Available online at www.sciencedirect.com

ScienceDirect

British Journal of Oral and Maxillofacial Surgery xxx (2023) xxx–xxx

BRITISH Journal of Oral and Maxillofacial Surgery

www.bjoms.com

Review

Velopharyngeal insufficiency, speech, and language impairment in craniofacial microsomia: a scoping review

Pauline A.E. Tio [a,*], Wietse Rooijers [a], Henriëtte H.W. de Gier [b], Henriëtte G. Poldermans [c], Maarten J. Koudstaal [a], Cornelia J.J.M. Caron [a]

[a] *The Dutch Craniofacial Centre, Department of Oral and Maxillofacial Surgery, Erasmus University Medical Centre, Sophia's Children's Hospital Rotterdam, Rotterdam, The Netherlands*

[b] *Department of Otorhinolaryngology, Erasmus University Medical Centre, Sophia's Children's Hospital Rotterdam, Rotterdam, The Netherlands*

[c] *Speech and Language Centre, Erasmus University Medical Centre, Sophia's Children's Hospital Rotterdam, Rotterdam, The Netherlands*

Received 11 July 2023; accepted in revised form 22 September 2023

Abstract

This review provides a comprehensive overview of the literature on velopharyngeal insufficiency, associated anomalies, and speech/language impairment in patients with craniofacial microsomia (CFM). A systematic search of the literature was conducted to identify records on VPI and speech impairment in CFM from their inception until September 2022 within the databases Embase, PubMed, MEDLINE, Ovid, CINAHL EBSCO, Web of Science, Cochrane, and Google Scholar. Seventeen articles were included, analysing 1,253 patients. Velopharyngeal insufficiency results in hypernasality can lead to speech impairment. The reported prevalence of both velopharyngeal insufficiency and hypernasality ranged between 12.5% and 55%, while the reported prevalence of speech impairment in patients with CFM varied between 35.4% and 74%. Language problems were reported in 37% to 50% of patients. Speech therapy was documented in 45.5% to 59.6% of patients, while surgical treatment for velopharyngeal insufficiency consisted of pharyngeal flap surgery or pharyngoplasty and was reported in 31.6% to 100%. Cleft lip and/or palate was reported in 10% to 100% of patients with CFM; these patients were found to have worse speech results than those without cleft lip and/or palate. No consensus was found on patient characteristics associated with an increased risk of velopharyngeal insufficiency and speech/language impairment. Although velopharyngeal insufficiency is a less commonly reported characteristic of CFM than other malformations, it can cause speech impairment, which may contribute to delayed language development in patients with CFM. Therefore, timely recognition and treatment of speech impairment is essential.

Keywords: Craniofacial microsomia; Velopharyngeal insufficiency; Speech disorders; Language disorders; Cleft palate

Introduction

Craniofacial microsomia (CFM) is a congenital malformation caused by the underdevelopment of the structures that arise from the first and second pharyngeal arches, which results in facial asymmetry.[1–5] With an incidence of approximately 1:3,500 to 1:5,600 births, CFM is considered to be the second most common congenital craniofacial malformation.[4] Structures most often affected include the mandible, ear, facial nerve, orbit, and soft tissues.[1–5] The most characteristic features include underdevelopment of the lower jaw and malformations of the outer ear.[6] The phenotype of patients with CFM is heterogenous and extracraniofacial malformations may be present, including malformations of the central nervous system, vertebrae, cardiorespiratory system, urogenital system, and limbs.[7] Furthermore, patients with CFM may suffer from functional impairments including feeding difficulties, hearing impairment, and obstructive sleep apnoea.[8–10]

In addition to the well-known features of CFM, a less commonly reported characteristic is velopharyngeal insufficiency (VPI).[11–13] VPI is a condition that involves incomplete closure of the soft palate, which results in the escape of nasal air during speech.[14–16] There is no consensus

* Corresponding author at: Department of Oral and Maxillofacial Surgery, Erasmus University Medical Center, Sophia's Children's Hospital Rotterdam, Doctor Molewaterplein 40, 3015 GD Rotterdam, The Netherlands.

