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Review

# Facial synkinesis outcome measures: A systematic review of the available grading systems and a Delphi study to identify the steps towards a consensus

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Systematic review;
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Synkinesia

**Abstract** *Introduction:* Facial synkinesis is characterised by abnormal and unintentional co-contractions of facial muscles caused by aberrant facial nerve healing, usually as a sequalae of facial palsy. The aim of this project is to propose a consensus for reporting this condition in the literature to facilitate the conduction of primary and secondary evidence studies, considering that no previous research has inquired in to this matter.
*Methods:* A systematic literature search was performed in MEDLINE and EMBASE databases, considering all the published articles on facial synkinesis. Studies that used a particular measuring system for this condition were included. Two authors independently assessed these articles focusing on the grading instruments utilised. The most commonly used instruments were analysed, and their basic components were incorporated in a modified Delphi survey, which was sent to a panel of experts.
*Results:* The systematic literature search retrieved 502 articles, of which 159 met the inclusion criteria. The two most commonly mentioned instruments were the House-Brackmann Scale and the Sunnybrook Facial Grading System. These were then followed by the Yanagihara scale, the Synkinesis Assessment Questionnaire, the eFace system and the Facial Clinimetric Evaluation. The modified Delphi study concluded that an ideal grading system for facial synkinesis should

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not only include a clinician-based evaluation of symmetry and signs of synkinesis, but also patient-reported symptoms.
*Conclusions:* Considering the characteristics of the studies found in the literature, the Sunnybrook Facial Grading System fits best with the ideal synkinesis measuring instrument described by the panel of experts. However, in order to satisfy the need to include patient-reported outcomes, the use of the Facial Clinimetric Evaluation as an adjunct to the Sunnybrook Facial Grading System is proposed.


## Contents



## Introduction

Facial synkinesis is one of the long-term sequalae of facial paralysis. This condition can be defined as abnormal and unintentional muscle contractions in a facial area when voluntary contraction in a different zone is attempted.[1,2] Contrasting with normal and expected co-contractions, facial synkinesis can be debilitating and painful. In severe cases, where chronic spasticity affecting agonist and antagonist muscles is present, facial synkinesis results in a pseudo-paralysis, producing the sensation of a tight face, instead of the classic flaccid manifestation of facial palsy.[3]

In recent years, several therapeutic interventions have been proposed for the treatment of facial synkinesis, including: physiotherapy, surgery and selective denervation using botulinum toxin injections. However, it has been alerted by authors conducting systematic reviews, the urgent necessity of standardising how this condition is reported.[4-6] The lack of a reliable, validated and wide-spread measure to evaluate pre-treatment and post-treatment outcomes stablishes a barrier not only for clinicians, but also for researchers.

Even though there has been recent progress to standardise reporting outcomes for facial palsy in general,[7-9] unfortunately this has not been replicated yet for facial synkinesis in particular. Unsurprisingly, both patients and clinicians have demonstrated their concern about this matter.[10]

The importance of this issue relies on that current evidence-based medicine (EBM) practice advocates for the conscious and judicious use of the best available empirical evidence for clinical decision-making.[11] Compared to other medical specialties, surgery has been particularly slow to adopt the principles of EBM, including plastic surgery.[12] Several barriers could explain this phenomenon, including the inherent difficulties associated with keeping the randomisation of patients blinded throughout surgical studies and the lack of adequate validated patient outcomes for certain conditions.[13]

The aim of this study was to identify the ideal method to grade patients with facial synkinesis. This would not only allow standardising how this condition is reported in the literature, reducing the heterogeneity in reporting outcomes. It could also potentially result in facilitating the conduction of meta-analysis to compare the findings of primary studies. Similarly, better documentation of facial synkinesis in facial palsy units would imply better recording of the patients' progression over time, affecting how clinicians monitor and audit the care they deliver.

## Materials and methods

In order to achieve the aims of this project a combination of two different methodologies were used. A broad systematic literature review, focusing on published articles that had included an instrument for measuring facial synkinesis, was sought to provide the necessary information on how facial synkinesis has been reported in the literature to date. This would help identify the most commonly used instruments for measuring this problem and their basic features.

Delphi survey methodology is a popular mean for achieving expert consensus. By using this technique, a panel of experts would be able to express their views in relation to their preferred synkinesis grading instrument and, at the same time, assess the importance of each basic component of the currently available systems. This would allow determining if a consensus could be reached on one of the currently available methods, and if that is not the case, at least identify which are the basic elements that an ideal instrument should possess.

The PRISMA Statement[14] was used as a guideline for the planning, conduction and reporting of the systematic literature review. As a first step, a detailed research protocol was prepared stating the eligibility criteria for studies to be considered (Table 1).

A systematic search strategy was designed to include as many facial synkinesis articles as possible, without any filters or limitations. Considering that the Medical Subject Headings (MESH) for synkinesis only includes articles

**Table 1** Systematic literature review article inclusion and exclusion criteria.

| Inclusion | Exclusion |
|---|---|
| Any scientific article that, as part of their study design, had measured facial synkinesis with a clinical grading system. | Articles not referring to facial synkinesis, such as studies focusing on facial spasm or synkinesis in other territories. |
| Articles written in English, Spanish, Portuguese, Italian and Greek were included, considering that the collaborators of this study are fluent in these languages. | Primary studies not measuring facial synkinesis with a clear instrument and secondary research not commenting on synkinesis grading systems. |
| | Withdrawn studies. |
| | Animal and laboratory-based research. |
| | Projects reporting facial synkinesis using nerve conduction studies only. |
| | Research protocols. |

published since 2005, a free text search was preferred to include previous reports: ("synkinesis" OR "synkinesia") AND ('face" or "facial"). This search was conducted on the 13th of February 2018 on MEDLINE and EMBASE databases. EndNote X8 (Clarivate Analytics, Pennsylvania, USA) software was used to eliminate duplicate entries. In a first instance, two independent assessors with previous experience in systematic reviews performed a title and abstract review of the articles retrieved, identifying those that were compatible with the inclusion and exclusion criteria (JEB and IK). A second full-text selection process was then conducted using the same above-mentioned inclusion and exclusion criteria.

For the data extraction process, a Microsoft Excel (Microsoft, Washington, USA) data gathering spreadsheet was used. The eligible articles were reviewed again in a parallel fashion by two authors independently (JEB and PK). Special attention was paid to identifying author information, journal and year of publication, specialty of the first author, country where the research was done and facial synkinesis instruments used.

No formal assessment of the risk of bias for each individual study was conducted. This was omitted considering that it had been previously established that an in-depth analysis of the methodological details and results of each article was out of the scope of this project.

For the modified Delphi study, the results from the systematic literature review were used as its first round. After identifying the most commonly used instruments for reporting facial synkinesis in the literature, these were deconstructed to obtain their basic elements. These findings were included in the second round of this modified Delphi study, as part of an online Delphi questionnaire.[15]

The first part of this survey contained a background information section to collect data regarding the respondents' place and scope of practice, years of experience and personal preference regarding facial synkinesis grading instruments. This was followed by a second section that included a series of Likert-scale questions for respondents to rate the importance of the basic synkinesis measuring elements identified in the literature. This was done to allow experts to express their preference related to particular ways of assessing facial palsy without committing necessarily with an instrument they were familiar with. Ethical approval for this study was obtained from the University of Oxford Medical Sciences Interdivisional Research Ethics Committee for review.

For the purpose of this study, experts in the field of facial palsy were defined as senior clinicians, such as substantive consultants or equivalent roles, in a specialty related with the diagnosis and treatment of facial palsy. Physiotherapists, occupational therapists and medical doctors specialised in neurology, neurosurgery, otorhinolaryngology – head and neck surgery, plastic surgery and maxillofacial surgery were invited to participate.

Facial Palsy UK, a registered charity based in England provided access to their medical advisory board, composed by thirty-nine experts that met the required inclusion criteria, based in the United Kingdom and abroad. Consensus was defined as a 75% of answers expressing moderate and extreme importance for a given element for assessing facial synkinesis.

## Results

In total, 855 publications were obtained from the systematic search. Of these, 477 were retrieved from EMBASE and 378 were acquired from MEDLINE. After the removal of 353 duplicate entries a list of 502 unique articles was obtained. A PRISMA flow-chart illustrating the study selection process can be found in Figure 1, detailing the reasons for exclusion at each stage.

A total of 159 articles, published between 1976 and 2018 were found in this systematic literature review, including a variety of study designs (Figure 2). A table summarising the main findings for each eligible article included in this review can be found in Table 3.

Ear, nose and throat (ENT) – Head and Neck Surgery was the predominant specialty among the authors responsible for the publications found, with 90 entries; followed by physiotherapists with 29 studies and plastic surgeons with 21 articles. Neurosurgeons authored 7 reports, maxillofacial surgeons and ophthalmologists wrote 5 papers each. For 12 entries the specialty of the first author was not mentioned or was unclear. The majority of the articles were published in journals that focus on otorhinolaryngology – head and neck surgery, with 84 articles forming part of this group. Plastic surgery related journals were the second most common, with 40 publications. In terms of countries

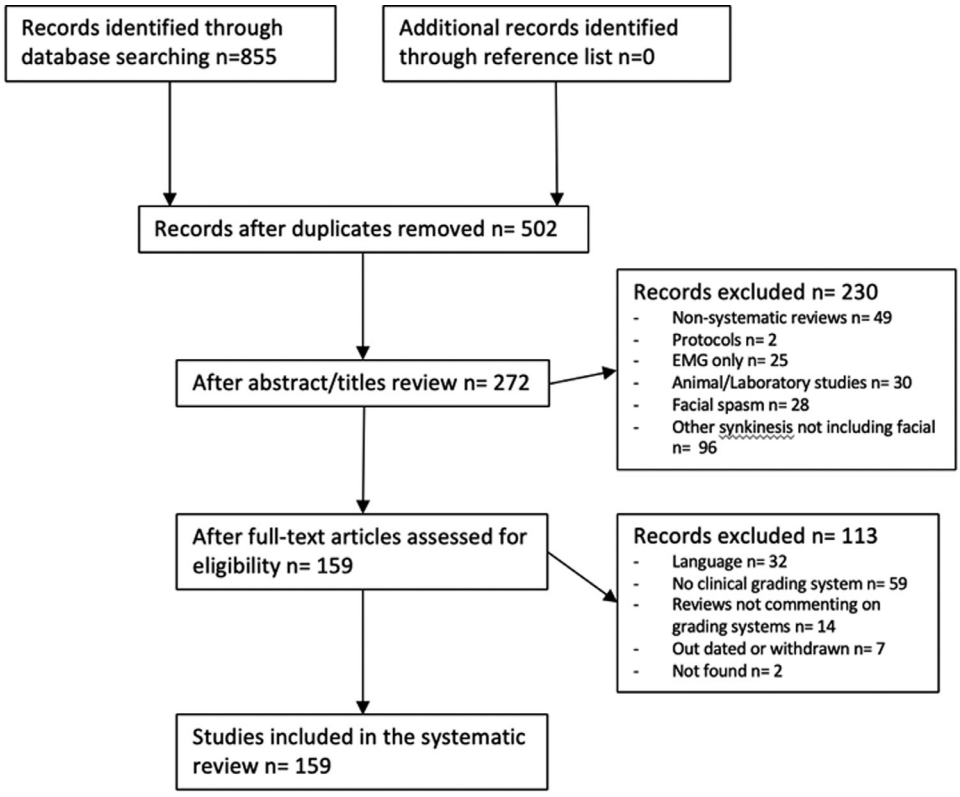


**Figure 1** Systematic literature review PRISMA flow-chart showing the study selection process for this project.

were the included research projects were undertaken, 54 studies were attributed to research group in the United States of America, followed by Japan with 20 cases, Italy with 14 and the United Kingdom with 9 entries.

Apart from the 9 systematic literature reviews found, the remaining 150 primary research articles reported a total of 11,554 cases, 859% of these being female and with a mean age of 45, 4 years. Most studies utilised just one grading system for measuring facial synkinesis, but in 38 articles more than one instrument was used. The House–Brackmann Facial Nerve Grading Scale[16] was the predominant choice for researchers, with 70 reports having used it. This was then followed by the Sunnybrook Facial Grading System[17] employed in 48 articles. The Yanagihara scale,[18] Synkinesis Assessment Questionnaire[19] and eFACE system[20] were used in 8 studies each. A total of 27 different named instruments were used in the articles included in this systematic literature review (Table 2). Apart from these described grading systems, 12 studies employed unnamed standardised measurement of facial landmarks and 2 reports utilised ad-hoc clinical grading questionnaires.

Most of the instruments identified were clinician-based, with 12 different named systems found, including the most popular ones. Nine different automatic, computerised systems were described, and 4 were patient-reported outcomes.

Considering the results obtained in the systematic literature review, the 5 most common grading instruments for facial synkinesis were examined: The House–Brackmann Facial Nerve Grading System, the Sunnybrook Facial Grading System, the Yanagihara facial nerve grading scale, eFACE and the Synkinesis Assessment Questionnaire.

Even though these instruments aim to measure the severity of the same condition, the approaches to do this differ slightly. Four main domains were identified in the analysed grading systems:

- Nature of the assessor: clinician-based versus patient-based.
- Nature of the deficit assessed: including evaluation of dynamic landmark excursion, static symmetry and synkinesis.
- Topographical approach: overall versus regional assessment.
- Nature of the assessment itself: subjective appraisal versus objective measurements.

These elements were considered to design a modified Delphi survey using the Online Surveys platform. This survey was sent to 39 experts on the 1st of June 2018, including three sections: the first one, focusing on the previously identified basic elements for measuring facial synkinesis; the second part inquiring on different instrument modalities, such as paper-based instruments, electronic questionnaires and computerised patient assessment; and the third section openly asking for the most relevant elements of any synkinesis grading instrument.

Three weeks after being sent to the potential respondents, on the 22nd of June 2018 the survey was closed obtaining a response rate of 28.2%. Respondents included 7 plastic surgeons, 2 physiotherapists, 1 ophthalmologist and 1 otorhinolaryngologist, which on average had 12.6 years of

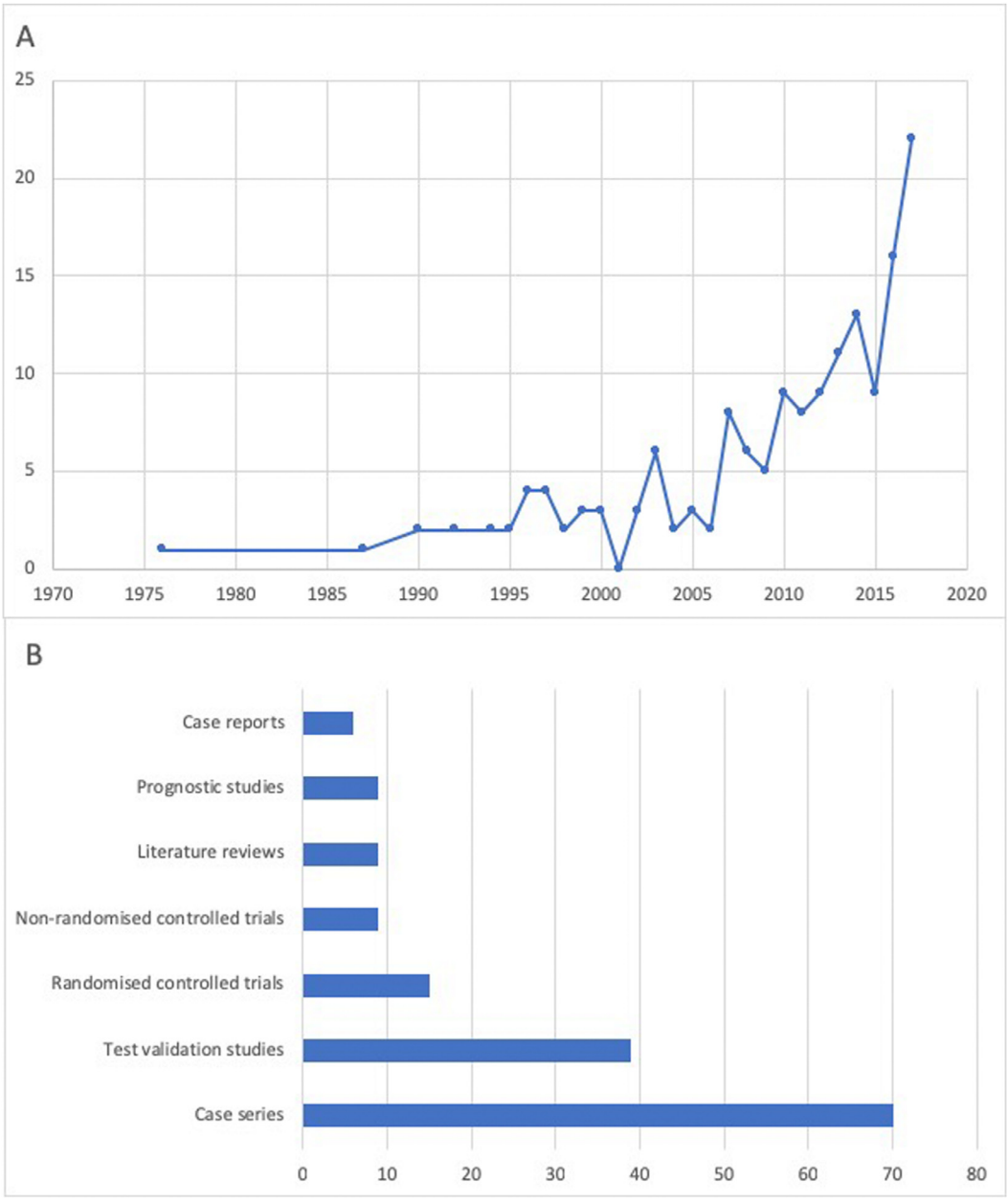


**Figure 2** (A) Graph showing the number of articles containing a synkinesis grading instrument published each year since 1976. (B) Column-graph showing the percentages for the different study designs present in the reviewed studies.

experience in their senior roles. Ten out of the eleven specialists were United Kingdom-based, with the single international respondent being from Chile. The Sunnybrook Facial Grading System was the most commonly used instrument by the panel of respondents (Figure 3).

The panel of experts recognised the overall subjective assessment of facial synkinesis done by the clinician as the single most important feature that should be included in a synkinesis grading instrument, with 100% of the respondents considering it moderately important or very important. There was also consensus on the importance of overall resting symmetry appraisal done by the clinician; regional assessment of synkinesis done by the clinician; patient self-assessment of synkinesis-related symptoms and patient self-assessment of the impact of synkinesis in their daily activities, as 81.8% of the panel considering these moderately or very important. Even though 72.7% of the respondents marked the assessment of dynamic facial excursion done by the clinician and regional resting symmetry appraisal as moderately or very important, these features did not reach the preestablished 75% threshold to define consensus. The item with the least level of consensus was the use of objective measurement of facial landmarks, with 2 respondents considering it slightly important, 5 answering neither important or non-important and 4 seeing it as very important.

In regard to the second part of the survey, none of the given options for delivering a synkinesis grading instrument reached a consensus. Options given to the panel were: paper-based clinician questionnaire, paper-based patient

**Table 2** Facial synkinesis grading instruments. Clinician-based instruments are shown in red, patient-based questionnaires in green; computerised systems in yellow and objective photographic or radiological measures in purple.

| Instrument | *n* | Instrument | *n* |
|---|---|---|---|
| House–Brackmann Facial Nerve Grading System | 70 | Facial paralysis recovery profile | 1 |
| Sunnybrook Facial Grading System (Toronto) | 48 | ADS | 1 |
| Yanagihara facial nerve grading scale | 8 | Facial CLIMA | 1 |
| eFACE | 8 | DVGate Motion Software | 1 |
| Synkinesis Assessment Questionnaire (SAQ) | 8 | Automated Facial Image Analysis (AFA) | 1 |
| Facial Clinimetric Evaluation Scale (FACE) | 5 | Post Paralysis Facial Synkinesis Examination Sheet | 1 |
| Facial Assessment by Computer Evaluation | 5 | Facial paresis questionnaire | 1 |
| PEAK Motion Measurement System | 4 | Maximal static response assay | 1 |
| Facial disability index | 4 | Facial asymmetry index | 1 |
| Freyss scale | 2 | 3D Ultrasound | 1 |
| Aesthetic and Functional Grading System | 2 | Glasgow Facial Palsy Scale (GFPS) | 1 |
| Sydney scale | 2 | OSCAR | 1 |
| Burres–Fisch linear measurement index | 2 | Unnamed clinical grading systems | 2 |
| CADS | 2 | Unnamed landmark measurement systems | 12 |
| May facial grading scale | 1 | | |

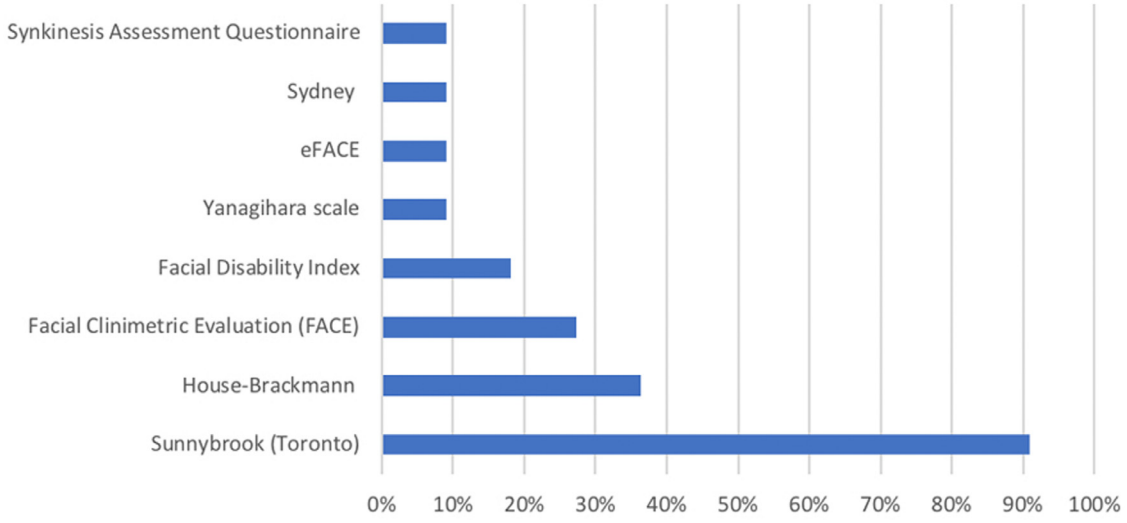


**Figure 3** Graph showing the responses for the following question: Which facial palsy grading instruments you routinely used for monitoring your patients?

questionnaire, smartphone-based clinician questionnaire, smartphone-based patient questionnaire and computerised assessment of facial landmarks.

The third part of the survey openly asking for any other important features of a synkinesis grading instruments did not obtained any new elements that had not been previously included by the research team.

On the 24th of June 2018 a third-round Delphi survey was sent to the recruited panel of experts that had responded the previous round. Only 3 of the participants responded to this third-round survey. In order to comply with the study protocol, no further attempts were made to contact the panel of experts to increase this response rate.

## Discussion

The incorporation of standardised instruments has become a priority for surgical specialties in order to address the challenges that the practice of evidence-based medicine implies. These not only allow an objective measurement of specific results, but also, if widely spread in the literature, favours the conduction of meta-analysis of studies that have used these instruments.

In general, for these instruments to demonstrate their quality 4 criteria should be assessed:

- Feasibility: capability of the instrument to be applied efficiently in a given context, which can be research, clinical or both.
- Reliability: capacity of the instrument to consistently measures the characteristic of interest, with an acceptable intra-rater and inter-rater reproducibility. Moreover, the grading system should present internal consistency, which means that items measuring the same feature should obtain similar results.
- Validity: confirms that an instrument measures what it intends to by comparing with other systems, such as “gold standards”.

**Table 3** Included studies in the systematic review.

| | Title | Study design | Outcome measure used | Specialty of first author | Country | |
|---|---|---|---|---|---|---|
| 1 | Adour et al. (1977) | Case series | Facial paralysis recovery profile | ENT | United States | Trauma |
| 2 | Yamamoto et al. (1988) | Prognostic | Yanagihara | ENT | Japan | Bell's palsy |
| 3 | Murata et al. (1990) | Diagnostic | Unnamed landmark measurement system | ENT | Japan | Bell's palsy |
| 4 | Jaaskelainen et al. (1990) | Case series | House-Brackmann | Neurosurgery | Finland | Acoustic neurinoma |
| 5 | Neely et al. (1992) | Diagnostic | House-Brackmann<br>Unnamed computerised analysis | ENT | United States | Bell's palsy |
| 6 | McKennan et al. (1992) | Case series | House-Brackmann | ENT | United States | Trauma |
| 7 | Laskawi et al. (1994) | Case series | Unnamed landmark measurement system | ENT | Germany | Bell's palsy |
| 8 | Johnson et al. (1994) | Diagnostic | Maximal Static Response Assay | Plastic surgery | United States | |
| 9 | Segal et al. (1995) | Non-randomized controlled trial | House-Brackmann | ENT | Canada | Bell's palsy |
| 10 | Segal et al. (1995) | Case series | House-Brackmann | Plastic surgery | Canada | Acoustic neurinoma |
| 11 | Moran and Neely (1996) | Case series | Unnamed computerised analysis | ENT | United States | Acoustic neurinoma |
| 12 | Ross et al. (1996) | Diagnostic | Sunnybrook<br>House-Brackmann | ENT | Canada | |
| 13 | Armstrong et al. (1996) | Case series | House-Brackmann | ENT | UK | Bell's palsy |
| 14 | Neely and Neufeld (1996) | Diagnostic | Facial Paresis Questionnaire | ENT | United States | |
| 15 | Brach et al. (1997) | Case series | Unnamed landmark measurement system | Physiotherapy | United States | |
| 16 | Laskawi (1997) | Case series | House-Brackmann | ENT | Germany | Acoustic neurinoma |
| 17 | Rickenmann et al. (1997) | Diagnostic | Fisch<br>House-Brackmann | ENT | Switzerland | |
| 18 | Brach et al. (1997) | Case series | Sunnybrook<br>House-Brackmann<br>FDI | Physiotherapy | United States | Bell's palsy |
| 19 | Bajaj-Luthra et al. (1998) | Diagnostic | Maximal Static Response Assay<br>Sunnybrook | Plastic surgery | United States | |

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**Table 3** (*continued*)

| | Title | Study design | Outcome measure used | Specialty of first author | Country | |
|---|---|---|---|---|---|---|
| 20 | Rodel (1998) | Case series | House-Brackmann | ENT | Germany | Acoustic neurinoma |
| 21 | Hammerschlag (1999) | Case series | House-Brackmann | ENT | United States | Acoustic neurinoma |
| 22 | Gantz et al. (1999) | Diagnostic | House-Brackmann | ENT | United States | Bell's palsy |
| 23 | Dulguerov et al. (1999) | Review | | ENT | Switzerland | |
| 24 | Kayhan et al. (2000) | Diagnostic | Sunnybrook | ENT | United States | Bell's palsy |
| 25 | Linstrom et al. (2000) | Diagnostic | PEAK Motus Motion Measurement System | ENT | United States | Healthy individuals |
| 26 | Targan and Alon (2000) | Case series | House-Brackmann | ENT | United States | Bell's palsy |
| 27 | Linstrom (2002) | Diagnostic | House-Brackmann PEAK Motus Motion Measurement System | ENT | United States | Tumours |
| 28 | Linstrom et al. (2002) | Diagnostic | House-Brackmann PEAK Motus Motion Measurement System | ENT | United States | Acoustic neurinoma |
| 29 | Frey and Giovanoli (2002) | Review | | Plastic surgery | Austria | |
| 30 | Hato et al. (2003) | Non-randomized controlled trial | Yanagihara | ENT | Japan | Bell's palsy |
| 31 | Cronin and Steenerson (2003) | Case series | May House-Brackmann | ENT | United States | Acoustic neurinoma |
| 32 | Beurskens and Heymans (2003) | RCT | House-Brackmann | Physiotherapy | Netherlands | |
| 33 | Yen et al. (2003) | Diagnostic | House-Brackmann | ENT | United States | Vestibular schwannoma |
| 34 | VanSwearingen et al. (2003) | Case series | Sunnybrook | Physiotherapy | United States | Acoustic neurinoma |
| 35 | Nakamura et al. (2003) | RCT | Dvgate Motion Software | ENT | Japan | Herpes zoster |
| 36 | Coulson et al. (2004) | Cross-sectional | Sunnybrook House-Brackmann FDI | Physiotherapy | Australia | |
| 37 | Beurskens and Heymans (2004) | Case series | House Brackmann | Physiotherapy | Netherlands | Acoustic neurinoma |
| 38 | Coulson et al. (2005) | Diagnostic | Sunnybrook | Physiotherapy | Australia | Acoustic neurinoma |

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**Table 3** (*continued*)

| | Title | Study design | Outcome measure used | Specialty of first author | Country | |
|---|---|---|---|---|---|---|
| 39 | Wu et al. (2005) | Diagnostic | Sydney House-Brackman PEAK Motus Motion Measurement System | ENT | United States | Bell's palsy |
| 40 | Dalla Toffola et al. (2005) | Case series | House-Brackmann | Physiotherapy | Italy | Bell's palsy |
| 41 | Beurskens and Heymans (2006) | RCT | Sunnybrook | Physiotherapy | Netherlands | Bell's palsy |
| 42 | Coulson et al. (2006) | Non-randomized controlled trial | House-Brackmann Sunnybrook | Physiotherapy | Australia | Vestibular schwannoma |
| 43 | Hato et al. (2007) | RCT | House-Brackmann Yanagihara | ENT | Japan | Bell's palsy |
| 44 | Kondo et al. (2007) | Case series | Yanagihara | ENT | Japan | Vestibular schwannoma |
| 45 | Mehta et al. (2007) | Diagnostic | SAQ | ENT | United States | |
| 46 | Yamamoto et al. (2007) | Case series | House-Brackmann | Plastic surgery | Japan | Bell's palsy |
| 47 | Kitisomprayoonkul et al. (2007) | Prognostic | House-Brackmann | Physiotherapy | Thailand | Bell's palsy |
| 48 | On et al. (2007) | Diagnostic | Sunnybrook | Physiotherapy | Turkey | Bell's palsy |
| 49 | Manikandan (2007) | RCT | Sunnybrook | Physiotherapy | India | Bell's palsy |
| 50 | Rogers et al. (2007) | Case series | Automated Facial Image Analysis (AFA) Sunnybrook | Plastic surgery | United States | |
| 51 | Engstrom et al. (2008) | RCT | House-Brackmann Sunnybrook | ENT | Sweden | Bell's palsy |
| 52 | Takanami et al. (2009) | Case report | House-Brackmann | ENT | Japan | Iatrogenic |
| 53 | Leonetti et al. (2008) | Case report | House-Brackmann | ENT | United States | Parotid tumour |
| 54 | Frey et al. (2008) | Case series | Unnamed landmark measurement system | Plastic surgery | Austria | |
| 55 | Cardoso et al. (2008) | Review | | Physiotherapy | Brazil | |
| 56 | Husseman and Mehta (2008) | Review | | ENT | United States | |
| 57 | Biglioli et al. (2009) | Case series | Functional and aesthetic grading system | Maxillo-Facial Surgery | Italy | Acoustic neurinoma |

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**Table 3** (*continued*)

| | Title | Study design | Outcome measure used | Specialty of first author | Country | |
|---|---|---|---|---|---|---|
| 58 | Kecskes et al. (2009) | Non-randomized controlled trial | Sunnybrook<br>House-Brackmann<br>Yanagihara<br>Freyss | ENT | France | Facial nerve schwannoma |
| 59 | Vrabec et al. (2009) | Diagnostic | House-Brackmann | ENT | United States | |
| 60 | Bodenez et al. (2010) | Case series | House-Brackmann | ENT | France | Bell's palsy |
| 61 | Venail et al. (2009) | Case series | House-Brackmann | ENT | France | Vestibular schwannoma |
| 62 | Zhang et al. (2010) | Case Series | Sunnybrook | Plastic surgery | China | Trauma |
| 63 | Beurskens et al. (2010) | Diagnostic | Unnamed questionnaire | Physiotherapy | Netherlands | Bell's palsy |
| 64 | Axelsson et al. (2011) | RCT | Sunnybrook | ENT | Sweden | Bell's palsy |
| 65 | Toffola et al. (2010) | Case series | Sunnybrook | Physiotherapy | Italy | Bell's palsy |
| 66 | Neely et al. (2010) | Diagnostic | Sunnybrook | ENT | United States | Bell's palsy |
| 67 | Navarrete and Junyent (2010) | | House-Brackmann<br>Sunnybrook | ENT | Spain | Bell's palsy |
| 68 | Kim et al. (2010) | Prognostic | House-Brackmann | ENT | Korea | Herpes zoster |
| 69 | Cai et al. (2010) | | House-Brackmann | Maxillo-Facial Surgery | China | |
| 70 | Krishnan et al. (2010) | Case series | Unnamed landmark measurement system | Neurosurgery | Germany | Tumour |
| 71 | Takemoto et al. (2011) | Prognostic | Yanagihara | ENT | Japan | Bell's palsy |
| 72 | Monini et al. (2011) | Case series | House-Brackmann<br>Sunnybrook | ENT | Italy | Tumour |
| 73 | Clapham et al. (2011) | Case series | House-Brackmann | Physiotherapy | UK | Acoustic neurinoma |
| 74 | Henstrom, et al. (2011) | Diagnostic | House-Brackmann | ENT | United States | |
| 75 | Wilkinson et al. (2011) | Case series | House-Brackmann | ENT | United States | Facial nerve schwannoma |
| 76 | Hirai et al. (2011) | Case report | House-Brackmann | ENT | Japan | Tumour |
| 77 | Pourmomeny and Zadmehr (2011) | Diagnostic | Sunnybrook<br>Unnamed landmark measurement system | Physiotherapy | Iran | Bell's palsy |
| 78 | Henstrom et al. (2011) | Case series | Facial clinimetric evaluation (FACE) | ENT | United States | Bell's palsy |
| 79 | Filipo et al. (2012) | Case series | Sunnybrook<br>SAQ | ENT | United States | Bell's palsy |

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**Table 3** (*continued*)

| | Title | Study design | Outcome measure used | Specialty of first author | Country | |
|---|---|---|---|---|---|---|
| 80 | Berg et al. (2012) | RCT | Sunnybrook<br>House-Brackmann | Plastic surgery | Norway | Bell's palsy |
| 81 | Axelsson et al. (2012) | RCT | Sunnybrook | ENT | Sweden | Bell's palsy |
| 82 | Saito (2012) | Diagnostic | Unnamed landmark measurement system | ENT | Japan | |
| 83 | Azuma et al. (2012) | Case series | House-Brackmann | ENT | Japan | Bell's palsy |
| 84 | Terzis and Karypidis (2012a) | Case series | Sunnybrook | Plastic surgery | United States | Cogenital |
| 85 | Terzis and Karypidis (2012b) | Case series | Sunnybrook | Plastic surgery | United States | Iatrogenic |
| 86 | Wernick Robinson et al. (2012) | Review | | ENT | United States | |
| 87 | Griffin and Kim (2012) | Review | | ENT | | |
| 88 | Chuang et al. (2013) | Case series | Unnamed landmark measurement system | Plastic surgery | Taiwan | Bell's palsy |
| 89 | Bran et al. (2014) | Case series | Unnamed questionnaire | ENT | Germany | Bell's palsy |
| 90 | Le Clerc et al. (2013) | Non-randomized controlled trial | House-Brackmann<br>Sunnybrook<br>Freyss | ENT | France | Vestibular schwannoma |
| 91 | Nicastri et al. (2013) | RCT | House-Brackmann | ENT | Italy | Bell's palsy |
| 92 | Wang et al. (2013) | Non-randomized controlled trial | House-Brackmann | ENT | China | Vestibular schwannoma |
| 93 | Nonaka et al. (2013) | Prognostic | House-Brackmann | Neurosurgery | United States | Vestibular schwannoma |
| 94 | Cayir and Set (2013) | Case report | House-Brackmann | | Turkey | Bell's palsy |
| 95 | Pourmomeny et al. (2014) | RCT | Sunnybrook | Physiotherapy | Iran | Bell's palsy |
| 96 | Cecini et al. (2013) | Case series | Facial assessment by computer evaluation (FACE)<br>Sunnybrook | Physiotherapy | Italy | Bell's palsy |

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**Table 3** (*continued*)

| | Title | Study design | Outcome measure used | Specialty of first author | Country | |
|---|---|---|---|---|---|---|
| 97 | Kleiss et al. (2013) | Diagnostic | Facial assessment by computer evaluation (FACE)<br>Glasgow Facial Palsy Software | ENT | United States | |
| 98 | Niziol et al. (2014) | Review | | | UK | |
| 99 | Placheta et al. (2014) | Case series | Unnamed landmark measurement system | Plastic surgery | Austria | |
| 100 | Couch et al. (2014) | Case series | Sunnybrook<br>Facial Clinimetric Evaluation Scale | Ophthalmology | United States | |
| 101 | Dall'Angelo et al. (2014) | Case series | Sunnybrook | Physiotherapy | Italy | Bell's palsy |
| 102 | Lee et al. (2015) | Case series | Sunnybrook | ENT | Korea | |
| 103 | Dalla Toffola et al. (2014) | Case series | House-Brackmann | Physiotherapy | Italy | Tumour |
| 104 | Mancini et al. (2014) | Prognostic | House-Brackmann | ENT | Italy | Bell's palsy |
| 105 | Fattah et al. (2014) | Cross-sectional survey | | Plastic surgery | UK | |
| 106 | Bianchi et al. (2014) | Case series | Functional and aesthetic grading system | Maxillo-Facial Surgery | Italy | Congenital |
| 107 | Lindsay et al. (2014) | Case series | Facial grading scale<br>Facial clinimetric Evaluation (FACE)<br>Facial assessment by computer evaluation (FACE) | ENT | United States | Vestibular schwannoma |
| 108 | Bhama et al. (2014) | Case series | Facial clinimetric Evaluation (FACE) | ENT | United States | Skin cancer |
| 109 | Hontanilla et al. (2014) | Case series | FACIAL CLIMA | Plastic surgery | Spain | Bell's palsy |
| 110 | Volk et al. (2014) | | 3D Ultrasound scan | ENT | Germany | Tumour |
| 111 | Fattah et al. (2015) | Review | | Plastic surgery | UK | |
| 112 | Yoshioka (2015) | Case series | Sunnybrook | Plastic surgery | Japan | Bell's palsy |
| 113 | Fujiwara et al. (2015) | Prognostic | Sunnybrook | ENT | Japan | Bell's palsy |

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**Table 3** (*continued*)

| | Title | Study design | Outcome measure used | Specialty of first author | Country | |
|---|---|---|---|---|---|---|
| 114 | Chuang et al. (2015) | Case series | Post-paralysis facial synkinesis examination sheet | Plastic surgery | Taiwan | Bell's palsy |
| 115 | Leonetti et al. (2015) | Diagnostic | Unnamed landmark measurement system | ENT | United States | Tumour |
| 116 | Haykal et al. (2015) | Case series | Facial assessment by computer evaluation | Plastic surgery | Canada | |
| 117 | Zhang et al. (2015) | Non-randomized controlled trial | House-Brackmann | Neurosurgery | China | Acoustic neurinoma |
| 118 | Pourmomeny et al. (2015) | RCT | Sunnybrook | Physiotherapy | Iran | Bell's palsy |
| 119 | Banks et al. (2015) | Diagnostic test study | eFACE | ENT | United States | Bell's palsy |
| 120 | Di Stadio (2015) | Diagnostic | ADS | ENT | Italy | Bell's palsy |
| 121 | Wasano et al. (2016) | Diagnostic | Yanagihara | ENT | Japan | Bell's palsy |
| 122 | Kleiss et al. (2016) | Diagnostic | SAQ<br>Sunnybrook<br>House-Brackmann | ENT | Netherlands | Bell's palsy |
| 123 | Wei and Diels (2016) | Case series | SAQ<br>Sunnybrook | Ophthalmology | United States | Bell's palsy |
| 124 | Mandrini et al. (2016) | Case series | Sunnybrook | Physiotherapy | Italy | Bell's palsy |
| 125 | Albathi et al. (2016) | Case series | Smile recovery scale<br>Facial asymmetry index<br>House-Brackmann | ENT | United States | Tumour |
| 126 | Kochhar et al. (2016) | Case series | Facial assessment by computer evaluation<br>House-Brackmann | ENT | United States | |
| 127 | Kim and Choi (2016) | RCT | House-Brackmann<br>Sunnybrook | ENT | South Korea | Bell's palsy |
| 128 | Mohamed et al. (2016) | Case series | House-Brackmann | ENT | Japan | Tumour |
| 129 | Socolovsky et al. (2016) | Case series | House-Brackmann | Neurosurgery | Argentina | |
| 130 | Sinha et al. (2016) | Case series | House-Brackmann | Ophthalmology | United States | Bell's palsy |
| 131 | Jindal and Jindal (2016) | Case series | House-Brackmann | Physiotherapy | India | Bell's palsy |

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**Table 3** (*continued*)

| | Title | Study design | Outcome measure used | Specialty of first author | Country | |
|---|---|---|---|---|---|---|
| 132 | Stankovic and Colovic (2016) | Non-randomized controlled trial | Sunnybrook | Physiotherapy | Serbia | Bell's palsy |
| 133 | Gaudin et al. (2016) | Diagnostic | FDI<br>eFACE | ENT | United States | |
| 134 | Banks et al. (2016) | Diagnostic | Sunnybrook<br>eFACE | ENT | United States | |
| 135 | Malhotra et al. (2016) | Diagnostic | CADS | Ophthalmology | UK | Bell's palsy |
| 136 | Tan et al. (2016) | Diagnostic | CADS | Ophthalmology | UK | Bell's palsy |
| 137 | Neville et al. (2017) | Case series | SAQ | Physiotherapy | UK | |
| 138 | Patel et al. (2017) | Case series | SAQ | ENT | United States | Vestibular schwannoma |
| 139 | Thomas et al. (2017) | RCT | SAQ | ENT | United States | |
| 140 | Fujiwara et al. (2017) | Case series | Sunnybrook | ENT | Japan | Herpes zoster |
| 141 | Kasahara et al. (2017) | Non-randomized controlled trial | Sunnybrook | Physiotherapy | Japan | Bell's palsy |
| 142 | Maria and Kim (2017) | Case series | Sunnybrook | ENT | Korea | Bell's palsy |
| 143 | Azuma et al. (2017) | Diagnostic | Unnamed landmark measurement system | ENT | Japan | Healthy individuals |
| 144 | Meier-Gallati and Scriba (2017) | Diagnostic | OSCAR | ENT | Switzerland | Bell's palsy |
| 145 | Akulov et al. (2017) | RCT | Fisch<br>House-Brackmann<br>House-Brackmann<br>Yanagihara<br>Sunnybrook | Neurosurgery | Russia | Tumour |
| 146 | Biglioli et al. (2017) | Case series | House-Brackmann | Maxillo-Facial Surgery | Italy | Iatrogenic |
| 147 | Biglioli et al. (2017) | Case series | House-Brackmann | Maxillo-Facial Surgery | Italy | Bell's palsy |
| 148 | Choi et al. (2017) | Prognostic | House-Brackmann | ENT | South Korea | Bell's palsy |
| 149 | Yetiser (2017) | Case report | House-Brackmann | ENT | Turkey | Petrosectomy |
| 150 | Yoshioka (2017) | Case series | House-brackmann | Neurosurgery | Japan | Tumour |

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**Table 3** (*continued*)

| | Title | Study design | Outcome measure used | Specialty of first author | Country | |
|---|---|---|---|---|---|---|
| 151 | Aranha et al. (2017) | Case report | Sunnybrook | Physiotherapy | India | Bell's palsy |
| 152 | van Veen et al. (2017) | Case series | Facial Clinimetric Evaluation (FACE)<br>SAQ | Plastic surgery | Netherlands | Bell's palsy |
| 153 | Jowett et al. (2017) | Case series | Unnamed landmark measurement system | ENT | United States | Bell's palsy |
| 154 | Chong et al. (2017) | Diagnostic | eFACE<br>FDI<br>Sunnybrook<br>House-Brackmann<br>Sydney | ENT | United States | Bell's palsy |
| 155 | Jowett et al. (2017) | Case series | eFACE<br>House-Brackmann | ENT | United States | Lyme's disease |
| 156 | Phillips et al. (2017) | Prognostic | eFACE<br>House-Brackmann | ENT | United States | Bell's palsy |
| 157 | Banks, et al. (2017) | Diagnostic | eFACE | ENT | United States | |
| 158 | Banks. et al. (2017) | Diagnostic | eFACE | ENT | United States | |
| 159 | Cooper et al. (2018) | Case series | Sunnybrook | Plastic surgery | UK | Bell's palsy |

- Responsiveness: the ability of the instrument to detect minor but clinically relevant subtleties, being this particularly relevant for patients undergoing regular monitoring.

To the knowledge of the authors, this article contains the only systematic review, to date, focusing on facial synkinesis and the instruments that have been used to measure this condition.

It can be noted that the number of articles on facial synkinesis being published each year has been steadily growing. In part, this could follow a general tendency in medical literature, with an exponential increase of publications in recent years.[21] However, it might also reflect a recent recognition of the importance of synkinesis in the literature

The predominance of otorhinolaryngology, as the leading specialty in terms of research related to facial synkinesis needs to be appraised carefully. Current and future systematic reviews on this subject could potentially over-represent the preferences, practice and views of ENT specialists. This potential bias does not just reside in the authorship of the published studies, but also in the fact that there is a large proportion of these published in otorhinolaryngology journals. Over-representation could imply, for example, the recruitment of patients with pathologies usually referred to this specialty, such as Ramsay-Hunt syndrome, facial and vestibular nerve neuromas and middle ear aetiologies in despite of more common causes, such as Bell's palsy.

In terms of the quality of the evidence found in this systematic review, the majority of the study designs fall in the lower categories as per the Oxford Centre for Evidence-Based Medicine levels of evidence. While 51% of the studies were case series and case reports, only 10% of these were randomised controlled trials.

By far, the two most commonly used instruments were the House–Brackmann Facial Nerve Grading System and the Sunnybrook Facial Grading System, present in 46% and 31% of the primary research articles found, respectively. These were then followed by an atomised variety of measuring scales, without any of these being able to surpass 6% of presence among eligible studies. In total, 27 different named instruments were identified, which can be classified in four major groups:

- Clinician-based questionnaires
- Patient-reported outcomes
- Automatic computerised analysis
- Manual objective measurements.

Clinician-based reporting outcomes were the category with highest representation in the sample of studies included in our review. This seems to be not only a tendency affecting scientific reports, but also replicates in clinical

practice according to a recent survey organised by the Sir Charles Bell Society.[8]

The House–Brackmann scale[16] consists in a six mutually-exclusive categories, describing several degrees of severity. These contain a clinical description of gross disfigurement, dynamic disturbances and asymmetry at rest that are associated with each category. In relation to facial synkinesis, this classification does not include a formal assessment of this feature, although it is briefly mentioned in its second and third degree, with no description included in the more severe grades. The formal application of the House-Brackmann scale consists in trying to classify patients to one of these six-categories. This inevitably presents a challenge for clinicians, particularly considering that the signs and symptoms of facial palsy do not always follow a linear pattern, with patients presenting sequalae of different severity in different territories or affecting different functions.

An amended House–Brackmann system was proposed in 2009 to address these issues,[22] including a formal assessment of weakness and synkinesis along with a new scoring system. However, for the purpose of this study it was challenging to ascertain if studies claiming to use the House-Brackmann system were using the 1985 version or the 2009 modified one. For this reason, it was impossible to count their appearances in the literature as two separate entities.

The Sunnybrook, or Toronto, Facial Grading System[17] is another clinician-based assessment system has three sections that aim to measure resting symmetry, voluntary movement and synkinesis. For each dimension, a topographical approach is used to evaluate different facial areas, resulting in a series of marks for each section. The obtained scores are then weighted using an equation that considers dynamic symmetry the most important aspect. This results in a composite score that ranges from 100, for normal facial nerve function, to 0 in cases of complete paralysis.

Even though it can be argued that the weighting of these three different components is arbitrary, the Sunnybrook system presents two main advantages compared the House-Brackmann scale. First, it formally assesses signs of facial synkinesis from the clinician's point of view, feature which is absent in the original House–Brackmann classification. Secondly, its continuous scoring system is able capture more subtleties than fixed 6 categories instrument. This allows not only a more detailed documentation of a patient's progression, but also presents an advantage for researchers interested in comparing different treatment options.

The two most commonly utilised patient-reported outcomes found in our review were the Synkinesis Assessment Questionnaire (SAQ) and the Facial Clinimetric Evaluation Instrument (FACE). The former consists in a ten-question survey for patients to score the severity of their symptoms. All these queries focus on synkinesis-related symptoms, resulting in a global score that ranges from 20 to 100.[19] The FACE is a more comprehensive instrument, including 15 Likert-style statements that patients are asked to grade, depending on their frequency and severity. These 15 questions include: three statements focusing on difficulties when attempting particular facial expressions; three assessing synkinesis-related symptoms; three measuring ophthalmologic sequalae; two appraise eating-related difficulties; and four focus on the social impact of facial palsy.[23]

Even though the Facial Assessment by Computer Evaluation developed by Hadlock and Urban[24] and the PEAK Motion Measurement System described by Linstrom et al.[25] were used in 5 and 4 articles, respectively; the use of automatic computerised assessment systems is not widespread in the literature.

The Delphi survey conducted for this study was able to recognise a consensus among five essential elements that an ideal synkinesis grading system should include:

- Overall subjective assessment of facial synkinesis done by clinician.
- Regional subjective assessment of synkinesis done by clinician.
- Overall resting symmetry done by clinician.
- Self-assessment of synkinesis related symptoms by patients.
- Self-assessment of impact of synkinesis in daily activities done by patients.

The Sunnybrook Facial Grading System, second most popular instrument in our systematic review, has proven to be a useful and reliable outcome measure since its introduction two decades ago, demonstrating its validity, reproducibility and responsiveness.[6,26] The Sunnybrook system includes a thorough assessment of resting symmetry and synkinesis, overall and in specific territories, which were consensus features in our Delphi study. It is important to mention that this instrument was also the most popular among the panel of experts. However, in order to fully comply with the results of our research, the Sunnybrook Facial Grading System requires the complementation of a patient-reported outcomes element.

It seems reasonable to use instruments that consider the patients' views, preferences and satisfaction for monitoring their care and conduct clinical research.[27] The SAQ and FACE are patient-based questionnaires that have demonstrated good reliability, responsiveness and validity for assessing patients with facial palsy.[19,23,28] These are specific for patients with facial palsy, which is not the case for the more generic Facial Disability Index (FDI). Both, the SAQ and FACE are able to assess the remaining elements that the Sunnybrook system fails to. While the SAQ provides an in-depth assessment of synkinesis it lacks the breadth that FACE provides, considering that the latter covers not only synkinesis, but also physical, psychological and social aspects associated with this condition.

Several limitations can be identified in this study, particularly related to low response rates, which are a well-recognised challenge for this type of studies.[29] There is a strong case to support the argument that a panel of 11 experts is far from being representative of a much more numerous specialist population. Strategies to confront this would have been to pay for participation or to organise a face-to-face initial round, implying a financial and logistic cost. Even though it would have been reassuring to have a better recruitment rate, this does not invalidate the responses received from the experts that kindly replied to our invitation.

To the knowledge of the authors, there is no clear answer regarding how Delphi studies should address potential biases arising from poor recruitment and arbitrary definition of consensus. For this reason, the author has intended

to detail the exact methodology applied, leaving further interpretation of these issues to the readers of this article.

Despite its limitations, we believe that this study demonstrates the importance of measuring synkinesis in the context of an overall assessment of facial palsy by the clinician, while including patient's views in the evaluation of this condition. Associated utilisation of the Sunnybrook Facial Grading System and a patient reported outcomes instrument, such as the FACE, is able to provide a thorough appraisal of this condition. In this way, the clinical examination incorporated in the Sunnybrook instrument can be complemented by a self-reported patient evaluation of their symptoms, disability and impact on their quality of life.

So far, joint use of these instruments has been so far an exception in the literature, with only two studies incorporating this design. Despite its limitations, to the knowledge of the author, this study is the first research project that has focused on the way facial synkinesis has been historically reported in the literature, making a proposal for uniformity.

The introduction of information technologies for the measuring of facial palsy and synkinesis is an ongoing phenomenon at present. The author and the recruited panel of experts agree on the statement that the utilisation of a reliable objective system could revolutionise the way we monitor patients suffering from facial disability. However, it seems that, so far, such instrument is still not available. Until then joint use of a clinician-based assessment through the Sunnybrook Facial Grading System and a patient-reported outcome should present a reliable and comprehensive alternative.

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