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Review

Assessing speech at three years of age in the cleft palate population: a scoping review of assessment practices

Beth Fitzpatrick†‡, Jane Coad†¶, Debbie Sell§ and Tanya Rihtman†‖

†Faculty of Health and Life Sciences, Coventry University, Coventry, UK
‡Speech and Language Therapy, Birmingham Children's Hospital, Birmingham, UK
§Centre for Outcomes and Experience Research in Children's Health, Illness and Disability, Great Ormond Street Hospital NHS Foundation Trust, London, UK
¶School of Health Sciences Nottingham University, Nottingham, UK
‖Faculty of Health and Life Sciences, Oxford Brookes University, Oxford, UK

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Abstract

Background: There is no consensus in the UK regarding the types of speech samples or parameters of speech that should be assessed at 3 years of age in children with cleft palate ± cleft lip (CP±L), despite cleft units routinely assessing speech at this age. The standardization of assessment practices would facilitate comparisons of outcomes across UK cleft units; earlier identification of speech impairments—which could support more timely treatments; and more reliable recording of therapy impacts and surgical interventions.
Aims: To explore assessment practices used to assess speech in 3-year-old children with CP±L, including speech parameters, methods of assessment and the nature of the speech sample used.
Methods & Procedures: A broad examination of the literature was undertaken through the use of a scoping review conducted in accordance with Joanna Briggs Institute guidelines. Search terms were generated from a preliminary search and then used in the main search (Medline, CINAHL, Embase, AMED and PsycINFO).
Main Contribution: A combination of approaches (medical, linguistic, developmental and functional) is required to assess CP±L speech at age 3. A developmental approach is recommended at this age, considering the complexity of speech profiles at age 3, in which typically developing speech processes may occur alongside cleft speech characteristics. A combined measure for both nasal emission and turbulence, and an overall measure for velopharyngeal function for speech, show potential for assessment at this age. Categorical ordinal scales are frequently used; the use of continuous scales has yet to be fully explored at age 3. Although single-word assessments, including a subset of words developed for cross-linguistic comparisons, are frequently used, more than one type of speech sample may be needed to assess speech at this age validly. The lack of consensus regarding speech samples highlights a need for further research into the types of speech samples 3-year-olds can complete; the impact of incomplete speech samples on outcome measures (particularly relevant at this age when children may be less able to complete a full sample); the impact of different speech samples on the validity of assessments; and the reliability of listener judgements.
Conclusions & Implications: Whilst a medical model and linguistic approaches are often central in assessments of age-3 cleft speech, this review highlights the importance of developmental and functional approaches to assessment. Cross-linguistic single-word assessments show potential, and would facilitate the comparison of UK speech outcomes with other countries. Further research should explore the impact of different speech samples and rating scales on assessment validity and listener reliability.

Keywords: cleft palate, cleft lip and palate, speech assessment, speech analysis, age 3 years, preschool.

Address correspondence to: Tanya Rihtman, Faculty of Health and Life Sciences, Oxford Brookes University, Jack Straws Lane, Marston, Oxford, OX3 0FL; e-mail: trihtman@brookes.ac.uk

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What this paper adds

What is already known on the subject

Although speech is typically assessed at 3 years of age in UK cleft units, assessment methods vary. This prevents cross-unit outcome comparisons and is a barrier to measuring the impact of therapy and surgical interventions. There is a need to explore assessment practices broadly to guide the direction of assessments at 3 years of age in the UK.

What this paper adds to existing knowledge

This review highlights the importance of a developmental approach to assessment in the assessment of children with CP±L at 3 years of age. Whilst there is consensus as to the parameters of assessment, there is no such consensus regarding speech samples or methods of assessment.

What are the potential or actual clinical implications of this work?

Children at age 3 are at a unique stage of speech development. Assessment procedures and outcomes used with older age groups require adaptation to meet the needs of 3-year-olds in terms of both the speech sample used and the need to consider developmental and functional outcomes. Whilst further research is needed to provide a foundation for decisions regarding the selection of speech samples, and the impact of different speech samples and methods of assessment on the reliability of listener judgements, cross-linguistic single-word assessments show potential and would facilitate international comparisons of outcomes.

Introduction

The speech outcome of individuals with a repaired cleft palate ± cleft lip (CP±L) is a central focus of all cleft teams, as speech outcomes are a primary measure of surgical success (Grunwell and Sell 2001: 68). To this end, a significant focus of cleft research has been on developing speech assessment procedures and outcome measures. Perceptual speech assessment with a basis in phonetic transcription is described as the 'gold standard' for the assessment of speech in individuals with CP±L (Howard 2011: 127). However, there are significant challenges in designing assessment procedures and protocols which allow for speech outcomes to be assessed over a lifespan, as particular speech samples and assessment materials may be more appropriate at certain ages, and assessment objectives may differ across age groups. For example, in children over 5 years and adults speech assessment may focus on the identification of persisting speech difficulties and the impact of anatomical changes (e.g., adenoid atrophy, dentition and occlusion) on speech production (Sell and Pereira 2015). However, for younger children, a focus of assessment may be to examine the impact of the cleft on the process of normal speech sound development (phonology as well as articulation) (Chapman and Willadsen 2011: 25).

Existing protocols such as the Cleft Audit Protocol for Speech—Augmented (CAPS-A) (John *et al.* 2006, Sell *et al.* 2009) and the Americleft modification of the CAPS-A, CAPS-A-AM (Chapman *et al.* 2016), have only been validated and tested for reliability on children aged 5 and above. This is understandable given the original intended use of the CAPS-A in the audit of speech outcomes at age 5 years, an established age for assessing speech outcomes, hence the focus of much attention in the last two decades. Only the Swedish Articulation and Nasality Test (SVANTE) (Lohmander *et al.* 2005, 2009), a valid and reliable assessment protocol available in Swedish and Norwegian, has been developed for use with children under 5 years with CP±L (Lohmander *et al.* 2017a). The Universal Parameters (UPS) (Henningsson *et al.* 2008), and the Pittsburgh Weighted Values for Speech Symptoms Associated with Velopharyngeal Incompetence (PWSS) (McWilliams and Philips 1979) are alternative assessment protocols; however, neither has been comprehensively validated (Prathanee *et al.* 2011, Dudas *et al.* 2006).

Although UK cleft units routinely carry out speech assessments at age 3, unlike age 5, there is no agreed assessment procedure or outcome measures. Agreed procedures and outcome measures at age 3 would have several benefits. First, valid and reliable outcome measures could be used to identify children at risk of poor speech outcomes at age 5 and encourage timely referral for intervention. Outcome measures would also facilitate the comparison of outcomes across cleft units before 5 years, with the potential to identify variability in outcomes and implement quality improvement initiatives in a similar way to established practices in the UK at age 5 (Cleft Registry and Audit Network (CRANE) 2019). In addition, it could permit the collection of comparative longitudinal data, providing an understanding of the impact of therapy and secondary surgery for speech through the comparison of outcomes at ages 3 and 5.

One well-recognized challenge in the assessment of speech in 3-year-olds is that at this age children go through a period of significant developmental change, refining and acquiring new skills related to their attention, language and speech (Dosman *et al.* 2012, McLeod

and Baker 2017: 202). As such, 3-year-old children can be highly variable in how they present in these areas despite being within normal developmental limits. Furthermore, variability in presentation can also result from the known delay in expressive language and speech (Cavalheiro *et al.* 2019) and an increased risk of neurodevelopmental disorders (Tillman *et al.* 2018). This variability presents a challenge when designing assessment protocols, in particular the selection of speech samples and the parameters of speech which can be assessed reliably. Given that children's speech sound systems are in a stage of developmental change (irrespective of the cleft palate), it also raises the question as to the focus of the assessment for children with cleft, that is, whether this focuses purely on those parameters considered core to assessment of cleft speech and/or should include an assessment of speech from a developmental perspective.

A broad scope of enquiry was required to examine the methods, parameters of assessment (whether developmental or cleft specific) and speech samples used in the assessment of speech in children with CP±L at age 3. This was achieved through the use of a scoping review methodology (Khalil *et al.* 2016). This methodology was selected given that the intention behind the work was to inform the future development of assessment procedures at age 3 in the UK. Rather than answer an effectiveness question (Peters *et al.* 2015), evaluate study quality or limit the review to a particular study design (Arksey and O'Malley 2005) or country of origin, the scoping review methodology enabled inclusion of a broad range of resource types, and the broad mapping of current practices, in order to clarify the key concepts, identify gaps in the evidence base and make recommendations for future practice in the UK. In doing so, the review identified the extent to which there is a consensus in the parameters of assessment, procedures and methods; differences and similarities in assessment at 3 years of age compared with practice at age 5; and the types of speech samples used (Munn *et al.* 2018).

Methods

Ethical review

Approval for the study was gained in accordance with the ethics and governance procedures at Coventry University (study number P68435).

Design

To strengthen methodological rigour and to allow replication, the scoping review was undertaken using the Joanna Briggs Institute (JBI) guidance for scoping reviews (Peters *et al.* 2015) and followed the process outlined below.

Objective/aim

To map the parameters of speech and types of speech samples used to assess speech in 3-year-old children with CP±L, with reference to the rating scales and methods used to assess them, and to consider key differences in speech assessments at age 3 and older age groups in order to inform the development of a UK assessment protocol.

Scoping review objectives

- To explore the parameters of speech typically assessed in 3-year-old children with CP±L, and to consider if they are core to the assessment of individuals with CP±L.
- To explore the methods and rating scales used to assess the identified parameters of speech.
- To explore the types of speech samples are used in the assessment of 3-year-old children with CP±L.
- To discuss how the parameters of speech assessment map onto different theoretical approaches to assessment.

Inclusion criteria

The inclusion criteria were developed with reference to the JBI methodology as follows:

- *Types of participants: children aged 3 with CP±L*. For inclusion in the review, the literature had to include specifically 3-year-olds with details provided as to how speech was assessed at this age (either the parameters of speech, the measurement scales used and/or the speech sample). This resulted in the inclusion of studies also assessing children at other ages, that is, longitudinal studies; however, only data referencing assessment at age 3 were included in the review.
- *Concept*: literature and sources had to address the following concepts: speech assessments, the types of speech samples and speech parameters assessed, including methods of assessment. This allowed for the inclusion of sources which investigated assessment processes as well as those reporting on speech outcomes.
- *Context*: to broaden the examination, no preference was given to sources from any specific country; however, resources were limited to English, or to those with an available translation. It was considered that the development of the Great Ormond Street Speech Assessment (GOS.SP.ASS'98) (Sell *et al.* 1999) marked the start of a new era in speech assessments and outcome measures; this informed the timeframe, with studies reviewed from 1998 to 2018.