E-mail address: p.tio@erasmusmc.nl (P.A.E. Tio).

https://doi.org/10.1016/j.bjoms.2023.09.008

in the literature on the aetiology and prevalence of VPI in CFM patients. It is proposed that the underdevelopment of the tensor veli palatini muscle, a derivative of the first branchial arch, and the levator veli palatini muscle, a derivative of the second branchial arch, causes VPI in CFM patients.[2] The trigeminal nerve, which also originates from the first branchial arch, is essential for motor innervation of the velum, and thus plays a critical role in velopharyngeal closure.[17] Therefore as an additional aetiology, unilateral palatal paralysis has been suggested as a cause of VPI in CFM.[11–13]

Preferably, the diagnosis of VPI is based on speech assessment, intraoral examination, and nasopharyngoscopy.[15,16] Common characteristics of VPI are hypernasality, nasal emission, and misarticulations, which result in speech distortions.[16] As a result, VPI is known to cause impaired speech, articulation errors, and decreased intelligibility, which are frequently described complications in CFM patients.[13,16,18–22]

Hearing impairment is a common problem in CFM patients. In the general population, unilateral hearing loss is associated with decreased outcomes in speech and language assessments.[23,24] In patients with CFM, delayed language skills and lower scores on receptive and expressive language measures are found compared to controls.[25] Moreover, a decreased intelligibility and disordered articulation were found in CFM patients compared to controls.[20] In addition to impaired hearing, VPI is also known to result in speech problems, which may contribute to the decreased intelligibility and articulation in these patients.[18] Therefore, early recognition of VPI is important for timely intervention and to prevent secondary developmental disorders.

Due to the scarcity of studies specifically addressing VPI in patients with CFM and the considerable heterogeneity in research pertaining to speech impairment in these patients, a scoping review was deemed the most appropriate design for this study. By conducting a comprehensive analysis of the available literature, this review aims to contribute to a better understanding of the current state of research on VPI and speech impairment in patients with CFM and to identify research gaps. It should therefore serve as a foundation for further investigations and inform future research directions on the topic of speech-related challenges faced by patients with CFM.

Methods

Search strategy

This review was guided by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses extension for scoping reviews (PRISMA-ScR) statement. A systematic search of the literature was conducted to identify records on VPI and speech impairment in CFM. The search was conducted within the databases Embase, PubMed, MEDLINE, Ovid, CINAHL EBSCO, Web of Science, Cochrane, and Google Scholar. Databases were searched from their inception until September 2022. Appendix A, presents the full search string of each database. Appendix B consists of a list of synonyms for CFM used in the search. In addition, reference lists of the included records were manually searched to identify additional records relevant to this study.

Two researchers (PAET and WR) independently screened the titles and abstracts of all records for relevance. Full texts of records without an abstract were reviewed for relevance. After the screening of titles and abstract, all relevant records underwent a full text review. Only original human studies concerning VPI and/or speech/language in CFM patients were included. Studies in other languages than English or Dutch were excluded. Case reports, conference abstracts, letters, notes, editorials, and studies including <10 CFM patients were excluded.

Data extraction

Data extracted from the included records consisted of: study design; number of included patients; prevalence of VPI; prevalence of speech/language impairment; treatments used for VPI and/or speech impairment; correlations between clinical manifestation of CFM and VPI and/or speech impairment. All records were graded on the quality of the evidence using the criteria of the Oxford Centre for Evidence-Based Medicine (CEBM).

Results

Study selection

A total of 6240 records was identified through the initial literature search. One additional record was identified through reference list searching. Following the screening based on title and abstract, 82 full-text records were screened for eligibility, of which 65 were excluded resulting in the inclusion of 17 records (Fig. 1).

Study characteristics

Of the 17 records included, 13 records were retrospective studies,[2,11–13,19,20,26–32] two were prospective studies,[33,34] and two were both retro- and prospective.[21,22] Among all records, three were case-control studies.[20,27,34] The characteristics of the included studies are described in Table 1. The average age of patients analysed ranged from 4.5 to 22.7 years. Patients with isolated microtia were included in six records.[20,27,28,32–34] A total of 1253 patients were analysed in this review. Three records were part of a multicentre longitudinal study from craniofacial centres across the United States and Canada.[20,27,34] Despite the fact that these records analysed the same patients, they were all included because they described different phases of the study. In total, 88 patients with CFM also had CL/P.

