-disciplinary team, providing expertise on non-pharmacological management, aiding symptom control, AC and cough enhancement (3). The expertise of respiratory physiotherapists encompasses skills, and knowledge in assessing and managing respiratory muscle impairment, and cough insufficiency, enhancing quality of life and end of life symptoms (7).

Exploring people's perceptions and experiences has a critical role in understanding the effectiveness of the management of chronic disease (8). The perspectives of those living with NMD (and their carers), who use mechanical insufflation-exsufflation (MI-E), one form of AC and respiratory physiotherapy, have been explored (9, 10, 11). However, the experiences of those with less advanced presentation or who do not utilise MI-E but may utilise other forms of AC and respiratory physiotherapy are currently unknown. Knowledge of the experiences and perceptions of this wider cohort of people with NMD would enhance patient-centred care. The aim of this study was to understand the perceptions and experiences of adults with NMD, and their carers, in relation to AC and respiratory physiotherapy in the community.

## Methods

### Study design

A qualitative design, using semi-structured interviews, was chosen to gain a rich understanding of people's lived experiences. This enabled focused information to be gathered with the flexibility to explore any new ideas that were raised during the interviews. Both participants and carers were interviewed as both parties are often essential in the respiratory management of those with NMD in the community setting.

### Ethics and approvals

The study was approved by the University College London (UCL) Research Ethics Committee (Ethics Project ID number 20809/001). Data were collected, handled, stored, and confidentiality maintained in-line with national General Data Protection Regulation (12) guidance and UCL ethics processes.

### Participants and setting

Participants were recruited if they matched the inclusion and exclusion criteria (Table 1).

Table 1: Inclusion criteria for participants or carers.

Inclusion criteria		Exclusion criteria
Participant	Carer	Participant and carer
<ul style="list-style-type: none"><li>Self-reported diagnosis of a neuromuscular disorder*.</li></ul>	<ul style="list-style-type: none"><li>A carer of an adult (over the age of 18) with a self-reported diagnosis of a neuromuscular disorder*.</li></ul>	<ul style="list-style-type: none"><li>Participant's diagnosis not a neuromuscular disorder such as a traumatic brain injury or stroke.</li></ul>
<ul style="list-style-type: none"><li>=/ &gt; 18 years.</li></ul>	<ul style="list-style-type: none"><li>=/ &gt; 18 years.</li></ul>	<ul style="list-style-type: none"><li>No participant perceived respiratory impairments or requirements.</li></ul>
<ul style="list-style-type: none"><li>English speaking.</li></ul>	<ul style="list-style-type: none"><li>English speaking.</li></ul>	<ul style="list-style-type: none"><li>Non-English speaking.</li></ul>
<ul style="list-style-type: none"><li>Able to read English.</li></ul>	<ul style="list-style-type: none"><li>Able to read English.</li></ul>	<ul style="list-style-type: none"><li>Unable to read English.</li></ul>
<ul style="list-style-type: none"><li>Access to the internet.</li></ul>	<ul style="list-style-type: none"><li>Access to the internet.</li></ul>	<ul style="list-style-type: none"><li>No access to the internet.</li></ul>
<ul style="list-style-type: none"><li>Able to participate in an interview via Microsoft Teams® or Zoom® (could be assisted by a carer).</li></ul>	<ul style="list-style-type: none"><li>Able to participate in an interview via Microsoft Teams® or Zoom®.</li></ul>	<ul style="list-style-type: none"><li>Unable to participate in an interview via Microsoft Teams® or Zoom®.</li></ul>

(NINDS, 2021)

Online interviews were used for participants' convenience, and protection from risk of infection during the COVID-19 pandemic. Unfortunately, it was not possible to recruit non-English speaking participants as this was an unfunded study without access to interpreters.

### **Recruitment and consent**

A description of the study was posted on the Motor Neurone Disease Association research noticeboard, newsletter and social media pages, inviting people to participate. In addition, an email was sent to people on the mailing list of a regional Neuromuscular Centre (NMC), also inviting them to participate in the study.

Potential participants were invited to contact the primary researcher (EH), at which point they were screened and given the opportunity to ask questions about the study. Following this, an information sheet and consent form were sent to potential participants via email. If they wished to take part in the study they were asked to complete and return the consent form and arrangements for the interview were made.

### **Study procedure**

A semi-structured interview guide was developed by the primary researcher (EH), drawing on her experience of working with people with NMD, and informed by the existing supporting literature. The interview guide was scrutinised by an NMD specialist physiotherapist, and an independent research physiotherapist; adjustments were made as appropriate. The initial participant interview was utilised, as a pilot interview, no adjustments were made according to the feedback following this, consequently the interview was included in the analysis of the findings. Interviews were conducted by the researcher in English, over Zoom® and audio and video recorded.

An overview of procedure of the interview is shown in [Figure 1](#).

## Start of call

- The information sheet was summarised, and participants were asked to reiterate their consent verbally.
- Participants were informed that they could stop the interview at any time, and that their recordings could either be kept or destroyed, depending upon their wishes.

## Recording

- A rapport and the health status of the person with a neuromuscular disorder, was initially established before moving on to explore the research aim and objectives.
- Questioning followed the semi-structured interview guide.
- Participants were asked whether there was any other information they would like to share.
- Participants were thanked for their time.

## End of call

### **Figure 1: Overview of the interview procedure.**

#### **Data analysis**

Recorded data were downloaded in mp4 format and transcribed, the auto-generated Zoom<sup>®</sup> caption aided transcription. Transcription was completed by the primary researcher (EH), who also completed the interviews. This aided familiarisation with the data prior to analysis. Once the audio and video recordings were transcribed, they were anonymised and all identifiable information within their content removed. The NVivo<sup>®</sup> software package was then used to support content analysis. This system of coding allowed for categories and subcategories to be identified. Content analysis was chosen as a research technique which systematically and objectively identifies special characteristics in the data as such interpreting, coding and grouping information into categories and subcategories and then themes, in-line with the approach by (13). Following discussion with an expert peer reviewer, themes were confirmed, and no further changes were made.

#### **Data management and protection**

Audio and video recordings were only accessed by the interviewer and were deleted once transcribed. No hard copies of data were kept. All written transcripts and information were electronically stored on the UCL University One Drive account to be kept on-file for 10 years.

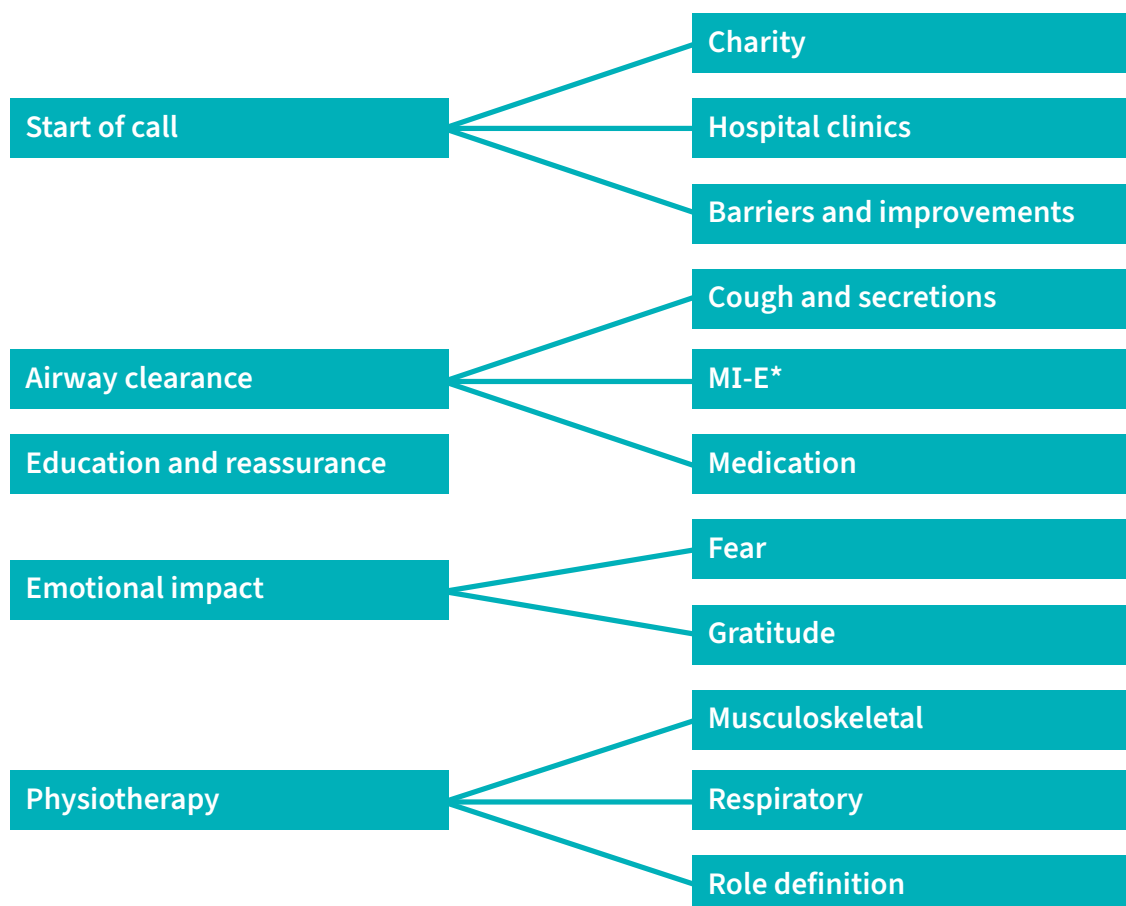
## Results

### Participant recruitment and demographics

Nine potential participants contacted the research team, seven of whom were eligible to participate. Participants included five people with NMD (four female; one male) and two carers, both relatives of two participants. Five interviews in total were conducted, with two having both the participant and relative participating. The NMD diagnoses were Pompe disease, muscular dystrophy, motor neurone disease and spinal muscular atrophy, with time from diagnosis ranging from 4–47 years (median, 32 years). Four people with NMD used overnight mechanical ventilation. Participants were interviewed between July to November 2021, interviews lasted between 25 and 54 minutes. The interview was paused for one participant, as they found discussing their disease progression emotional. Following the pause, they were happy to continue and for their data to be used.

### Thematic findings

Five key themes were identified with subthemes devised. The key themes were, 1) access to services, 2) airway clearance, 3) education and reassurance, 4) emotional impact and 5) physiotherapy (Figure 2). Quotations in relation to each of these key themes are included in Table 2.



\*Mechanical insufflation-exsufflation (MI-E).

📌 **Figure 2: Key themes and subthemes developed through thematic analysis of interview data.**

Table 2: Quotations in relations to the five key themes.

Theme	Quotation/quotations
Access to services	<ul style="list-style-type: none"> <li>‘I have been going to the NMC almost since I was diagnosed, in 2002. And so I normally, without COVID-19, have regular physiotherapy.’ <i>Participant 1.</i></li> <li>‘So the GPs or whoever that had been dealing with mum, had never been aware of it and never referred her to it.’ <i>Carer 2.</i></li> </ul>
Airway clearance	<ul style="list-style-type: none"> <li>‘I do that every day religiously, every day, at least twice a day. And if I’m not very well, I might even do it three or four times a day. And that just gives me confidence that I’ve tried to expel, as much as I can’. <i>Participant 1.</i></li> <li>‘I think they said to take the tablets, that would help to clear the mucus’. <i>Carer 2.</i></li> </ul>
Physiotherapy	<ul style="list-style-type: none"> <li>‘If I was to ask you, what do you think respiratory physio is?’ <i>Interviewer.</i> ‘Erm let me think, possibly a bit of question mark. I mean, I know that with that terrible illness where they do bash on their chest, to get the rubbish up. They have respiratory physio. But other than that, I don’t know much about it.’ <i>Patient 2.</i> ‘I don’t either’. <i>Carer 2.</i></li> </ul>
Education and reassurance	<ul style="list-style-type: none"> <li>‘Do you have access to advice and education?’ <i>Interviewer.</i> ‘I suppose it’s the world wide web now.’ <i>Carer 2.</i></li> </ul>
Emotional impact	<ul style="list-style-type: none"> <li>‘You have to get referred, so you can get your GP or your neurological consultant. I was just very fortunate I was in the right place at the right time, and when I got diagnosed, my consultant had literally just been to the neuromuscular center’. <i>Participant 1.</i></li> <li>‘Because of my cough now, I am always frightened to be on my own.’ <i>Participant 4.</i></li> </ul>

### Access to services

All participants reported attending hospital based respiratory clinics, two to three times per year. Four participants also reported relying heavily on the NMC and reported that they had been accessing the NMC services including physiotherapy, for many (10–19) years. Barriers to accessing NHS and NMC services had been a problem for four participants. Three participants explained that neither they nor their GP knew of services that were available to them, such as specialised hospital clinics (*Participant 2*) or charity services (NMC) (*Participants 1, 2 and 3*).

### Airway clearance

All participants reported excess secretions. *Participants 1, 3 and 4* reported needing routine AC, *participant 2* said they did not need support with secretions and had no airway

clearance regime, but their carer reported that every morning they continuously coughed to clear secretions. *Participant 5* identified a regular need to clear their throat and found this difficult. Three participants used MI-E daily (*participants 1, 3 and 4*), which was supplied and maintained by a hospital respiratory clinic, and their technique was reviewed in clinic. These participants reported that a family member assisted with the administration of MI-E. They expressed their dependence and appreciation of the use of MI-E. *Participant 2* reported that when excess secretions were a concern, medications had also been given.

### Physiotherapy

All five participants had accessed physiotherapy in the past from the NMC. None reported receiving physiotherapy outside of this charity, and all specified they accessed musculo-skeletal physiotherapy as shown in Table 3.

📄 **Table 3: Participant's and carer's access to and perceptions of physiotherapy.**

	<b>Access to physio-therapy?</b>	<b>Accessed physio-therapy at NMC?</b>	<b>Accessed physiotherapy outside of NMC?</b>	<b>Accessed MSK physio-therapy?</b>	<b>Perception of what respiratory physio-therapy entailed</b>
<i>Participant 1</i>	✓	✓	✗	✓	<ul style="list-style-type: none"> <li>• MI-E.</li> <li>• Breathing exercises.</li> <li>• Bed exercise.</li> </ul>
<i>Participant 2</i>	✓	✓	✗	✓	<ul style="list-style-type: none"> <li>• Unknown.</li> </ul>
<i>Participant 3</i>	✓	✓	✗	✓	<ul style="list-style-type: none"> <li>• Unclear.</li> <li>• Hydrotherapy, massage, acupuncture reflexology.</li> </ul>
<i>Participant 4</i>	✓	✓	✗	✓	<ul style="list-style-type: none"> <li>• Acute hospital based respiratory physio-therapy during admissions only.</li> </ul>
<i>Participant 5</i>	✓	✓	✗	✓	<ul style="list-style-type: none"> <li>• Unknown.</li> </ul>
<i>Carer 1</i>	NA	NA	NA	NA	<ul style="list-style-type: none"> <li>• Unknown.</li> </ul>
<i>Carer 2</i>	NA	NA	NA	NA	<ul style="list-style-type: none"> <li>• Acute hospital based respiratory physio-therapy during admissions only.</li> </ul>



*Participants 1, 2, 5 and carer 1* were unclear as to what respiratory physiotherapy was, no-one related their experience of physiotherapy with their airway clearance or respiratory needs.

### **Education and reassurance**

There was a strong theme of education and the need for reassurance amongst the narrative from all participants. Participants reported education and reassurance on respiratory management came from hospital respiratory clinics for MI-E (*participant 1 and 3*) and home mechanical ventilation use (all participants).

Participants and carers generally felt they needed to ask for support and education (*Participants 2 and 3, Carer 3*). The internet was identified by all participants as a primary source of information for all participants and carers with respiratory issues.

### **Emotional impact**

Participants expressed appreciation of the benefits of their ventilators and *participants 1, 2 and 4* were grateful for their MI-E device. *Participants 2 and 4* expressed a fear of not being able to breathe properly and *participant 1* expressed anxiety on recalling episodes when she had struggled to breathe. The concepts of fear and gratitude were also interconnected for *participants 1 and 3*, as they both were grateful for the support that they received, but feared it not being available.

## **Discussion**

This study explored the experiences and perceptions of respiratory care and physiotherapy in those with NMD and their carers in the community setting. All participants interviewed had sputum load and AC needs. Whilst all had access to GPs and hospital based respiratory clinics, all found accessing physiotherapy services challenging, relying upon third sector support to access musculoskeletal physiotherapy services only. There was a general lack of awareness of the role of respiratory physiotherapy. All participants identified a need for education and reassurance and reflected on the emotional impact attached to their respiratory symptoms and AC. Five key themes were identified that informed the above, and are discussed further.

### **Access to services**

Participants and carers perceived three key services for their respiratory care: hospital respiratory clinics, GP services and the NMC. Participants and carers were grateful for having access to the services at the NMC and relied heavily on this service for physiotherapy and emotional support. This centre is based in the northwest of England and referrals and uptake are predominately local to this region. This finding is corroborated by the findings by Rodger et al. (14) and Landfeldt et al. (15). Heavy reliance on a charity leaves this population vulnerable if services or funding are not available, as charity support should be an addition to NHS services, not instead of.

## **Airway clearance and respiratory physiotherapy**

This study corroborated perceived benefits and barriers of MI-E found by Siewers et al. (11) and Dale et al. (10) that included an initial lack of training and knowledge of the device. Respiratory clinics supporting the use of MI-E did not include a respiratory physiotherapist, and participants did not identify having any respiratory physiotherapy input. Outside of the use of MI-E and breathing exercises there was no mention of any management that might be included in a respiratory physiotherapist's scope of practice. This is despite all the participants reportedly needing to clear secretions daily and it being professionally recognised that AC and respiratory physiotherapy entail more than MI-E use. Respiratory services have been encouraged to include a specialist respiratory physiotherapist across the acute and community setting as a lack of specialist knowledge and unmanaged respiratory symptoms is evident (16, 17). Commissioning groups have been called on to recognise the value of specialist physiotherapy for neuromuscular participants and to commit funding to clinical support (18). This represents an area for ongoing clinical quality improvement and research.

## **Respiratory physiotherapy role identification**

GP support was a consistent theme throughout all the interviews. Ensuring that GPs have knowledge of referral options, and the role of respiratory physiotherapy may improve access to physiotherapy services.

NICE (6) advocated effective referrals to be a key priority for participants with NMD. However, participants and carers did not appear to be aware of the role or scope of respiratory physiotherapists. This suggests that either participants were receiving physiotherapy, but were not identifying it as such, or they were not accessing physiotherapy services. This highlights an area for potential education to raise the profile, role and benefits of respiratory physiotherapy, so that the needs of the NMD population can be met.

## **Education and reassurance**

Participants sought information from NMC and hospital clinics, although their main source of information was the internet. Drawbacks associated with internet use for participants include inaccurate information and access issues (19). Hence, it is important that information is disseminated clearly and unambiguously by respiratory physiotherapists working in partnership with third sector organisations to proactively provide and direct to trustworthy information sites clinicians to all participants with NMD to facilitate them in managing symptoms and to avoid hospital admissions.

## **Emotional impact**

Fear of choking to death is well documented in NMD and progressive respiratory diseases, as anxiety and respiratory diseases, may increase body sensations thus leading to panic (20). This anxiety is unpleasant for the person, is likely to have a negative effect on their quality of life, and has been linked to an increase in hospital admissions (21). Fear was also expressed in relation to the level of support available (or support being withdrawn). Fear surrounding cough and AC management is also a key feature of palliative care management

and it is crucial both are addressed in progressive conditions, to promote independent management (22). This further highlights the physical and psychological benefits, that access to specialised respiratory services, including respiratory physiotherapy, can have on adults with NMD.

### **Strengths and limitations**

Recruitment methods may have led to sampling bias in this population. Four of the five participants and carers lived within the Midlands and North-West of England, which means that their experiences may not be representative of the wider NMD community. Non-English-speaking participants were not eligible to participate, that limits the potential diversity of the populations interviewed. Positively, interviews were completed over Zoom®, with cameras on, as well as audio communication that allowed identification of non-verbal cues, and richer data to be gathered.

The understanding of AC and respiratory physiotherapy was limited and varied, between participants. This lack of identification with terms and concepts may have led to some participants and carers being deterred from volunteering for this study.

### **Future practice and research**

Increased advocacy for the role of AC and respiratory physiotherapy is needed for adults with NMD. Further exploration of service providers' experiences and perceptions on the same topic is suggested. Exploring reasons for the lack of clarity from participants and carers as to the role of AC and respiratory physiotherapy for adults with NMD is also needed. Research is recommended to address access to services, particularly when considering respiratory physiotherapy and AC.

## **Conclusion**

Respiratory hospital clinics, GP, and the NMC were key to the respiratory management of people living with NMD, yet all those interviewed had experienced barriers to accessing services. This was linked to a lack of knowledge of the services available. AC for three of the participants included the daily use of MI-E, yet beyond this, participants did not report other forms of AC, and respiratory physiotherapy. Participants were either unclear or unable to identify what respiratory physiotherapy was and did not report having access to a respiratory physiotherapist at either a clinic or charity settings. Reasons for this could include reduced exposure to respiratory physiotherapy and a lack of clarity over the role. Yet, all participants expressed the value of education and reassurance in terms of their respiratory management and the need to clear secretions daily. Participants and carers expressed fear of respiratory distress/inability to clear secretion and a fear a lack of services at the NMC. It therefore seems important that further research into the barriers to participants accessing specialised services, such as respiratory physiotherapy is needed.

## Key points

- People in the community with NMD are unclear or unable to identify the role and scope of respiratory physiotherapy and some report not having access to a respiratory physiotherapist at either a clinic or charity setting despite all identifying the need to clear secretions regularly.
- Many people with NMD are reliant on charity services and expressed fear of respiratory distress/inability to clear secretions and a fear a lack of services such as physiotherapy at the NMC.
- Further research is needed to ensure access to specialised services for NMD participants such as respiratory physiotherapy is available and accessible for participants.

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