- *Types of sources*: sources needed to be sufficiently detailed to enable the extraction of sufficient information about assessment procedures, therefore conference abstracts were excluded.
- *Search strategy*: in accordance with Peters *et al.* (2015), an initial limited database search was conducted using the EMBASE database to consolidate relevant search terms from the title, abstract and keywords of identified studies and explore controlled vocabulary. Cleft specific search terms as well as general terms such as 'speech assessment' were subsequently included. The final list of search terms is shown in table 1. Relevant medical subject headings (MeSH; a vocabulary used to index publications) were used in the database search. The electronic databases Medline, Cumulative Index of Nursing & Allied Health Literature (CINAHL), Embase, AMED and PsycINFO were used. Citation tracking was used to identify additional relevant sources.
- *Resource selection*: figure 1 outlines the study selection processes using the flowchart advised in the PRISMA Scoping Review Extension (PRISMA-ScR) flowchart (Tricco *et al.* 2016). Abstracts were screened for relevance to the objectives of the review and the inclusion criteria, first by title, then by abstract and, if necessary, using the full text. With the agreement of the research team and in an exception to the inclusion criteria, the normative data developed using the SVANTE assessment (Lohmander *et al.* 2017a) was included. Whilst the outcomes presented were not related to children with CP±L, the assessment protocol which was primarily designed to assess structurally based speech difficulties (i.e., cleft palate) and is suitable for children aged 3, provides a detailed account of the assessment procedure and is highly relevant to the objectives of this review. Consideration was also given to a large ongoing research project taking place in the UK, The Cleft Collective Speech and Language Study (https://www.nbt.nhs.uk/bristol-speech-language-therapy-research-unit/bsltru-research/cleft-speech-language-study). As part of the development of the study, a national survey had taken place regarding assessment practices in UK cleft centres at age 3. Given the specific relevance of this survey to the aims of this review, with the agreement of the research team, this was included and is referred to as Wren (2013).
- The final sources included in the review were discussed and confirmed by paired members of the research team. All but one were research papers that appeared in peer-reviewed journals.

Table 1. Search terms

Participant type		
Age	Diagnosis	Concept
preschool	cleft palate	articulation
toddler	cleft lip and palate	cleft speech characteristics
kindergarten		cleft type characteristics
nursery		compensatory articulation
3 years old		velopharyngeal dysfunction (VPD)
aged 3 years		velopharyngeal insufficiency (VPI)
3 years of age		voice quality/dysphonia
		speech articulation
		speech analysis
		speech assessment
		speech and language assessment

Charting the results

Studies meeting the inclusion criteria were read in full and charted according to the country of origin, study aim, participant details, methodology and methods, details of the speech assessment and speech sample, the parameters of speech assessed and the rating scales used. The parameters of speech assessed across the CAPS-A, CAPS-A-AM, SVANTE, PWSS and UPS provided a framework for analysis defining those parameters core to the assessment of speech in the CP±L population.

Results

A total of 35 sources were reviewed in the scoping review. These are listed in appendix A and detailed in table 2.

Demographic information

The sources originated from 12 countries (figure 2). A total of 34% originated from Sweden, with Scandinavian countries producing 48% of the sources reviewed.

Apart from one, almost all sources included were primary research and recruited samples of children with CP±L. The subtypes are presented in table 2. It is noteworthy that many of the Scandinavian sources were part of/sub-studies relating to the Scandcleft Trial (Lohmander *et al.* 2017a, 2017b, Willadsen *et al.* 2017), a large multicentre cross-linguistic randomized control trial, evaluating four different surgical protocols. The Scandcleft Trial only recruited participants with unilateral cleft lip and palate (UCLP), contributing to the high number of sources ($n = 13/33$; 39.4%), which only included participants with UCLP. Two sources did not record cleft type (Wren 2013 reporting assessment practices; Lohmander *et al.* 2017a reporting on non-cleft children) and were excluded in calculations regarding cleft type.

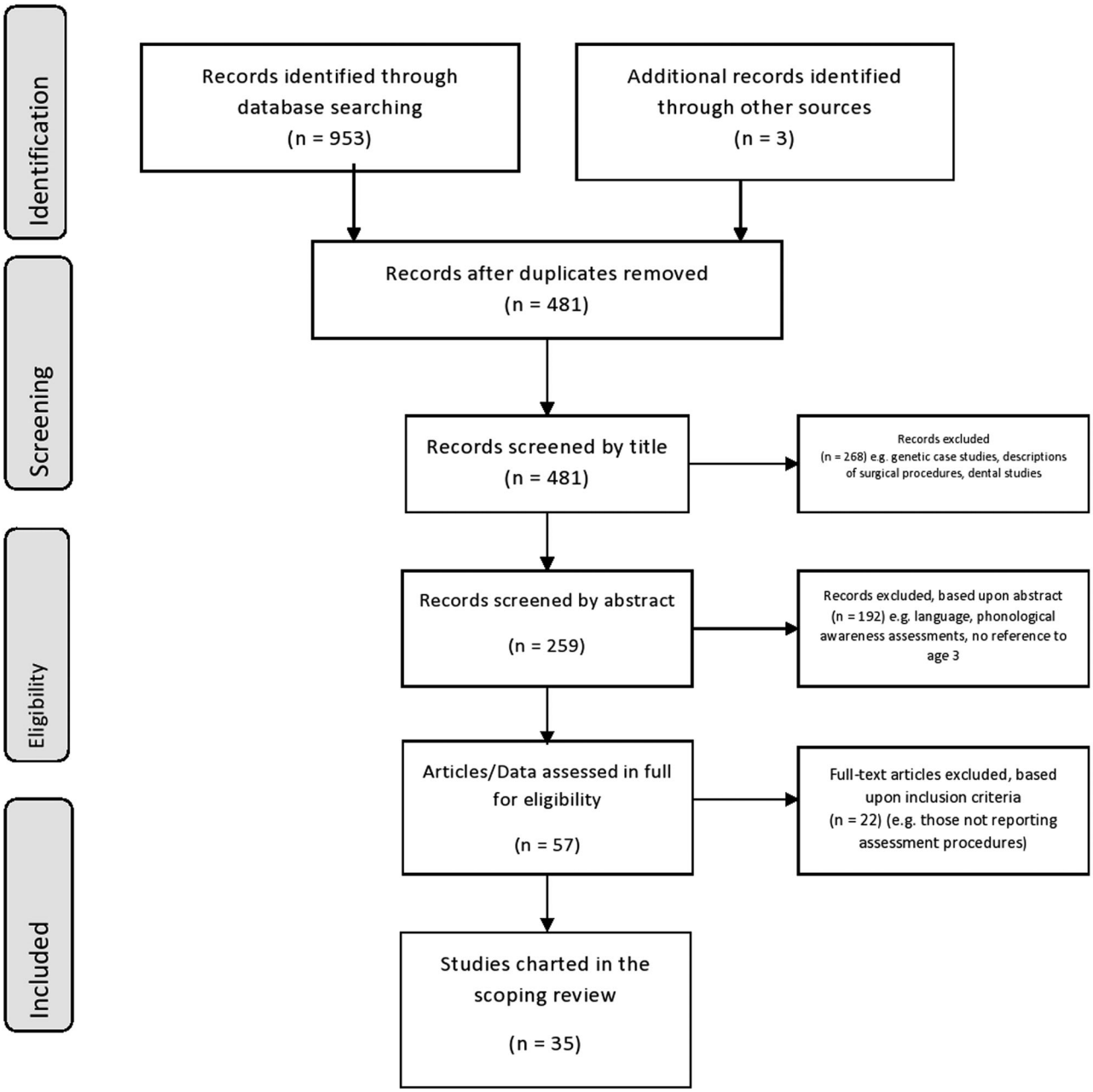

Figure 1. Adapted PRISMA Extension for Scoping Reviews (PRISMA-ScR) (Tricco *et al.* 2016) Flow Diagram selection of sources.

Which parameters of speech are typically assessed in 3-year-old children with CP±L, and are they core to the assessment of individuals with CP±L?

The parameters of speech assessed in each source are charted in table 3. Each source reported parameters that addressed their specific research aims. As such, not all sources assessed all the parameters identified in this review. Therefore, where percentages are presented, this refers only to those sources which directly assessed the parameter in question.

Consonant production

Almost all sources assessed consonant production. Although phonetic transcription underpinned the assessment of xxxxxx, a variety of methods were used (figure 3).