Prevalence of velopharyngeal insufficiency and related clinical characteristics

Nine records reported VPI in 0 to 55% of patients with CFM (Table 2).[2,11–13,19,21,29,31] Dellon et al[28] describe velopha-

Please cite this article as: Tio, Pauline A.E., Rooijers, Wietse, de Gier, Henriëtte H.W. et al. Velopharyngeal insufficiency, speech, and language impairment in craniofacial microsomia: a scoping review, *British Journal of Oral and Maxillofacial Surgery*, https://doi.org/10.1016/j.bjoms.2023.09.008

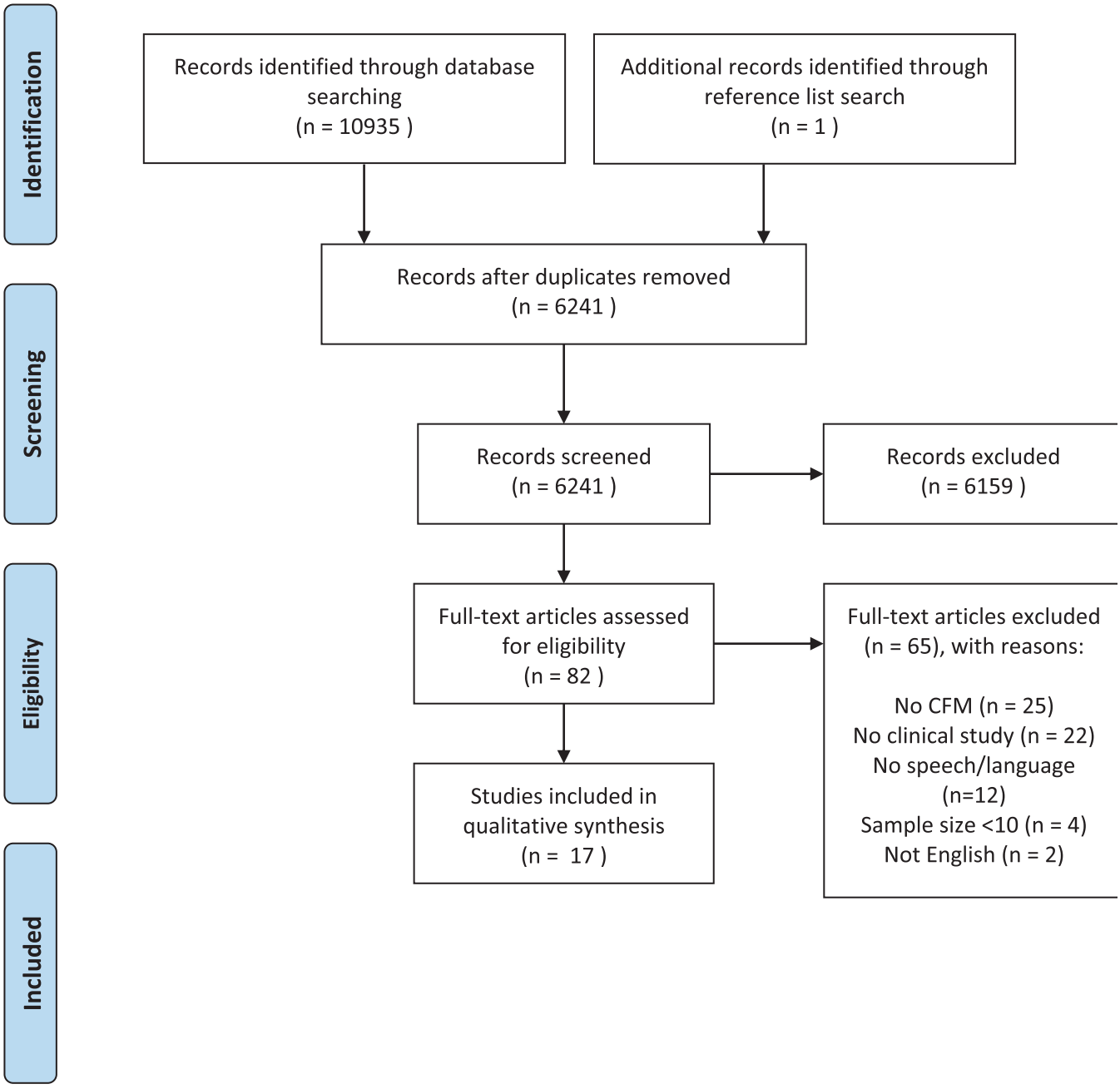

Fig. 1. PRISMA flow diagram.

ryngeal incompetence in 26.3% patients with microtia. Van Hövell Tot Westerflier et al[33] described VPI in 60% of patients with unilateral and 85% of patients with bilateral microtia, and hypernasality in 23% of patients with unilateral and 78% of patients with bilateral microtia. Seven records reported hypernasality in 12.5% to 55% of patients.[12,13,19–21,29,31] Two records reported unilateral hypodynamic palate (UHP) in 50% to 77.8% of patients.[11,13] Three records described a prevalence of palatal paralysis ranging from 36.8% to 50%.[2,13,28]

Cleft lip/palate was reported in five records, with a prevalence ranging between 10% to 100%.[12,20,27,29,30] In one record CL/P was part of the inclusion criteria.[29] Three records only reported the prevalence of cleft palate, ranging from 7.7% to 27%.[11,19,31] One record reported cleft lip/-palate/uvula in 27.8% of CFM patients.[22] Four records reported VPI or hypernasality in CFM patients without or with CL/P separately.[11,12,19,31] Funayama et al, Chan et al, and Shprintzen et al reported VPI in CFM patients without and with CL/P, in 14.6% versus 100%, 19.6% versus 100%, and 63% versus 33%, respectively.[11,12,31] D'Antonio et al[19] reported hypernasality in patients without and with CL/P, in 29% versus 70%, respectively. In addition, D'Antonio et al described perceptual evaluations showing symptoms of VPI in 20% among non-cleft patients and 26% of all patients evaluated.[19]

Please cite this article as: Tio, Pauline A.E., Rooijers, Wietse, de Gier, Henriëtte H.W. et al. Velopharyngeal insufficiency, speech, and language impairment in craniofacial microsomia: a scoping review, *British Journal of Oral and Maxillofacial Surgery*, https://doi.org/10.1016/j.bjoms.2023.09.008

Table 1
Study characteristics.

Year Author	CEBM level of evidence	Number of patients with CFM	Methodology	Inclusion criteria of the study
Chan, 2021[12]	3	68	Retrospective study	CFM
Chen, 2009[21]	3	167 + 65*	Retro- and prospective study	CFM
Cohen, 1995[26]	4	24	Retrospective study	OAVS
Collet, 2019[20]	3	107	Retrospective case-control	CFM
Collet, 2011[27]	3	136	Retrospective case-control	Facial asymmetry or HFM, including oculoauricular vertebral syndrome, GS, or microtia
D'Antonio, 1998[19]	3	41	Retrospective study	OAVS
Dellon, 1983[28]	4	19	Retrospective study	microtia
Dentino, 2016[29]	3	26	Retrospective study	HFM and cleft lip/palate
Funayama, 2007[11]	3	48+4**	Retrospective study	unilateral HFM or microtia without cleft palate; and 4 HFM patients with unilateral cleft palate
Goetze, 2017[30]	4	10	Retrospective study	OAVS criteria Strömland et al 2007
Grabb, 1965[2]	3	102	Retrospective study	First and second brachial arch syndrome
Johns, 2021[32]	3	169	Retrospective study	At least one of the following: microtia, mandibular hypoplasia and preauricular tag, mandibular hypoplasia and facial tag, mandibular hypoplasia and epibulbar dermoid, mandibular hypoplasia and a lateral oral cleft, preauricular tag and a lateral oral cleft, facial tag and epibulbar dermoid, or lateral oral cleft and an epibulbar dermoid.
Luce, 1977[13]	4	18	Retrospective study	HFM
Shprintzen, 1980[31]	4	22	Retrospective study	facio-auriculo-vertebral malformation complex
Strömland, 2007[22]	3	18	Retro- and prospective study	Abnormalities in at least two of the following areas: orocraniofacial, ocular, auricular, and vertebral.
van Hövell Tot Westerflier, 2019[33]	3	67	Prospective cohort	microtia
Wallace, 2018[34]	3	142	Prospective case-control	CFM

CEBM, Centre for Evidence-Based Medicine; CFM, craniofacial microsomia; HFM, hemifacial microsomia; OAVS, oculo-auriculo-vertebral syndrome.
* Chen et al 2009[21] analysed 167 patients in their retrospective study and 65 patients in their prospective study
** Funayama et al[11] 2007 analysed 48 HFM patient without cleft palate and four HFM patient with cleft palate.

Speech, language, and hearing impairment

Six records reported speech impairment in 35.4% to 74% of patients (Table 3).[13,19–22,27] Language problems were described in two records, with a prevalence ranging from 37% to 50%.[26,30] One record reported speech or language impairment in 42.2%.[32] Six records that reported on speech/language impairment also reported hearing impairment, with a prevalence ranging from 55% to 83%.[19,20,22,25,30,32]

Treatment of velopharyngeal insufficiency and speech impairment

Surgical treatment for VPI was reported in two records, ranging from 31.6% to 100% of patients (Table 4).[12,29] Surgical treatment consisted of pharyngoplasty and pharyngeal flap surgery. Treatment for speech impairment (23.7% to 59.6%) was described in four records.[12,20,32,34] Van Hövell Tot Westerflier et al[33] described treatment for speech impairment in 25% of patients with unilateral microtia and 75% with bilateral microtia.

Other associations in craniofacial microsomia with velopharyngeal insufficiency, speech, and language impairment

Three records reported that CFM patients with CL/P had a higher risk of VPI than non-cleft patients with CFM.[11,12,19] In addition, Chan et al[12] and Funayama et al[11] both suggested that CFM patients with CL/P have worse speech results than those without CL/P. Among CL/P patients in both records, only one patient had bilateral cleft lip and palate.[12]

Funayama et al[11] found that all CFM patients with VPI had UHP and that severity of mandibular hypoplasia and soft

Please cite this article as: Tio, Pauline A.E., Rooijers, Wietse, de Gier, Henriëtte H.W. et al. Velopharyngeal insufficiency, speech, and language impairment in craniofacial microsomia: a scoping review, *British Journal of Oral and Maxillofacial Surgery*, https://doi.org/10.1016/j.bjoms.2023.09.008

Table 2
Prevalence VPI and related clinical characteristics. Data are % (No./Total cohort.)

First author, year, and reference	VPI	Hypernasality/nasal air emission	Unilateral hypodynamic palate	Palatal paralysis	Cleft lip and/or palate
Chan, 2021[12]	27.9 (19/68)	27.9 (19/68) [a]			10.3 (7/68)
Chen, 2009[21]	12.5 (1/8)	12.5 (1/8) [b]			
Collet, 2019[20]		13.5 (13/96) [c]			15.5 (19/121)
Collet, 2011[27]					10% (14/136)
D'Antonio, 1998[19]	13 (3/23)	39 (16/41)			24.4 (10/41)[d]
Dellon, 1983[28]	26.3 (5/19) [e]			36.8 (7/19)	
Dentino, 2016[29]	9 (2/22)	18 (4/22)			100 (22/22) [f]
Funayama, 2007[11]	29.2 (7/24)		50 (24/48)		7.7 (4/52) [g]
Goetze, 2017[30]					10 (1/10)
Grabb, 1965[2]	0 (0/10)			50 (5/10)	
Luce, 1977[13]	33.3 (6/18)	33.3 (6/18)	77.8 (14/18)	50 (9/18)	
Shprintzen, 1980[31]	55 (12/22)	55 (12/22)			27 (6/22) [g]
Strömland, 2007[22]					27.8 (5/18) [h]
van Hövell Tot Westerflier, 2019[33]	60 (24/40) unilateral; 85 (23/27) bilateral	23 (9/40) unilateral; 78 (21/27) bilateral			

CFM, craniofacial microsomia; CL/P, cleft lip/palate; VPI, velopharyngeal insufficiency;
[a] VPI diagnosis was based on hypernasality/nasal air emission;
[b] only data of 8 patients;
[c] mild/moderate/severe;
[d] 6 overt palatal clefts and 4 submucous cleft palate;
[e] velopharyngeal incompetence;
[f] cleft lip with/without cleft palate;
[g] cleft palate;
[h] cleft lip/palate/uvula;

tissue deficiency were related to a significant increase of VPI/UHP. In addition, there was a significant association between the presence of VPI and macrostomia, and mental retardation. Luce et al[13] found that in patients with VPI soft tissue and skeletal deformities of the maxillary-malar complex were more severe. Chan et al[12] did not find an association between VPI and the severity of mandibular hypoplasia, soft tissue deficiencies or macrostomia.

Discussion

The aim of this review was to improve the understanding of VPI and speech-related challenges in patients with CFM. To achieve this goal, it was important to determine the prevalence of speech and language problems in CFM, with emphasis on VPI, identify patients at risk, and determine the effectiveness of treatment.

VPI was reported in 0 to 55% of patients, speech impairment in 35.4% to 74%, and language problems in 37% to 50%. The wide range of prevalence rates in VPI is primarily based on differences in diagnostic techniques and inclusion criteria. Several records used nasopharyngoscopy to diagnose VPI, whereas others based the diagnosis of VPI on the presence of hypernasality/nasal air emission. The wide range of prevalence rates in speech impairment can be attributed to the different criteria for speech impairment that were used; some records only reported articulation errors, whereas others reported speech impairment based on multiple diagnostic criteria (including hypernasality/hyponasality).

The wide range of prevalence rates of CL/P in CFM patients is based on a substantial difference in inclusion criteria. One record only included patients with CFM and CL/P, resulting in a 100% incidence of CL/P, whereas without this record the prevalence range is 10% to 27.8%. CL/P patients are at risk for speech and language delays.[35] CFM phenotypes with CL/P were found to have worse speech results than those without.[11,12] Among CFM patients with CL/P, there was a higher prevalence of VPI and hypernasality compared to the CFM patients without CL/P.[11,12,19] In contrast to these findings, only one record reported a decreased prevalence of VPI in CFM patients with CL/P compared to the noncleft CFM patients.[31] These findings suggest that in patients with both CFM and CL/P there is an increased prevalence of VPI and hypernasality compared to those without CL/P.

In order to develop targeted screening for VPI in CFM, we aimed to identify patient characteristics that are associated with an increased risk of VPI and speech/language impairment. Several records reported the relationship between CFM phenotypes and VPI besides the CL/P phenotype. Greater degrees of soft tissue deficiencies, mandibular hypoplasia, macrostomia, and skeletal deformities of the maxillary-malar complex were related to an increased prevalence of VPI.[11,13] In addition, developmental delays were found to correlate with VPI.[11] However, Chan et al[12] did not find an association between VPI and mandibular hypoplasia, soft tissue deficiencies, or macrostomia. In the literature various causes are implied as aetiology of VPI in CFM patients, including disruption of the tensor veli palatini muscle or unilateral palatal paralysis. Our hypothesis is that VPI in patients with CFM results from a combination of unilateral palatal paralysis and muscle underdevelopment, which results in asymmetry and fewer tissue of the palate.[31]

Please cite this article as: Tio, Pauline A.E., Rooijers, Wietse, de Gier, Henriëtte H.W. et al. Velopharyngeal insufficiency, speech, and language impairment in craniofacial microsomia: a scoping review, *British Journal of Oral and Maxillofacial Surgery*, https://doi.org/10.1016/j.bjoms.2023.09.008

Table 3
Speech, language, and hearing impairment. Data are % (No./Total cohort.)

Author	Speech impairment	Language problems	Hearing impairment
Chen, 2009[21]	38.2 (21/55) [a]		
Cohen, 1995[26]		37 (7/24)	
Collet, 2019[20]	35.4 (35/99)		70 (77/110)
Collet, 2011[27]	47.1 (64/136)		55 (75/136)
D'Antonio, 1998[19]	74 (14/19) [b]		78 (31/41)
Goetze, 2017[30]		50 (5/10) [c]	63 (5/8)
Johns, 2021[32]	42.2 (19/45) [d]		70.9 (117/141) [e]
Luce, 1977[13]	44.4 (8/18) [f]		
Strömland, 2007[22]	53.3 (8/15)		83 (15/18)

[a] include articulation errors/hypernasality/hyponasality;
[b] impaired articulation;
[c] language delays;
[d] speech or language impairment which was covered in an Individualized Educational Program;
[e] unilateral hearing loss;
[f] articulation errors

Table 4
Treatment VPI and speech impairment. Data are % (No./Total cohort.)

Author	Treatment VPI	Treatment speech impairment
Chan, 2021[12]	31.6 (6/19) [a]	45.5 (31/69)
Collet, 2019[20]		59.6 (68/114) [b]
Dentino, 2016[29]	100 (2/2)[c]	
Johns, 2021[32]		23.7 (40/169) [b]
van Hövell Tot Westerflier, 2019[33]		25 (10/40) unilateral; 78 (21/27) bilateral
Wallace, 2018[34]		59.2 (68/114) [d]

[a] 6 patients with CFM and CL/P who were diagnosed with VPI were treated with surgery (5 pharyngoplasty and 1 posterior pharyngeal flap), none of the patients with isolated CFM and VPI underwent corrective VPI surgery;
[b] speech therapy/language interventions;
[c] pharyngeal flap surgery;
[d] speech therapy.

As mentioned above, hearing loss is associated with speech and language problems. Therefore, it is not unexpected that records describing speech/language problems recorded a high prevalence of hearing impairment, with a prevalence ranging from 55% to 83%.[19,20,22,25,30,32] For example, Strömland et al[22] reported seven patients with bilateral hearing problems among eight patients with speech impairment. Collet et al[20] reported that in general adolescent patients with CFM scored lower on most measures of intelligibility, articulation and expressive language compared to unaffected controls. In CFM patients with a failed hearing screening and CFM patients with microtia and mandibular hypoplasia, the differences were most pronounced. No differentiation was made between unilateral or bilateral hearing problems, nor between distinct ear anomalies. Due to the fact that both hearing impairment and VPI are associated with speech problems and that both are prevalent in CFM patients, their distinct contribution to speech problems in CFM patients remains unknown.

For the purposes of evidence-based care for patients with CFM and VPI or speech impairment, we aimed to identify records describing the effectiveness of (surgical) interventions. However, few records reported treatment for speech or language impairment. Most of these records did not document what kind of speech therapy patients received. Furthermore, none of these records indicated the duration of the treatment or the established outcome. The available evidence concerning treatment options for these patients remains limited. However, it is concluded in these papers that early speech intervention is of high importance to improve outcomes.[11]

There is a general consensus that patients with CFM are, in addition to their hearing difficulties, also at risk for speech and language impairments.[6] In addition to hearing, also speech has a contributing factor in language development, and therefore speech impairment in CFM may also result in secondary developmental problems.[20,25] Therefore, timely recognition and treatment of speech and language impairment is essential. As suggested in the literature, CFM patients should be evaluated before the age of two years for speech impairment, alongside early hearing screening.[6] In addition, these patients should be monitored by speech and language therapists. In particular, patients with microtia plus mandibular hypoplasia require additional attention. CFM patients with CL/P are already monitored, because there is a greater risk of speech and language difficulties. It is recommended that if speech therapy is necessary, interventions are initiated as early as possible to prevent developmental delays and improve outcomes.

Limitations of this review include the descriptive character and lack of meta-analysis due to heterogeneity of the

Please cite this article as: Tio, Pauline A.E., Rooijers, Wietse, de Gier, Henriëtte H.W. et al. Velopharyngeal insufficiency, speech, and language impairment in craniofacial microsomia: a scoping review, *British Journal of Oral and Maxillofacial Surgery*, https://doi.org/10.1016/j.bjoms.2023.09.008

included studies. Future research should focus on the relationship between speech and language impairment and the CFM phenotype, in order to improve screening protocols. In addition, future research should focus on the effectiveness of treatment for VPI and speech impairment in CFM patients.

Conclusion

In conclusion, this review aimed to improve the understanding of VPI and speech-related challenges in patients with CFM. The prevalence of VPI and speech/language impairment was described, highlighting the variability in diagnostic techniques and criteria used among studies. In general, certain characteristics of CFM patients were found to have an increased risk of VPI, including CL/P, soft tissue deficiencies, mandibular hypoplasia, and developmental delays. Limited information was available on treatment options and the effectiveness of interventions for VPI and speech impairment in patients with CFM.

Conflict of interests

We have no conflicts of interest.

Ethics statement/confirmation of patient permission

Not applicable.

Acknowledgements

The authors would like to acknowledge Elise Krabbendam (Biomedical Information Specialist, Erasmus MC, Rotterdam, The Netherlands) for her assistance with the literature search.

Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.bjoms.2023.09.008.

References

1. Birgfeld CB, Luquetti DV, Gougoutas AJ, et al. A phenotypic assessment tool for craniofacial microsomia. *Plast Reconstr Surg* 2011;**127**:313–320.
2. Grabb WC. The first and second branchial arch syndrome. *Plast Reconstr Surg* 1965;**36**:485–508.
3. Murray JE, Kaban LB, Mulliken JB. Analysis and treatment of hemifacial microsomia. *Plast Reconstr Surg* 1984;**74**:186–199.
4. Poswillo D. The pathogenesis of the first and second branchial arch syndrome. *Oral Surg Oral Med Oral Pathol* 1973;**35**:302–328.
5. Rollnick BR, Kaye CI. Hemifacial microsomia and variants: pedigree data. *Am J Med Genet* 1983;**15**:233–253.
6. Renkema RW, Caron C, Heike CL, et al. A decade of clinical research on clinical characteristics, medical treatments, and surgical treatments for individuals with craniofacial microsomia: what have we learned? *J Plast Reconstr Aesthet Surg* 2022;**75**:1781–1792.
7. Renkema RW, Caron CJ, Pauws E, et al. Extracraniofacial anomalies in craniofacial microsomia: retrospective analysis of 991 patients. *Int J Oral Maxillofac Surg* 2019;**48**:1169–1176.
8. Caron CJ, Pluijmers BI, Joosten KF, et al. Feeding difficulties in craniofacial microsomia: a systematic review. *Int J Oral Maxillofac Surg* 2015;**44**:732–737.
9. Caron CJ, Pluijmers BI, Joosten KF, et al. Obstructive sleep apnoea in craniofacial microsomia: a systematic review. *Int J Oral Maxillofac Surg* 2015;**44**:592–598.
10. Rooijers W, Tio PA, van der Schroeff MP, et al. Hearing impairment and ear anomalies in craniofacial microsomia: a systematic review. *Int J Oral Maxillofac Surg* 2022;**51**:1296–1304.
11. Funayama E, Igawa HH, Nishizawa N, et al. Velopharyngeal insufficiency in hemifacial microsomia: analysis of correlated factors. *Otolaryngol Head Neck Surg* 2007;**136**:33–37.
12. Chan CH, Hu AC, Jain NS, et al. Velopharyngeal insufficiency in craniofacial microsomia: prevalence, diagnosis, and treatment. *J Craniofac Surg* 2021;**32**:2771–2773.
13. Luce EA, McGibbon B, Hoopes JE. Velopharyngeal insufficiency in hemifacial microsomia. *Plast Reconstr Surg* 1977;**60**:602–606.
14. Glade RS, Deal R. Diagnosis and management of velopharyngeal dysfunction. *Oral Maxillofac Surg Clin North Am* 2016;**28**:181–188.
15. Kummer AW, Marshall JL, Wilson MM. Non-cleft causes of velopharyngeal dysfunction: implications for treatment. *Int J Pediatr Otorhinolaryngol* 2015;**79**:286–295.
16. Dudas JR, Deleyiannis FW, Ford MD, et al. Diagnosis and treatment of velopharyngeal insufficiency: clinical utility of speech evaluation and videofluoroscopy. *Ann Plast Surg* 2006;**56**:511–557.
17. Perry JL. Anatomy and physiology of the velopharyngeal mechanism. *Semin Speech Lang* 2011;**32**:83–92.
18. Hashemi Hosseinabad H, Washington KN, Boyce SE, et al. Assessment of intelligibility in children with velopharyngeal insufficiency: the relationship between intelligibility in context scale and experimental measures. *Folia Phoniatr Logop* 2022;**74**:17–28.
19. D'Antonio LL, Rice Jr RD, Fink SC. Evaluation of pharyngeal and laryngeal structure and function in patients with oculo-auriculo-vertebral spectrum. *Cleft Palate Craniofac J* 1998;**35**:333–341.
20. Collett BR, Chapman K, Wallace ER, et al. Speech, language, and communication skills of adolescents with craniofacial microsomia. *Am J Speech Lang Pathol* 2019;**28**:1571–1581.
21. Chen EH, Reid RR, Chike-Obi C, et al. Tongue dysmorphology in craniofacial microsomia. *Plast Reconstr Surg* 2009;**124**:583–589.
22. Strömland K, Miller M, Sjögreen L, et al. Oculo-auriculo-vertebral spectrum: associated anomalies, functional deficits and possible developmental risk factors. *Am J Med Genet A* 2007;**143A**:1317–1325.
23. Anne S, