Consensus research priorities for facial palsy: a Delphi survey of patients, carers, clinicians and researchers

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Abstract

Despite the potential negative impact of facial palsy, major gaps in understanding persist surrounding the treatment and care of those affected. This collaborative three-round Delphi process aimed to identify priorities for future facial palsy research, from the perspective of clinicians, researchers, patients and carers. It also determined whether the research priorities of patients and carers aligned with those of health professionals and researchers.

In Round one participants (n=85) were asked to generate research questions via focus groups or an online or postal survey. In Rounds two (n=72) and three (n=78) participants were asked to rate the priorities identified on a 5 point ordinal scale.

36 questions reached the definition of "high priority" consensus. Seven of each groups' top ten research priorities were shared. Prioritised questions included the provision and

pathways of care, the psychosocial impact of living with facial palsy and the effective management of side-effects.

Establishing the research priorities in the field of facial palsy is a significant first step in ensuring the future research agenda is focussed on topics that are considered important by both patients and health professionals.

Background

Facial palsy is an umbrella term referring to a weakness of the facial muscles caused by a variety of temporary or permanent conditions that affect the facial nerve, for example Bells Palsy, Ramsay Hunt Syndrome or Acoustic Neuroma. It is estimated that more than 100,000 people in the UK are affected by facial palsy[1], which can result in appearance changes alongside a wide range of physical symptoms, including difficulties with speech, facial expressions and eye function. Despite the potential negative impact of facial palsy on psychosocial functioning and quality of life, major gaps in understanding persist surrounding the treatment and care of those affected. Recognising the pressing need to develop a collaborative research agenda with the potential to address these gaps, the charity, Facial Palsy UK[2] sought to conduct a research priority setting exercise. To achieve this, they established a collaboration with the Centre for Appearance Research at the University of the West of England, UK[3] - an internationally acclaimed centre of research excellence in the field of disfigurement.

A Delphi technique was chosen as the most appropriate methodology to develop a facial palsy research agenda. Based upon the premise that a group consensus is more valid and reliable than individual perspectives,[4] the technique aims to obtain consensus on an important issue using a structured multi-staged survey involving a group of 'experts' or

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informed individuals. The Delphi technique has been used extensively in research priority setting exercises for health conditions, assisting researchers, funders and policy makers to formulate research questions with the greatest potential to result in benefit for patients.[5] As the process is often completed remotely, its permits participation from people drawn from diverse geographical areas, and unlike face-to-face methods, it avoids a situation in which those with particular experience or expertise might dominate the process. In recent years online (or 'eDelphi') methods are increasingly being favoured, offering several advantages over paper-based methods, including the potential for faster set-up and recruitment phases, the likelihood of accurate data analysis (as survey responses are entered directly into associated databases) and a higher level of data security.[6]

Variability exists between studies in the precise methods employed within the broader Delphi approach[7]. However, when used to set research priorities, the process typically begins with an open response round to generate a list of research questions participants believe need to be addressed. These responses are analysed, summarised and a new questionnaire developed based on the results obtained. Following this, two further rounds are usually conducted to move towards achieving consensus, where respondents are asked to rate the importance of the research priorities.[5] The present study can be considered modified, as it utilised a combination of focus groups and online or postal surveys to gather data for the initial open-response round.

The primary aim of this study was to use the Delphi technique to develop a set of facial palsy research priorities as identified by health professionals, researchers, patients and carers. The secondary objective was to determine if the research priorities of patients and carers aligned with those of health professionals and researchers. The results of this study will provide a focus for future grant applications and a source of reference to help researchers and funders determine where to allocate scarce research resources to achieve better outcomes for patients living with facial palsy.

Participants

There are no universally agreed criteria for the selection of experts for a Delphi study. However, it is essential to involve as many legitimate stakeholders as possible to ensure that the interests of all relevant parties are considered.[8] When setting research priorities for health conditions, there is a recognised need to include clinicians, researchers, patients and significant others who have experienced the impact of the condition (e.g.carers). This first-hand experience can help to foster ownership of the priorities amongst all those involved in the process, and increase the chance of their participation and/or cooperation in future research.[9]

The current study included all groups considered to have a legitimate stake in the research agenda for facial palsy. This broad stakeholder involvement was considered beneficial to minimise the risk of any potential research topic being overlooked, and to ensure that the priorities corresponded with those who may implement the research or could benefit from its outcome. Clear inclusion/exclusion criteria were established from the outset. All respondents were required to live in the UK (thus having experience of the condition and/or its treatment within the NHS) and have time to take part in the process. Patient respondents were required to have first-hand experience of facial palsy (any origin), or to be a close relative or carer. Health professional and researcher respondents were eligible to participate if they had at least 6 months experience of working with patients with facial palsy or conducting research in the field.

In order to recruit the expert panel, letters of invitation were sent to all members subscribing to the contact list of the charity Facial Palsy UK and to their Medical Advisory Board. In addition, snowball sampling was employed as invitations encouraged health professionals and researchers to share the invitation with suitable others. An information sheet was

attached to the invitation, informing potential participants of the purpose of the study, in addition to providing assurances of confidentiality and the opportunity to withdraw at any time. In order to be sent the first round survey, participants were required to register their interest using a short online form. This provided researchers with the information necessary to confirm their eligibility to participate. Consent to take part in Round 1 was obtained on paper for those who attended focus groups and at the start of the survey for those participating online. Ongoing consent was inferred by submission of the subsequent rounds.

Ethical approval was obtained from the University of the West of England's Research Ethics committee. Data were collected between February 2017- June 2017.

Methods

Round One

Round One employed three different data collection methods to obtain a 'long list' of respondents' research priorities. Two one-hour focus groups were held in London in February 2017, one with patients (n= 4) and the other health professionals/researchers (n= 5). These were facilitated by researchers from the Centre for Appearance Research. The topic of discussion was "What are the research priorities for facial palsy". For those unable to attend the focus groups, an invitation to propose their research priorities via an online survey hosted on the survey tool Qualtrics[10] was distributed .Those who did not want to participate online could do so via a postal survey. Additional information was requested from all participants including gender, age, and type of facial palsy for patients, and profession and years of experience for health professionals. Participants responding to the online or postal surveys were asked to propose a maximum of five research topics in response to the same headline question as the focus groups, namely "What are the research priorities for facial palsy".

The transcripts of the focus groups were transcribed verbatim and the research topics were extracted by the research team and combined with responses to the survey for analysis. Data were analysed qualitatively by the first author using inductive content analysis[4], whereby the research questions/topics proposed were organised and similar responses grouped together to reduce the number of items. In order to maintain methodological rigour and because content analysis has the potential to introduce researcher bias[12], this analysis was reviewed by two further members of the research team who commented on the analysis and agreed on the questions to go forward to the second round. Researchers kept the language of the questions as true to the original data as possible.

Round Two

In Round Two, the 'long list' of research topics was distributed electronically or by post to all those that registered to take part in the study. Participants were asked to rate how important they believed each research topic was using a 5-point Likert scale ranging from 1 (very important) and 5 (very unimportant). This survey remained open for two weeks and two reminders to complete the survey were sent during this time. Following this round, any questions failing to reach a rating of 'important' or 'very important' amongst 70% of respondents were removed from the list for Round Three.

Round Three

In Round Three, the consensus list was distributed once more electronically or by post to all of those who registered to take part in the study. In the previous round, the majority of topics had been rated as 'very important' or 'important', possibly reflecting respondents' perceptions that there is an overwhelming and general lack of research in the field.

Therefore to provide focus and elicit a more purposeful outcome, in this round, participants were asked to prioritise the research topics they wanted addressed within the next 5 years. Respondents were also provided with group priority ratings for each question from Round Two (e.g. 'in the last round 78% of the group rated this priority as important or very important'), enabling respondents to consider their response in light of the group's response.

Analysis plan and statistical considerations

Data from round two and three collected were exported from Qualtrics, cleaned (e.g. incomplete responses deleted) and analysed in SPSS. In addition, as this was an online survey and the link could be shared, some responses were received from individuals who had not registered to take part in the study from the outset. These data were deleted in order to maintain the integrity and authenticity of the data as the demographic information necessary to enforce the inclusion/exclusion criteria was absent. Descriptive statistics [mean, standard deviation (SD) and percentage] were calculated for the numbers of respondents rating each question as 'important' or 'very important'.

A universally agreed minimum level of consensus does not exist for a Delphi study but typically ranges from 50% to 80%.[11] Due to the lack of previous research in the field, a relatively high level of consensus about the most urgent research questions was anticipated in this study. Taking this into consideration, in addition to reviewing previous priority setting exercises, a target minimum consensus level was set at 70% from the outset. Therefore, a specific research question or topic was required to achieve an agreement of 'very important' or 'important' from at least 70% of the expert panel before it could be confirmed a priority.

The pragmatic decision was made to end the process after three rounds, based on previous work suggesting that additional rounds produce insignificant changes in opinion[13.] Following

the analysis of Round Three data, results were ranked according to two groups - patients/carers and health professionals/researchers – to identify the level of agreement between them in the questions rated most important. This was deemed appropriate, due to the imbalance in numbers of participants in the two groups. Means and SDs were used when ranking the importance of the research topics in Round Three. Greater importance was associated with higher means and smaller SD.

Results

Round One

The flow of respondents across all three rounds is presented in Fig.1 and their characteristics in Table 1 and 2. In total 85 people responded to Round One. The sample comprised of 67 patients/carers and 18 health professionals/researchers. The majority of patient respondents were female (n= 57, 85.1%) and had experienced acquired facial palsy (n= 55, 88.7 %) predominantly caused by Bells Palsy (n=25, 40.3%) Ramsay Hunt syndrome (n=8, 12.9%) or Acoustic Neuroma (n=10, 16.1%). In total 82.3% (n=51) were experiencing facial palsy at the time of the study and 74.5% (n=38) of these reported they were receiving some treatment. Health professional respondents (n=18) were primarily surgeons (n=9, 58%) and physiotherapists (n=5, 27.8%), with between 1 and 40 years' experience of working with facial palsy patients or conducting research and represented most geographical areas of the UK.

During Round One, 274 research questions were proposed by the 85 respondents, via focus groups (n=9), online survey (n=75) or postal survey (n=1). Twenty-one of these could not be formulated into a research question due to insufficient content or context and were removed.

The remaining questions were analysed and grouped using content analysis. In total, 45 research questions remained for prioritisation in the next round. The topics proposed varied from general research areas e.g. 'clear treatment pathways' to more specific questions e.g. 'What is the best protocol for managing facial palsy including technique/timing and sequence?'.

Round Two

In total, 72 people responded to Round two via online survey (n=71) and postal survey (n=1). They comprised of 54 patients or carers and 18 health professionals or researchers. During this round, three questions failed to reach the agreed levels of consensus amongst the group and were removed, leaving 42 research priorities to proceed for further rating in the final round.

Round Three

In total, 78 people responded to Round three, via online survey (n=77) and postal survey (n=1). They comprised of 62 patients or carers and 16 health professionals or researchers. Retention across all three rounds was 71%. During Round three, six questions failed to reach a sufficiently high level of consensus amongst all the respondents and were removed, leaving 36 research priorities to be ranked. The research priorities that reached group consensus are identified in Table 3 and have been ranked according to both health professional/researcher and patient/carer priority. High levels of consensus were demonstrated, with many research priorities reaching 100% agreement of 'important' or 'very important'. Some priorities reaching consensus in the patient/carer group did not reach consensus agreement in the health professional/carer group (n= 6).

Discussion

The aim of this modified Delphi study was to identify the research priorities for facial palsy according to key stakeholders not normally in communication with each other. It also aimed to establish if the research priorities of patients and carers aligned with those of health professionals and researchers After three rounds, the research priorities for were established and ranked for both patients/carers and health professionals/researchers. Seven out of the top 10 research priorities for each sub-group were shared, revealing a high level of consensus amongst stakeholders about the questions that require urgent attention.

The retention of respondents was excellent during the exercise, with 71% of the original panel taking part in all three rounds. Stakeholder commitment to take part in consecutive rounds are considered reflective of the level of interest or investment in the topic[4], indicating that a high level of importance is attached to advancing research in the field of facial palsy amongst those participating.

Previous research has reported that the provision of care for patients affected by facial palsy in the United Kingdom is inadequate and potentially detrimental to patient outcomes.[14, 15] For participants in this study, the importance of improving standards of care was evidenced by the number of research questions in the top 10 relating to this issue. Notably, health professionals and patients agree that research should systematically evaluate the impact of current facial palsy care on functional and psychological outcomes. Patients have prioritised more research to establish what treatment options (both surgical and non-surgical) are useful and to understand what information patients require upon diagnosis. Health

professionals recognise the need for significant advancements in the clinical management of facial palsy and in the urgent need to establish coordinated patient care pathways.

Research questions regarding synkinesis and eye functionality were attributed a high level of importance, (featuring in the 'top 5' of both patients and health professionals priorities), this may highlight the side effects that are either neglected, or the most challenging for patients and health professionals to manage effectively. Health professionals believed that research is required to standardise the ways clinicians' measure functional and psychological aspects of facial palsy. This supports previous reports of significant levels of heterogeneity in the current assessment of facial palsy and recommendations that clinicians should agree a common set of measures pending the development and evaluation of a definitive core data set.[16,17]

Lastly, both groups agreed that establishing the psychological impact of facial palsy was a priority. Whilst research has explored the psychological impact of living with facial disfigurement generally[18-21], there is a significant absence of research specific to facial palsy and therefore the support needs of those living with the condition is unknown. Future research should consider understanding the lived experiences of those living with facial palsy, both in the UK and worldwide.

As with any Delphi study, there are limitations to the present study. Firstly, this study was advertised using email, which required participants to have email access and computer skills, and might have excluded some from being invited or feeling confident in taking part. Secondly, content analysis was used to analyse the qualitative data from the open response round, and although steps were taken to minimise any researcher bias, this remains a possibility. Lastly, one health professional respondent stated in their additional comments that some research priorities are already being addressed (e.g. establishing core outcome measures). Therefore, it is important that those currently undertaking facial palsy research

ensure their findings are widely disseminated and are accessible to others. Furthermore, when consulting these research priorities in the future, professionals will need to review whether a significant gap in evidence remains.

To the best of our knowledge, this is the first study worldwide to establish the research priorities for facial palsy. As with previous research priority setting exercises, these results have implications for research, education and practice. This exercise is a significant first step to ensure a more focused and coordinated approach to research in a field that has been largely overlooked to date. Sharing these results with researchers, clinicians and funding bodies will help to focus future research agendas on the areas that will have the greatest importance and potential benefits for those living with facial palsy.

Acknowledgements

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