The first principal method used to assess consonant production was to report summary patterns by grouping errors according to their place of articulation, or the broader categories of passive or active characteristics (see Hutters and Brøndsted 1987 for a description of

Table 2. Papers included in the review recorded by author and date, country of origin, methods and methodology, and speech assessment

Author (date)	Country of origin	Age of the participants	Cleft type*	Methodology and methods	Speech assessment
Frey *et al.* (2018)	USA	15–36 months	CPL	CPL recruited from a larger randomized experimental group design	Spontaneous speech sample
Willadsen *et al.* (2018)	Denmark	36 months, mean = 3.0 years (range = 2.83–3.23 years)	UCLP NCCG	Randomized control trial (randomized by age at hard palate repair)	Single-word naming using the naming test developed in the Scandcleft study (Lohmander *et al.* 2009)
Chacon *et al.* (2017)	Australia	3-year-olds: 2;10–3;11 years 5-year-olds: 4;10–5;09 years	CPO UCLP BCLP	Cross-sectional observation study	GOS.SP.ASS × six sentences DEAP Articulation subtest (Dodd *et al.* 2002), single words Informal list of 15 monosyllabic and 10 polysyllabic words
Larsson *et al.* (2017)	Sweden	Adopted group: 38 months (range = 35–43 months); control group (also cleft): 37 months (range = 34–42 months)	UCLP	Cohort study	SVANTE (Lohmander *et al.* 2005, 2015). Single-word naming and spontaneous speech sample.
Lohmander *et al.* (2017a)	Sweden	Normative data gained at 3, 5, 7, 10, 16 and 19 years 3-year-olds: mean age 2;11 years (range = 2;8–3;3 years)	NCCG—normative data	Normative referencing	SVANTE (Lohmander *et al.* 2005, 2015). Single-word naming, sentence repetition, spontaneous speech sample
Raud Westberg *et al.* (2017)	Sweden	Longitudinal assessment at ages 1, 1;6 and 3 years	UCLP	Randomized control trial	Single-word naming using the naming test developed in the Scandcleft study (Lohmander *et al.* 2009)
Safaiean *et al.* (2017)	Iran	3–5 years	CPL NCCG	Cohort study	Persian Speech Intelligibility Test (Heydari *et al.* 2011) which involves picture naming
Swanson *et al.* (2017)	USA	Initial assessment at age 3 years	SMCP	Retrospective review	Pittsburgh Weighted Speech Scoring (McWilliams and Philips 1979) (no information provide as to what the speech sample was)
Klintö *et al.* (2016)	Sweden	Mean = 36 months (range = 35–38 months)	UCLP NCCG	Randomized control trial (some participants) Prospective longitudinal	Single-word naming using the naming test developed in the Scandcleft study (Lohmander *et al.* 2009)

(Continued)

Table 2. Continued

Author (date)	Country of origin	Age of the participants	Cleft type*	Methodology and methods	Speech assessment
El Ezzi *et al.* (2015)	Switzerland	3 years (no specific age range)	UCLP CPO	Retrospective cohort study	Not stated
Klintö *et al.* (2015) (the same procedure as Klintö *et al.* 2014a)	Sweden	Median = 36 months (range = 35.5–37 months)	UCLP NCCG	Prospective comparative study at 5 years and retrospective comparative study at 3 years	Single-word naming using the naming test developed in the Scandcleft study (Lohmander *et al.* 2009)
Klintö *et al.* (2014a)	Sweden	Median = 36 months (range = 35.5–37 months)	UCLP NCCG	Randomized control trial (randomized by age at hard palate repair)	Single-word naming using the naming test developed in the Scandcleft study (Lohmander *et al.* 2009) Sample of spontaneous speech
Klintö *et al.* (2014b)	Sweden	Group 1: mean age = 36 months Group 2: mean age = 36.9 months	UCLP	Prospective comparison study	Single-word naming using the naming test developed in the Scandcleft study (Lohmander *et al.* 2009) Sample of spontaneous speech
Wren (2013)	UK	n.a. Report on assessment practises of speech and language therapists	n.a.	Survey of 14 UK cleft units (% of cleft units)	South Tyneside Assessment of Phonology (STAP) (Armstrong and Ainley 1992): 21.42% Diagnostic Evaluation of Articulation & Phonology (Dodd *et al.* 2002): 28.57% PACS TOYS (Harding and Grunwell 1995): 28.57% Phonological Screening Assessment (PSA) (Stevens and Isles 2001): 14.28% Hart Screen Speech Assessment: 14.28% CLEAR Phonology Screening Assessment (CLEAR Resources 2006): 28.57% Great Ormond Street Speech Assessment (GOS.SP.ASS) (Sell *et al.* 1999): 85.71% Renfrew Action Picture Test (Renfrew 2011): 14.28% Preschool Language Scale Version 3 (PLS-3) (Boucher and Lewis 1997): 21.42% Spontaneous speech: 64.28%
Willadsen (2012)	Denmark	3-years-old group: 36 months	UCLP	Randomized control trial (age at hard palate repair)	Single-word naming using the naming test developed in the Scandcleft study (Lohmander *et al.* 2009)
Willadsen and Poulsen (2012)	Denmark	36 months	UCLP NCCG	Cross-sectional study	Single-word naming using the naming test developed in the Scandcleft study (Lohmander *et al.* 2009)

(Continued)

Table 2. Continued

Author (date)	Country of origin	Age of the participants	Cleft type*	Methodology and methods	Speech assessment
Dayashankara *et al.* (2011)	India	Mean age = 27.7 months	BCLP UCLP CPO	Prospective cohort study	Not stated
Hamming *et al.* (2009)	USA	Younger age group: 3–4 years (up to 4.11 years)	UCLP BCLP CPO SMCP	Retrospective longitudinal	Not stated
Chapman *et al.* (2008)	USA	33–42 months; median = 39 months	CPL	Prospective longitudinal	Goldman–Fristoe Test of Articulation (Goldman and Fristoe 1986), single-word naming, spontaneous speech sample
Gugsch *et al.* (2008)	Germany	Mean age at baseline = 3;11 years (range = 3.6–4.9 years)	UCLP BCLP CPO	Longitudinal prospective	Voice recordings in standardized form
Lohmander and Persson (2008)	Sweden	Longitudinal speech assessments at 18 months and 3, 5 and 7 years	UCLP NCCG	Prospective longitudinal study	Single-word picture naming test
Hodge and Gotzke (2007)	Canada	Age range = 3;5–6;7 years Mean = 4;6 years	UCLP BCLP CPO	Prospective between-groups design	Single-word repetition Spontaneous speech sample
Frederickson *et al.* (2006)	USA	Age range = 2;9–3;8 years	UCLP NCCG	Prospective cross-sectional study	Goldman Fristoe Test of Articulation (Goldman and Fristoe 1986), single-word naming
Lohmander *et al.* (2006)	Sweden	Longitudinal speech assessments at 3, 5, 7 and 10 years	UCLP	Prospective longitudinal study (3-year results retrospectively collected)	Assessment not stated at age 3, data collected from records
Persson *et al.* (2006)	Sweden	Longitudinal speech assessments at 3, 5, 7 and 10 years	CPO	Longitudinal study (3–7 years)	Single-word naming
		Age 3: mean age = 3;0 years (range = 3;0–3;3 years)	NCCG		Spontaneous speech if unable to complete naming

(Continued)

Table 2. Continued

Author (date)	Country of origin	Age of the participants	Cleft type*	Methodology and methods	Speech assessment
Chapman (2004)	USA	Longitudinal assessment, final assessment at 39 months	UCLP BCLP	Multi-site longitudinal study	Spontaneous speech sample
Konst *et al.* (2003)	The Netherlands	Longitudinal follow up at age 2, 2.6 and 3 years	UCLP	Randomized control trial	Spontaneous speech sample
Morris and Ozanne (2003)	Australia	Assessment at ages 2 and 3 years	UCLP BCLP CPO	Comparative groups—longitudinal	Spontaneous speech sample Goldman Fristoe Test of Articulation (Goldman and Fristoe 1986), single-word naming
Zanzi *et al.* (2002)	Switzerland	Age 3.5 years	UCLP BCLP	Retrospective review	Spontaneous speech/interview
Hattee *et al.* (2001)	UK	Longitudinal assessment at 9 months, 18 months and 3 years	UCLP CPO	Longitudinal study	South Tyneside Assessment of Phonology (STAP) (Armstrong and Ainley 1992)
Hutters *et al.* (2001)	Denmark	Mean = 3;1 years (range = 2;11–3;3 years)	UCLP BCLP	Prospective cross-sectional study	Picture naming
Pulkkinen *et al.* (2001)	Finland	Longitudinal assessment at 3, 6, and 8 years	UCLP CPO	Longitudinal	Not stated
Gunther *et al.* (1998)	USA	Assessment at age 3 years	CPL	Retrospective review	Pittsburgh Weighted Speech Scale (McWilliams and Philips 1979)—no information on type of speech sample
Lohmander-Agerskov (1998)	Sweden	Longitudinal assessment at 9 months, 18 months, and 3, 5, 7, 10, 16 and 19 years	UCLP BCLP	Results combined from a longitudinal study—retrospective and a cross-sectional study	Picture naming
Lohmander-Agerskov *et al.* (1998)	Sweden	Assessment at 3 and 5 years	UCLP BCLP CPO	Longitudinal study	Picture naming

Notes: UCLP, unilateral cleft lip and palate; BCLP, bilateral cleft lip and palate; CPO, cleft palate only; SMCP, submucous cleft palate; CPL, cleft palate ± cleft lip (used when the specific type of cleft is not stated); NCCG, non-cleft control group.
For additional references for assessments, see Armstrong and Ainley (1992), Boucher and Lewis (1997), CLEAR Resources (2006), Goldman and Fristoe (1986), Grunwell and Harding (1995), Heydari *et al.* (2011), Stevens and Isles (2011) and Renfrew (2011).

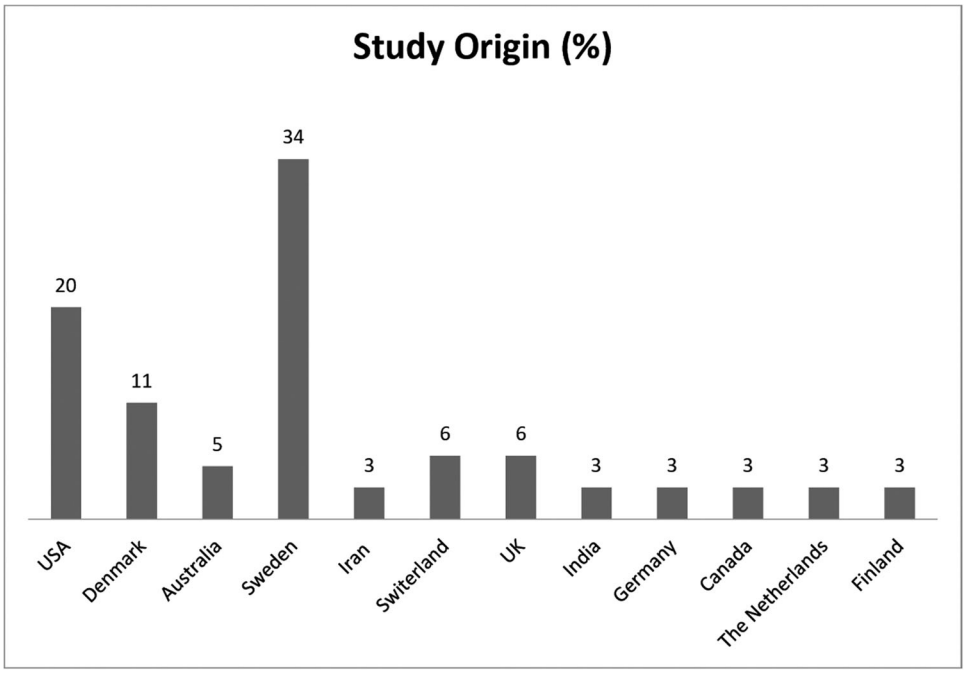

Figure 2. Study origin.

these processes). Such summary patterns can be viewed as a specific measure of cleft speech characteristics (CSCs) and feature in the CAPS-A, CAPS-A-M, UPS and SVANTE protocols. Despite a strong trend across the studies to report on consonant articulation in summary categories, different summary patterns were used. For example, Safaiean *et al.* (2017) used the UPS summary patterns, whilst Persson *et al.* (2006) identified compensatory articulation using a three-step process (yes/no scale → categorizing type of compensatory articulation as retracted oral/pharyngeal/glottal/active nasal fricative/other → recording frequency of error). Chapman *et al.* (2008) and Hutters *et al.* (2001) both recorded the frequency with which CSCs/compensatory articulations occurred. The studies highlight that at age 3 there is a consensus on the need to report CSCs, even within the context of a developing sound system, albeit using different summary categories.

The second principal method was percentage consonants correct (PCC) (Shriberg and Kwiatkowski 1982). PCC was also used in its adjusted form (PCC-A) (table 3) in which age-appropriate speech distortions are classed as correct productions (Shriberg *et al.* 1997). For example, Klintö *et al.* (2014a, 2014b, 2015, 2016) used PCC-A, with simplifications of phonemes used by more than 10% of the children scored as correct (Klintö *et al.* 2016: 151). Unlike the summary patterns that specifically focus on CSCs, PCC is a broader measure of all the speech errors including CSCs, developmental and other disordered speech patterns. PCC does not differentiate between articulatory or phonologically based errors. This 'combined approach' is recommended by Lohmander *et al.* (2017a) as articulation errors (arising from structural or functional abnormalities) may be 'phonologized' (Harding and Grunwell 1995) and become integrated into a child's sound system alongside or replacing developmental phonological patterns. Summary patterns similarly provide phonological information, for example, *backing* is an example of a common phonological process arising from retracted articulation (Chapman 2003, Willadsen 2012). UK cleft units did not appear to use PCC in the assessment of 3-year-olds (Wren 2013).

Other measures of consonant production included consonant/phoneme inventory which was used in 85.7% of UK cleft units at age 3 (Wren 2013). This also appeared in several other studies including the SVANTE (Lohmander *et al.* 2017a, Klintö *et al.* 2014a, Morris and Ozanne 2003, Chapman *et al.* 2008, Chapman 2004, Konst *et al.* 2003, Hattee *et al.* 2001). The criteria for inclusion in the inventory varied across the studies. For example (Hattee *et al.* 2001) rated the presence of consonants, Morris and Ozanne (2003) required phonemes to be heard twice to be included, and Lohmander *et al.* (2017a) reported correct productions in more than 50% of the targets.

Several studies reported on both cleft and developmental phonological processes (Willadsen *et al.* 2018, Chacon *et al.* 2017, Klintö *et al.* 2014a, 2014b, 2016, Willadsen 2012, Konst *et al.* 2003, Morris and Ozanne 2003, Hutters *et al.* 2001). This provides a broader perspective of an individual's phonological development

Table 3. Summary of the parameters of speech assessed: papers that used percentage consonants correct (P CC) were categorized as assessing both articulation and phonology

	Parameters of assessment							
			Resonance					
Author (date)	Articulation	Phonology	Hypernasality	Hyponasality	Nasal airflow errors (NAE)	Velopharyngeal function	Intelligibility	Voice
Frey *et al.* (2018)							√ Categorization	
Willadsen *et al.* (2018)	√ PPC adjusted for age: age-appropriate distortions were counted as correct, as were accompanying nasality or NAE if the production did not cross a phoneme boundary	√						
Chacon *et al.* (2017)	√ PCC PCC-R: in which sound distortions were also counted as correct Percentage vowels correct	√	√					
Larsson *et al.* (2017)	√ PCC adjusted for age: misarticulations of /s, ɕ/ were scored as correct at age 3 with reference to typically developing 3-year-olds. Audible nasal leakage or weak articulation was scored correctly PCC by articulatory manner PCC by articulatory placement	√			√ Nasal air leakage	√ Binary rating scale of yes/no to indicate competent velopharyngeal function		
Lohmander *et al.* (2017a)	√ PCC	√	√	√	√ Nasal air leakage	√ Three-point scale to describe velopharyngeal function for speech	√ Three-point ordinal scale	
Raud Westberg *et al.* (2017)	√ PCC adjusted for age	√						
Safaiean *et al.* (2017)	√ PCC	√	√			√ Orthographic transcription by naïve listeners	√	
Swanson *et al.* (2017)	√		√		√ Nasal emission/ nasal escape/nasal turbulence	√ Composite score		√ Hoarseness/ breathiness, volume/ tension

(Continued)

Table 3. Continued

Author (date)	Parameters of assessment							
			Resonance					
	Articulation	Phonology	Hypernasality	Hyponasality	Nasal airflow errors (NAE)	Velopharyngeal function	Intelligibility	Voice
Klintö *et al.* (2016)	√ PCC adjusted: substitutions and simplifications of phonemes used by more than 10% of children are scored correctly	√						
El Ezzi *et al.* (2015)	√		√	√	√ Audible nasal emission	Borel–Maisonny SIMPLIFIED score, which uses a four-point scale with descriptors	√ Not directly assessed but scalar point d) on Borel–Maisonny refers to 'no intelligibility'	√ No information provided as to method
Klintö *et al.* (2015) (the same procedure as Klintö 2014a)	√ PCC adjusted: substitutions and simplifications of phonemes used by more than 10% of children are scored correctly	√						
Klintö *et al.* (2014a)	√ PCC adjusted: substitutions and simplifications of phonemes used by more than 10% of children are scored correctly	√						
Klintö *et al.* (2014b)	√ PCC adjusted: substitutions and simplifications of phonemes used by more than 10% of children are scored correctly	√	√	√				
Wren (2013)	Consonant inventory: 85.71% CSCs: 78.57%	50%	Resonance (hyper/hypo): 92.85%	Nasal emission: 85.71% Nasal turbulence: 78.57%		42.85%	78.57%	
Willadsen (2012)	√ PPC adjusted for age: age-appropriate distortions were counted as correct, as were accompanying evidence of nasality or NAE if the production did not cross a phoneme boundary	√						

(Continued)

Table 3. Continued

Author (date)	Parameters of assessment							
			Resonance					
	Articulation	Phonology	Hypernasality	Hyponasality	Nasal airflow errors (NAE)	Velopharyngeal function	Intelligibility	Voice
Willadsen and Poulsen (2012)	√ PPC adjusted for age: age-appropriate distortions were counted as correct, as were accompanying evidence of nasality or NAE if the production did not cross a phoneme boundary	√				√ Orthographic transcription by naïve listeners		
Dayashankara *et al.* (2011)	√		√			√ Referred to as clinical diagnosis by surgeon and SLT	√ Categorization	
Hamming *et al.* (2009)	√		√	√		√ Method not stated		√ (i) Hoarseness and other dysphonias rated as inconsistent/consistent (ii) Consistent dysphonias rated as mild/moderate/severe
Chapman *et al.* (2008)	√ Total consonants correct PCC by articulatory manner	√	√					
Gugsch *et al.* (2008)	√ vowels							√ Changes in the fundamental frequency of vowel production after surgery
Lohmander and Persson (2008)	√ PCC PCC by articulatory manner PCC by articulatory placement	√	√		√ Nasal air leakage			√

(Continued)

Table 3. Continued

Author (date)	Parameters of assessment							
			Resonance					
	Articulation	Phonology	Hypernasality	Hyponasality	Nasal airflow errors (NAE)	Velopharyngeal function	Intelligibility	Voice
Hodge and Gotzke (2007)							√ Orthographic transcription by naïve listeners	
Frederickson *et al.* (2006)	√ PCC	√	√					
Lohmander *et al.* (2006)	√		√		√ Nasal air leakage	√ Composite measure of velopharyngeal insufficiency, formed by combining the two highest scores for hypernasality, nasal leakage and weak pressure consonants	√ Categorization	
Persson *et al.* (2006)	√		√	√	√ Audible nasal emission	√ Five-point scale to provide a global measure of velopharyngeal impairment		
Chapman (2004)	√		√	√				
Konst *et al.* (2003)	√	√			√ Nasal escape			
Morris and Ozanne (2003)	√	√						
	PCC: with consonant distortions classified as correct PCC by articulatory manner							

(Continued)

Table 3. Continued

Author (date)	Parameters of assessment							
			Resonance					
	Articulation	Phonology	Hypernasality	Hyponasality	Nasal airflow errors (NAE)	Velopharyngeal function	Intelligibility	Voice
Zanzi *et al.* (2002)	√				√ Nasal emission	√ Borel–Maisonny score, which uses a five-point scale with descriptors		
Hattee *et al.* (2001)	√							
Hutters *et al.* (2001)	√	√						
Pulkkinen *et al.* (2001)	√		√		√ Nasal emission			
Gunther *et al.* (1998)			√		√ Nasal escape	√ Composite score		√ Hoarseness rated on a four-point scale
Lohmander-Agerskov (1998)	√		√	√	√ Nasal escape-velopharyngeal friction sounds		√ Categorization (also included voice)	√ Voice quality and intelligibility rated as normal, mildly distorted, severely distorted
Lohmander-Agerskov *et al.* (1998)	√		√	√	√ Nasal escape-velopharyngeal friction sounds		√ Categorization	

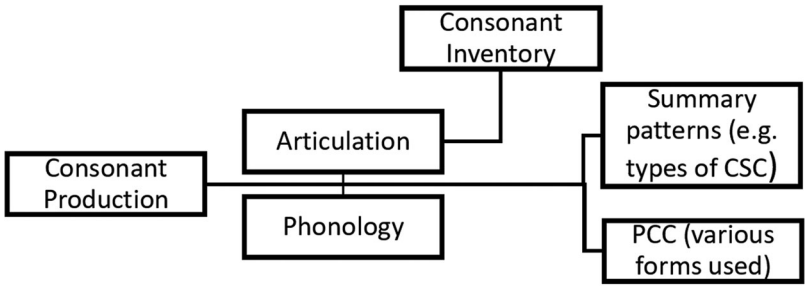

Figure 3. Methods used to assess consonant production.

from both a cleft and developmental perspective. In contrast, whilst 78.57% of UK cleft units assess CSCs at age 3, only 50% were reported to assess phonology (Wren 2013). Whilst there may be some overlap between measures of CSCs and phonology, this indicates that a comprehensive phonological assessment may not be considered a priority for assessment in UK cleft units at age 3.

The sources reviewed highlight the importance, at 3 years of age, of assessing consonant production both from a cleft and developmental perspective. The methods used in the studies to assess consonant production demonstrate that both PCC and summary patterns can provide information about articulation and phonology.

Resonance

Both judgements of hypernasality and hyponasality are made in the CAPS-A, CAPS-A-AM, UPS, PWSS and SVANTE outcome measures, indicating that these are two parameters core to the assessment of speech in the CP±L population. Of those studies assessing resonance, hypernasality was reported to be specifically assessed in 84.2% of the studies, as shown in table 3. Ordinal scales were most frequently used, although scales and descriptors varied. For example, Lohmander *et al.* (2006) used a five-point scale with descriptors, Chapman *et al.* (2008) used a four-point scale, and Pulkkinen *et al.* (2001) used a binary scale rating the presence/absence of hypernasality. Whereas hypernasality was sometimes assessed in the absence of hyponasality, hyponasality was always reported alongside hypernasality (table 3). Wren (2013) reported that hypernasality and hyponasality are assessed in 92.85% of UK cleft units at age 3.

Although, those studies assessing resonance predominately assessed hypernasality, there was no consensus as to the type or length of scale used.

Nasal airflow errors (NAE)

NAE is a generic term to describe air escaping inappropriately through the nose during the production of oral pressure consonants (Sweeney 2011: 200). NAE featured in several of the studies reviewed and it is therefore also considered core to the assessment of cleft speech at this age. Wren (2013) separated out the parameters of nasal emission and turbulence. In contrast, other studies only reported on audible nasal emission (although this term was possibly used to refer to all NAE, that is, both nasal emission and nasal turbulence) or used a single measure to encapsulate all types of nasal airflow, as per the CAPS-A-AM and SVANTE protocols. As with measures of resonance, a variety of scales were used, for example, Konst *et al.* (2003) measured the percentage of target consonants realized with nasal escape, whereas Lohmander and Persson (2008) used a five-point scale to measure the frequency with which nasal air leakage occurred.

Velopharyngeal function

An overall measure of velopharyngeal function based on perceptual speech assessment featured in some studies, as shown in table 3. This parameter was assessed in addition to those associated with velopharyngeal impairment, that is, hypernasality and NAE. Methods of assessment varied significantly, with some studies using a composite score (Swanson *et al.* 2017, Lohmander *et al.* 2006, Gunther *et al.* 1998), whilst others used rating scales (Zanzi *et al.* 2002, Persson *et al.* 2006, El Ezzi *et al.* 2015, Lohmander *et al.* 2017a, Larsson *et al.* 2017). Dayashankara *et al.* (2011) referred to the use of a clinical diagnosis, and Hamming *et al.* (2009) did not report the methods used. Wren (2013) did not record if an overall measure of velopharyngeal function was used in UK cleft units at age 3.

Intelligibility

Whitehill *et al.* (2011) stated that a key objective of all cleft teams is for their patient's speech to be understood and that intelligibility is a measure of how successfully cleft teams have achieved this (Whitehill *et al.* 2011: 293). Whilst the SVANTE protocol assesses

intelligibility, there are well-reported challenges in both defining and measuring intelligibility (Whitehill 2002). Despite these challenges, intelligibility was assessed in 11 studies, using methods of orthographic transcription, or categories with descriptors (table 3). From a clinical perspective, fewer than half of the UK cleft units (42.85%) reported that intelligibility was assessed at age 3 (Wren 2013).

Voice

Only 20% of sources reported on the parameter of voice; this is summarized in table 3. In contrast, voice was reported on in 78.57% of UK cleft units Wren (2013). One explanation may be that UK SLTs routinely screen voice (as a parameter of assessment on the CAPS-A, and in clinical assessment using the GOS.SP.ASS) and apply these or similar categories to the assessment of voice at age 3. As such there may be a different culture in the assessment of voice in the UK at age 3 in comparison with other countries.

What methods and rating scales are used to assess the parameters of speech identified?

Methods of assessment

Phonetic transcription, as previously stated, underpinned measures of consonant production. This was supplemented in the studies of Chacon *et al.* (2017), Chapman *et al.* (2008), Chapman (2004) and Morris and Ozanne (2003) by computer software to aid the analysis of articulation and phonology. Computer-based analysis was also used by Gugsch *et al.* (2008) in the evaluation of the voice-specific measures of formants and fundamental frequency. Orthographic transcription using naïve listeners was used in studies assessing intelligibility, as shown in table 3.

Categorical rating scales were commonly used (Frey *et al.* 2018, Chacon *et al.* 2017, Lohmander *et al.* 2006, 2017a, Swanson *et al.* 2017, El Ezzi *et al.* 2015, Klintö *et al.* 2014b, Dayashankara *et al.* 2011, Hamming *et al.* 2009, Chapman *et al.* 2008, Lohmander and Persson 2008, Frederickson *et al.* 2006, Persson *et al.* 2006, Chapman 2004, Konst *et al.* 2003, Zanzi *et al.* 2002, Gunther *et al.* 1998, Lohmander-Agerskov 1998, Lohmander-Agerskov *et al.* 1998); however, binary scales also featured (Larsson *et al.* 2017, Hamming *et al.* 2009, Pulkkinen *et al.* 2001). Despite this, there was limited commonality in the categorical scales across the studies with a range of scales, number of scalar points and descriptors used. As an alternative to categorical scales, Hodge and Gotzke (2007) used a continuous scale to measure intelligibility, and Chapman *et al.* (2008) used direct magnitude estimation (DME) to measure articulation proficiency and hypernasality.

What types of speech samples are used in the assessment of 3-year-old children with CP±L?

The type of speech samples used is outlined in table 2. A total of 20% of the studies did not provide any information about this. Of the remaining studies, single-word naming was most frequently used, in some instances in combination with other speech samples. A total of eight sources (29%) used the picture naming test developed as part of the Scandcleft Trial, referred to as the restricted word list (table 2). This picture naming test has been developed in seven different languages to facilitate cross-linguistic speech comparisons (Cleft Palate International Speech Issues (CLISPI) n.d.) through the assessment of phonetically similar units of speech across language and context (Lohmander *et al.* 2009: 348).

Samples of spontaneous speech were also frequently used occurring in 37.14% of the sources, sometimes in addition to single-word naming. Wren (2013) reported that 64.28% of UK cleft units used a spontaneous speech sample in the assessment of 3-year-olds with CP±L. Sentence repetition was used by Chacon *et al.* (2017); participants repeated six sentences taken from the GOS.SP.ASS (Sell *et al.* 1999). The GOS.SP.ASS was frequently used across UK cleft units with 85.7% using this assessment at age 3 (Wren 2013). However, no information is provided as to the type of speech sample used, that is, if it was used in its most common form (using the accompanying sentences) or if short phrases or single words were used.

Discussion

This scoping review aimed to map assessment practices at age 3 in the CP±L population. It found that the evaluation of palatal structure and function through the assessment of resonance, NAE, overall measures of velopharyngeal function and CSCs was a central feature of assessment in many of the sources reviewed, even at this young age. Whilst this approach to assessment is in line with a medical model, it was used alongside a linguistic approach to assessment which was underpinned by phonetic transcription. Although many of the sources had the same assessment objective, that is, to assess palatal function for speech, the assessment methods varied across the studies. The majority of studies reported NAE in combination (e.g., both emission and turbulence in a single measure) including the SVANTE, which is designed to measure speech outcomes at age 3. In contrast, UK cleft units reported nasal emission and turbulence separately, this may be historical and

recorded as per the CAPS-A and the GOS.SP.ASS. There is a need for consensus in the UK as to whether the objective at age 3 is to assess the type or overall presence of NAE. To guide this decision-making, further research is needed to understand the implications of rating nasal emission and turbulence separately or in combination, on listener reliability.

This scoping review indicates that an overall measure of velopharyngeal function was assessed using varying methods. This included the use of validated and unvalidated rating scales and clinical opinions. There is an emphasis on overall measures in the reporting of speech outcomes in the CP±L population and this has been recommended by the International Consortium for Health Outcome Measurement (ICHOM) (2017) for use at older ages. The overall measure VPC-Rate (Lohmander *et al.* 2009) shows potential for use with this age group; it has been shown to be reliable and efficient (Lohmander *et al.* 2017b) and has featured in several studies reporting outcomes associated with the Scandcleft Trial, and used in a similar form in the SVANTE. In the UK, an overall score for resonance and NAE is used to measure 5-year speech outcomes against National Standards for Speech (Britton *et al.* 2014) and a velopharyngeal composite score using the CAPS-A has also been validated (Pereira *et al.* 2013). Further research should compare the reliability of hypernasality, NAE and overall velopharyngeal function at age 3 to support decisions regarding the selection of outcome measures at this age.

A linguistic approach underpinned by phonetic transcription allowed for both phonetic and phonological analysis of consonant production and the subsequent identification of cleft specific CSCs and calculations of PCC. The frequent reporting of consonant production using cleft summary patterns highlights the importance of this approach during the preschool years. Again, challenges in comparing speech outcomes across the studies arise from the use of different criteria and summary patterns (Sell 2005) and the need for consensus is relevant both at age 3 years and for other age groups. PCC-A, using a controlled speech sample, allows for cross-linguistic comparisons and takes into consideration sound distortions occurring as part of normal development, which is particularly appropriate for assessment at age 3. PCC-A may, therefore, offer a partial solution although it has the disadvantage of not capturing the qualitative nature of speech errors. In addition, to be meaningful, PCC-A needs to be used alongside normative or comparison data (longitudinal data or data to compare groups), which may account for why PCC-A has not been adopted in the UK assessments at age 3. Perhaps a solution to the need for normative/comparison data may be to compare PCC-A at ages 3 and 5 to monitor progress, or to use assessments with available normative data, that is, the DEAP (Dodd *et al.* 2002), although this has the disadvantage of not being specifically designed to assess cleft speech.

The assessment of speech from a developmental perspective has not been a core feature in the reporting of cleft speech outcomes (which may account for why the assessment of phonology only featured in 50% of UK cleft centres; Wren 2013). However, this scoping review highlights the use of both PCC-A and developmental phonological processes in speech assessments at age 3 demonstrating the use of a combined linguistic and developmental approach to consider delayed, typical and atypical speech production. The importance of a developmental approach is evidenced by studies in this review, as both Chacon *et al.* (2017) and Hutters *et al.* (2011) reported that children with CP±L present with more developmental phonological processes at 3 years of age than their non-cleft peers.

The assessment of intelligibility in the studies included in this review recognizes the need for a functional approach to speech assessment. Whilst not a cleft specific outcome measure, a functional approach to assessment at age 3 is important given that many children start attending pre-school education in the UK at this age and thus interact with a wider social group. Most studies in the review used ordinal scales to assess intelligibility despite evidence questioning their validity to measure this parameter (Whitehill *et al.* 2011). In the context of the International Classification of Functioning, Disability and Health: Children and Youth Version (ICF-CY; World Health Organization (WHO) 2007) McLeod *et al.* (2012: 649) report that intelligibility is influenced by both production factors (body functions) and contextual factors (environmental factors). Safaiean *et al.* (2017), Willadsen and Poulsen (2012) and Hodge and Gotzke (2007) used orthographic transcription by naïve listeners. This method addresses concerns regarding contextual factors and the validity of expert (cleft SLT) listeners rating intelligibility but would be impractical to employ in clinical practice. Indeed, The Intelligibility in Context Scale (ICS) (McLeod at al. 2012), which uses parent ratings of intelligibility, shows potential for use with the cleft population. The ICS is validated and recommended by ICHOM (2017) for use at ages 5 and 12 years, but has yet to be validated for use at age 3; the challenges and complexity of measuring intelligibility at this age persist.

In addition, to intelligibility, the reliability and validity of using ordinal/categorical scales to measure resonance and NAE has been challenged by growing evidence to suggest that ratio or category-ratio scales may be more valid and reliable measures (Yamashita *et al.* 2018, Baylis *et al.* 2015). However, in the studies in this review, only Hodge and Gotzke (2007) and Chapman *et al.* (2008) used non-categorical scales, using a continuous scale and DME, respectively, with good levels

of reliability reported in both studies. The impact of different rating scales on the validity and reliability of listener judgements has therefore yet to be fully explored in 3-year-old children with CP±L and further research is required to determine if findings at this age mirror that of other age groups.

Three types of speech samples were used in the studies: single-word production, spontaneous speech samples and, more infrequently, sentence repetition. The variety of assessments used to assess single words is striking and well exemplified by Wren (2013) which indicates there is no preferred single-word assessment to evaluate speech at age 3 in the UK. The use of supplementary assessment materials or unnamed picture naming assessments suggests existing assessments may not be wholly adequate for the comprehensive assessment of speech at age 3 in the CP±L population. The restricted word list (Lohmander *et al.* 2009), developed for cross-linguistic comparisons, would facilitate multi-centre research and comparisons of outcomes internationally. However, given its design to allow cross-linguistic comparisons it does not assess the full range of oral pressure consonants in English and there is a need to expand upon this assessment to assess speech more comprehensively (as per the SVANTE). Nonetheless, this review highlights its potential use in speech assessments at age 3.

Speech samples may need to be used in combination to comprehensively assess speech at age 3 when combining approaches to assessment, that is, using a medical model and linguistic, developmental and functional approaches. Most common in the studies are single-word naming samples, given that this is an age-appropriate task and there is evidence at age 5 that single-word naming samples enhance listener reliability in judgements of consonant production (Klintö *et al.* 2011). However, evidence that there may be variability in 'speech performance between single words and conversational speech' (Sweeney 2011: 206) indicates that this sample may not be sufficient in isolation, particularly when measuring the core parameters of hypernasality, NAE and consonant production. Although 37.14% of the studies used a spontaneous speech sample, Klintö *et al.* (2014b), Persson *et al.* (2006), and Lohmander and Persson (2008) favoured single words to assess resonance and NAE. Klintö *et al.* (2014b) used only single-words reporting that a 'representative and standardized speech sample with connected speech' (277) could not be achieved, thus highlighting the challenges of spontaneous speech samples at this age. An alternative is sentence or phrase repetition. Wren (2013) found that 85.7% of UK cleft units used the GOS.SP.ASS and the accompanying speech sample (Sell *et al.* 1999) in addition to single-word assessments. The familiarity UK cleft SLTs have with the GOS.SP.ASS may help them to support 3-year-olds in completing the GOS.SP.ASS sentences, sometimes as short phrases, and may facilitate reliable listener judgements. The SVANTE also includes both single word and sentence-level speech samples.

Limitations

The parameters of speech identified in the study are those established as core to the assessment of cleft speech and do not constitute novel information. The number of sources ($n = 8/35$; 22.9%) included in this review that report on outcomes associated with the Scandcleft Trial is a testament to the success of this research group in generating new knowledge. However, it is important to recognize the impact this has on the overall results of this scoping review, introducing an element of bias into the results given that the same assessment principles and processes were used across this subset of studies. The inclusion of sources in languages other than English would have provided a more diverse overview of assessment practices at age 3 in other parts of the world and could potentially have informed UK practice.

Conclusions

Whilst there are core parameters fundamental to the assessment of cleft speech, this scoping review also highlights the unique assessment requirements of 3-year-old children, particularly with reference to the selection of the speech sample and the importance of using a developmental approach to assessment. The need to assess functional outcomes at this age, that is, intelligibility, in addition to cleft specific outcomes, that is, resonance and NAE, is highlighted.

Unlike age 5, speech outcomes at age 3 in the CP±L population have not been a central focus in the UK or internationally; this is perhaps reflected in the variety of assessment methods noted in this review. However, more recent studies relating to the Scandcleft study highlight the potential of the restricted word list (Lohmander *et al.* 2009) for use in assessments at age 3, particularly as this allows for cross-linguistic comparisons. This scoping review highlights the need for a consolidated approach to assessing CP±L speech at age 3; however, further research is needed to provide a foundation for assessments at this important age in order to develop assessment protocols. This should consider the extent to which 3-year-old children with CP±L can complete different speech samples and the impact this has on the parameters of speech which can be assessed and the validity and reliability of listener judgements of those parameters using different measures.

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References

Arksey, H. and O'malley, L., 2005, Scoping studies: Towards a methodological framework. *International Journal of Social Research Methodology*, **8**(**1**), 19–32.

Armstrong, S. and Ainley, M., 1992, *South Tyneside Assessment of Phonology (STAP)* (Notuhumberland: Stass Publications).

Baylis, A., Chapman, K. and Whitehill, T. L., 2015, Validity and reliability of visual analogue scaling for assessment of hypernasality and audible nasal emission in children with repaired cleft palate. *Cleft Palate-Craniofacial Journal*, **52**(**6**), 660–670.

Boucher, J. and Lewis, V., 1997, *Preschool Language Scale—3 (UK)* (London: Psychological Corporation).

Britton, L., Albery, L., Bowden, M., Harding-Bell, A., Phippen, G. and Sell, D., 2014, A cross-sectional cohort study of speech in five-year-olds with cleft palate ± lip to support development of national audit standards: benchmarking speech standards in the United Kingdom. *Cleft Palate-Craniofacial Journal*, **51**(**4**), 431–451.

Cavalheiro, M. G., Lamônica, D. A. C., De Vasconsellos H., Simone R. and Maximino, L. P., 2019, Child development skills and language in toddlers with cleft lip and palate. *International Journal of Pediatric otorhinolaryngology*, **116**, 18–21.

Chacon, A., Parkin, M., Broome, K. and Purcell, A., 2017, Australian children with cleft palate achieve age-appropriate speech by 5 years-of-age. *International Journal of Pediatric Otorhinolaryngology*, **103**, 93–102.

Chapman, K. L., 2004, Is presurgery and early postsurgery performance related to speech and language outcomes at 3 years of age for children with cleft palate? *Clinical Linguistics and Phonetics*, **18**(**4–5**), 235–257.

Chapman, K. L., Baylis, A., Trost-Cardamone, J., Cordero, K. N., Dixon, A., Dobbelsteyn, C., Thurmes, A., Wilson, K., Harding-Bell, A. and Sweeney, T., 2016, The Americleft Speech Project: a training and reliability study. *Cleft Palate-Craniofacial Journal*, **53**(**1**), 93–108.

Chapman, K. L., Hardin-Jones, M. A., Goldstein, J. A., Halter, K. A., Havlik, R. J. and Schulte, J., 2008, Timing of palatal surgery and speech outcome. *Cleft Palate-Craniofacial Journal*, **45**(**3**), 297–308.

Chapman, K. L., Hardin-Jones, M. and Halter, K. A., 2003, The relationship between early speech and later speech and language performance for children with cleft lip and palate. *Clinical Linguistics and Phonetics*, **17**(**3**), 173–197.

Chapman, K. L. and Willadsen, E., 2011, The development of speech in children with cleft palate. In S. Howard and A. Lohmander (eds), *Cleft Palate Speech: Assessment and Intervention* (Chichester: Wiley-Blackwell), pp. 23–42.

Clear Resources, 2006, *CLEAR Phonology Screening Assessment* (Spilsby: CLEAR Resources).

Cleft Palate International Speech Issues (CLISPI), n.d., *Speech Material to be Collected at Different Ages* (available at: https://www.clispi.org/en/node/17) (accessed on 11 August 2018).

Cleft Registry and Audit Network (CRANE), 2019, *Annual Report on Cleft Lip and/or Palate 2019* (available at: https://crane-database.org.uk/?!.iD=3l) (accessed on 1 October 2019).

Dayashankara, R. J., Singh, S., Suma, G. N., Gokkula, K., Kumar, V. and Sursh, B. S., 2011, Comparative study of 2 palatoplasty techniques to assess speech and fistula in primary cleft palate patients. *Journal of Dentistry for Children*, **78**(**1**), 13–18.

Dodd, B., Zhu, H., Crosbie, S., Holm, A. and Ozanne, A., 2002, *Diagnostic Evaluation of Articulation and Phonology (DEAP)* (London: Psychology Corporation).

Dosman, C. F., Andrews, D. and Goulden, K. J., 2012, Evidence-based milestone ages as a framework for developmental surveillance. *Paediatrics and Child Health*, **17**(**10**), 561–568.

Dudas, J. R., Deleyiannis, F. W., Ford, M. D., Jiang, S. and Losee, J. E., 2006, Diagnosis and treatment of velopharyngeal insufficiency: Clinical utility of speech evaluation and videofluoroscopy. *Annals of Plastic Surgery*, **56**(**5**), 511–517.

El Ezzi, O., Herzog, G., Broome, M., Trichet-Zbinden, C., Hohlfeld, J., Cherpillod, J. and De Buys Roessingh, A. S., 2015, Grommets and speech at three and six years in children born with total cleft or cleft palate. *International Journal of Pediatric Otorhinolaryngology*, **79**(**12**), 2243–2247.

Frederickson, M. S., Chapman, K. L. and Hardin-Jones, M., 2006, Conversational skills of children with cleft lip and palate: a replication and extension. *Cleft Palate-Craniofacial Journal*, **43**(**2**), 179–188.

Frey, J. R., Kaiser, A. P. and Scherer, N. J., 2018, The influences of child intelligibility and rate on caregiver responses to toddlers with and without cleft palate. *Cleft Palate-Craniofacial Journal*, **55**(**2**), 276–286.

Goldman, R. and Fristoe, M., 1986, *Goldman–Fristoe Test of Articulation* (Circle Pines Minnesota: American Guidance Service).

Grunwell, P. and Harding, A., 1995, *PACS TOYS* (Windsor: NFER-Nelson).

Grunwell, P. and Sell, D., 2001, Speech and cleft palate/velopharyngeal anomalies. In A. C. H. Watson, D. A. Sell and P. Grunwell (eds), *Management of Cleft Lip and Palate* (London: Whurr), pp. 68–86.

Gugsch, C., Dannhauer, K. and Fuchs, M., 2008, Evaluation of the progress of therapy in patients with cleft lip, jaw and palate, using voice analysis—a pilot study. *Journal of Orofacial Orthopedics*, **69**(**4**), 257–267.

Gunther, E., Wisser, J. R., Cohen, M. A. and Brown, A. S., 1998, Palatoplasty: Furlow's double reversing Z-plasty versus intravelar veloplasty. *Cleft Palate-Craniofacial Journal*, **35**(**6**), 546–549.

Hamming, K. K., Finkelstein, M. and Sidman, J. D., 2009, Hoarseness in children with cleft palate. *Otolaryngology—Head and Neck Surgery*, **140**(**6**), 902–906.

Harding, A. and Grunwell, P., 1995, Active versus passive cleft-type speech characteristics: implications for surgery and therapy. *International Journal of Language and Communication Disorders*, **30**(**S1**), 242–247.

Hattee, C., Farrow, K., Harland, K., Sommerlad, B. and Walsh, M., 2001, Are we ready to predict speech development from babble in cleft lip and palate children? *International Journal of Language and Communication Disorders*, **36**(**S1**), 115–120.

HENNINGSSON, G., KUEHN, D. P., SELL, D., SWEENEY, T., TROST-CARDAMONE, J. E. and WHITEHILL, T. L., 2008, Universal parameters for reporting speech outcomes in individuals with cleft palate. *Cleft Palate-Craniofacial Journal*, **45**(**1**), 1–17.

HEYDARI, S., TORABI NEZHAD, F., AGHA RASOULI, Z. and HOSEYNI, F., 2011, Development of speech intelligibility measurement test for 3 to 5 years old normal children. *Bimonthly Audiology-Tehran University of Medical Sciences*, **20**(**1**), 47–53.

HODGE, M. and GOTZKE, C. L., 2007, Preliminary results of an intelligibility measure for English-speaking children with cleft palate. *Cleft Palate-Craniofacial Journal*, **44**(**2**), 163–174.

HOWARD, S., 2011, Phonetic transcription for speech related to cleft palate. In S. Howard S. and A. Lohmander (eds), *Cleft Palate Speech: Assessment and Intervention* (Chichester: Wiley-Blackwell), pp. 127–144.

HUTTERS, B., BAU, A. and BRØNDSTED, K., 2001, A longitudinal group study of speech development in Danish children born with and without cleft lip and palate. *International Journal of Language and Communication Disorders*, **36**(**4**), 447–470.

HUTTERS, B. and BRØNDSTED, K., 1987, Strategies in cleft palate speech—with special reference to Danish. *Cleft Palate Journal*, **24**(**2**), 126–136.

INTERNATIONAL CONSORTIUM FOR HEALTH OUTCOME MEASUREMENT (ICHOM), 2017, *Cleft Lip and Palate Data Collection version 3.0.6*. (available: http://www.ichom.org/medical-conditions/cleft-lip-palate/) (accessed on 10 June 2018).

JOHN, A., SELL, D., SWEENEY, T., HARDING-BELL, A. and WILLIAMS, A., 2006, The cleft audit protocol for speech—augmented: A validated and reliable measure for auditing cleft speech. *Cleft Palate-Craniofacial Journal*, **43**(**3**), 272–288.

KHALIL, H., PETERS, M., GODFREY, C. M., MCINERNEY, P., SOARES, C. B. and PARKER, D., 2016, An evidence-based approach to scoping reviews. *Worldviews on Evidence-Based Nursing*, **13**(**2**), 118–123.

KLINTÖ, K., OLSSON, M., FLYNN, T., SVENSSON, H. and LOHMANDER, A., 2014a, Phonology in Swedish-speaking 3-year-olds born with cleft lip and palate and the relationship with consonant production at 18 months. *International Journal of Language and Communication Disorders*, **49**(**2**), 240–254.

KLINTÖ, K., SALAMEH, E. and LOHMANDER, A., 2015, Verbal competence in narrative retelling in 5 year olds with unilateral cleft lip and palate. *International Journal of Language and Communication Disorders*, **50**(**1**), 119–128.

KLINTÖ, K., SALAMEH, E. and LOHMANDER, A., 2016, Phonology in Swedish-speaking 5-year-olds born with unilateral cleft lip and palate and the relationship with consonant production at 3 years of age. *International Journal of Speech–Language Pathology*, **18**(**2**), 147–156.

KLINTÖ, K., SALAMEH, E., SVENSSON, H. and LOHMANDER, A., 2011, The impact of speech material on speech judgement in children with and without cleft palate. *International Journal of Language and Communication Disorders*, **46**(**3**), 348–360.

KLINTÖ, K., SVENSSON, H., ELANDER, A. and LOHMANDER, A., 2014b, Speech and phonology in Swedish-speaking 3-year-olds with unilateral complete cleft lip and palate following different methods for primary palatal surgery. *Cleft Palate-Craniofacial Journal*, **51**(**3**), 274–282.

KONST, E. M., RIETVELD, T., PETERS, H. F. and PRAHL-ANDERSEN, B., 2003, Phonological development of toddlers with unilateral cleft lip and palate who were treated with and without infant orthopedics: a randomized clinical trial. *Cleft Palate-Craniofacial Journal*, **40**(**1**), 32–39.

LARSSON, A., SCHÖLIN, J., MARK, H., JÖNSSON, R. and PERSSON, C., 2017, Speech production in 3 year old internationally adopted children with unilateral cleft lip and palate. *International Journal of Language and Communication Disorders*, **52**(**5**), 626–636.

LOHMANDER, A., BORELL, E., HENNINGSSON, G., HAVSTAM, C., LUNDEBORG, I. and PERSSON, C., 2005, *SVANTE Svenskt artikulations- och nasalitets-test, manual* [In Swedish], 2nd ed. (Lund: Studentlitteratur).

LOHMANDER, A., FRIEDE, H., ELANDER, A., PERSSON, C. and LILJA, J., 2006, Speech development in patients with unilateral cleft lip and palate treated with different delays in closure of the hard palate after early velar repair: a longitudinal perspective. *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery*, **40**(**5**), 267–274.

LOHMANDER, A., LUNDEBORG, I. and PERSSON, C., 2017a, SVANTE—The Swedish Articulation and Nasality Test—Normative data and a minimum standard set for cross-linguistic comparison. *Clinical Linguistics and Phonetics*, **31**(**2**), 137–154.

LOHMANDER, A. and PERSSON, C., 2008, A longitudinal study of speech production in Swedish children with unilateral cleft lip and palate and two-stage palatal repair. *Cleft Palate-Craniofacial Journal*, **45**(**1**), 32–41.

LOHMANDER, A., PERSSON, C., WILLADSEN, E., LUNDEBORG, I., ALALUUSUA, S., AUKNER, R., BAU, A., BOERS, M., BOWDEN, M. and DAVIES, J., 2017b, Scandcleft randomised trials of primary surgery for unilateral cleft lip and palate: 4. Speech outcomes in 5-year-olds-velopharyngeal competency and hypernasality. *Journal of Plastic Surgery and Hand Surgery*, **51**(**1**), 27–37.

LOHMANDER, A., WILLADSEN, E., PERSSON, C., HENNINGSSON, G., BOWDEN, M. and HUTTERS, B., 2009, Methodology for speech assessment in the Scandcleft project—an international randomized clinical trial on palatal surgery: experiences from a pilot study. *Cleft Palate-Craniofacial Journal*, **46**(**4**), 347–362.

LOHMANDER-AGERSKOV, A., 1998, Speech outcome after cleft palate surgery with the Gothenburg regimen including delayed hard palate closure. *Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery*, **32**(**1**), 63–80.

LOHMANDER-AGERSKOV, A., SÖDERPALM, E., FRIEDE, H. and LILJA, J., 1998, A comparison of babbling and speech at pre-speech level, 3, and 5 years of age in children with cleft lip and palate treated with delayed hard palate closure. *Folia Phoniatrica et Logopaedica*, **50**(**6**), 320–334.

MCLEOD, S. and BAKER, E., 2017, *Children's Speech: An Evidence-Based Approach to Assessment and Intervention* (Boston, MA: Pearson).

MCLEOD, S., HARRISON, L. J. and MCCORMACK, J., 2012, The intelligibility in context scale: Validity and reliability of a subjective rating measure. *Journal of Speech, Language, and Hearing Research*, **55**(**2**), 648–656.

MCWILLIAMS, B. and PHILIPS, B. J., 1979, *Velopharyngeal Incompetence: Audio Seminars in Speech Pathology* (Philadelphia, PA: W.B. Saunders).

MORRIS, H. and OZANNE, A., 2003, Phonetic, phonological, and language skills of children with a cleft palate. *Cleft Palate-Craniofacial Journal*, **40**(**5**), 460–470.

MUNN, Z., PETERS, M. D., STERN, C., TUFANARU, C., MCARTHUR, A. and AROMATARIS, E., 2018, Systematic review or scoping review? guidance for authors when choosing between a systematic or scoping review approach. *BMC Medical Research Methodology*, **18**(**1**), 143.

PEREIRA, V. J., SELL, D. and TUOMAINEN, J., 2013, Effect of maxillary osteotomy on speech in cleft lip and palate: Perceptual outcomes of velopharyngeal function. *International*

Journal of Language and Communication Disorders, **48**(**6**), 640–650.

PERSSON, C., LOHMANDER, A. and ELANDER, A., 2006, Speech in children with an isolated cleft palate: a longitudinal perspective. *Cleft Palate-Craniofacial Journal*, **43**(**3**), 295–309.

PETERS, M. D., GODFREY, C. M., KHALIL, H., MCINERNEY, P., PARKER, D. and SOARES, C. B., 2015, Guidance for conducting systematic scoping reviews. *International Journal of Evidence-Based Healthcare*, **13**(**3**), 141–146.

PRATHANEE, B., LORWATANAPONGSA, P., ANANTAPONG, D. and BUAKANOK, N., 2011, Thai speech parameters for patients with cleft palate in a universal reporting system. *Asia Pacific Journal of Speech, Language and Hearing*, **14**(**1**), 31–49.

PULKKINEN, J., HAAPANEN, M., PAASO, M., LAITINEN, J. and RANTA, R., 2001, Velopharyngeal function from the age of three to eight years in cleft palate patients. *Folia Phoniatrica et Logopaedica*, **53**(**2**), 93–98.

RAUD WESTBERG, L., HÖGLUND SANTAMARTA, L., KARLSSON, J., NYBERG, J., NEOVIUS, E. and LOHMANDER, A., 2017, Speech outcome in young children born with unilateral cleft lip and palate treated with one- or two-stage palatal repair and the impact of early intervention. *Logopedics Phoniatrics Vocology*, **44**(**2**), 58–66.

RENFREW, C. E., 2011, *Action Picture Test* (Milton Keynes: Speechmark).

SAFAIEAN, A., JALILEVAND, N., EBRAHIMIPOUR, M., ASLESHIRIN, E. and HIRADFAR, M., 2017, Speech intelligibility after repair of cleft lip and palate. *Medical Journal of The Islamic Republic of Iran*, **31**(**1**), 500–504.

SELL, D., 2005, Issues in perceptual speech analysis in cleft palate and related disorders: a review. *International Journal of Language and Communication Disorders*, **40**(**2**), 103–121.

SELL, D., HARDING, A. and GRUNWELL, P., 1999, GOS.SP.ASS'98: an assessment for speech disorders associated with cleft palate and/or velopharyngeal dysfunction (revised). *International Journal of Language and Communication Disorders*, **34**(**1**), 17–33.

SELL, D., JOHN, A., HARDING-BELL, A., SWEENEY, T., HEGARTY, F. and FREEMAN, J., 2009, Cleft Audit Protocol for Speech (CAPS-A): a comprehensive training package for speech analysis. *International Journal of Language and Communication Disorders*, **44**(**4**), 529–548.

SELL, D. and PEREIRA, V., 2015, Speech: perceptual and functional outcomes. In J. Losee. and R. E. Kirschner (eds), *Comprehensive Cleft Care* (London: McGraw Hill), pp. 689–714.

SHRIBERG, L. D. and KWIATKOWSKI, J., 1982, Phonological disorders III: A procedure for assessing severity of involvement. *Journal of Speech and Hearing Disorders*, **47**(**3**), 256–270.

SHRIBERG, L. D., AUSTIN, D., LEWIS, B. A., MCSWEENY, J. L. and WILSON, D. L., 1997, The percentage of consonants correct (PCC) metric: Extensions and reliability data. *Journal of Speech, Language, and Hearing Research*, **40**(**4**), 708–722.

STEVENS, N. and ISLES, D., 2011, *Phonological Screening Assessment (PSA)* (London: Taylor & Francis).

SWANSON, J. W., MITCHELL, B. T., COHEN, M., SOLOT, C., JACKSON, O., LOW, D., BARTLETT, S. P. and TAYLOR, J. A., 2017, The effect of Furlow palatoplasty timing on speech outcomes in submucous cleft palate. *Annals of Plastic Surgery*, **79**(**2**), 156–161.

SWEENEY, T., 2011, Nasality—assessment and intervention. In S. Howard and A. Lohmander (eds), *Cleft Palate Speech: Assessment and Intervention* (Chichester: Wiley–Blackwell), pp. 199–220.

TILLMAN, K. K., HAKELIUS, M., HÖIJER, J., RAMKLINT, M., EKSELIUS, L., NOWINSKI, D. and PAPADOPOULOS, F. C., 2018, Increased risk for neurodevelopmental disorders in children with orofacial clefts. *Journal of the American Academy of Child and Adolescent Psychiatry*, **57**(**11**), 876–883.

TRICCO, A. C., LILLIE, E., ZARIN, W., O'BRIEN, K., COLQUHOUN, H., KASTNER, M. and WILSON, K., 2016, A scoping review on the conduct and reporting of scoping reviews. *BMC Medical Research Methodology*, **16**(**1**), 15.

WHITEHILL, T. L., 2002, Assessing intelligibility in speakers with cleft palate: a critical review of the literature. *Cleft Palate-Craniofacial Journal*, **39**(**1**), 50–58.

WHITEHILL, T. L., GOTZKE, C. and HODGE, M., 2011, Speech intelligibility. In S. Howard and A. Lohmander (eds), *Cleft Palate Speech: Assessment and Intervention* (Chichester: Wiley-Blackwell), pp. 293–304.

WILLADSEN, E., 2012, Influence of timing of hard palate repair in a two-stage procedure on early speech development in Danish children with cleft palate. *Cleft Palate-Craniofacial Journal*, **49**(**5**), 574–595.

WILLADSEN, E., BOERS, M., SCHÖPS, A., KISLINGMØLLER, M., NIELSEN, J. B., JØRGENSEN, L. D., ANDERSEN, M., BOLUND, S. and ANDERSEN, H. S., 2018, Influence of timing of delayed hard palate closure on articulation skills in 3-year-old Danish children with unilateral cleft lip and palate. *International Journal of Language and Communication Disorders*, **53**(**1**), 130–143.

WILLADSEN, E., LOHMANDER, A., PERSSON, C., LUNDEBORG, I., ALALUUSUA, S., AUKNER, R., BAU, A., BOERS, M., BOWDEN, M. and DAVIES, J., 2017, Scandcleft randomised trials of primary surgery for unilateral cleft lip and palate: 5. Speech outcomes in 5-year-olds—consonant proficiency and errors. *Journal of Plastic Surgery and Hand Surgery*, **51**(**1**), 38–51.

WILLADSEN, E. and POULSEN, M., 2012, A restricted test of single-word intelligibility in 3-year-old children with and without cleft palate. *Cleft Palate-Craniofacial Journal*, **49**(**3**), 6–16.

WORLD HEALTH ORGANIZATION (WHO), 2007, International Classification of Functioning, Disability and Health. *Children and Youth Version* (Geneva: WHO).

WREN, Y., 2013, A survey of the assessment procedures at 24 and 36 months in UK cleft centres. *The Cleft Collective Speech and Language Study* (unpublished survey).

YAMASHITA, R. P., BORG, E., GRANQVIST, S. and LOHMANDER, A., 2018, Reliability of hypernasality rating: comparison of 3 different methods for perceptual assessment. *Cleft Palate-Craniofacial Journal*, **55**(**8**), 1–12.

ZANZI, M., CHERPILLOD, J. and HOHLFELD, J., 2002, Phonetic and ontological results after early palate closure in 18 consecutive children presenting with cleft lip and palate. *International Journal of Pediatric Otorhinolaryngology*, **66**(**2**), 131–137.

Appendix A: Papers included in the scoping review

Chacon *et al.* (2017)
Chapman (2004)
Chapman *et al.* (2008)
Dayashankara *et al.* (2011)
El Ezzi *et al.* (2015)
Frederickson *et al.* (2006)
Frey *et al.* (2018)
Gugsch *et al.* (2008)
Gunther *et al.* (1998)
Hamming *et al.* (2009)
Hattee *et al.* (2001)
Hodge and Gotzke (2007)
Hutters *et al.* (2001)
Klintö (2015)
Klintö *et al.* (2014a, 2014b, 2016)
Konst *et al.* (2003)
Larsson *et al.* (2017)
Lohmander and Persson (2008)
Lohmander *et al.* (2006, 2017a)
Lohmander-Agerskov (1998)
Lohmander-Agerskov *et al.* (1998)
Morris and Ozanne (2003)
Persson *et al.* (2006)
Pulkkinen *et al.* (2001)
Raud Westberg *et al.* (2017)
Safaiean *et al.* (2017)
Swanson *et al.* (2017)
Willadsen and Poulsen (2012)
Willadsen *et al.* (2012, 2018)
Wren (2013)
Zanzi *et al.* (2002)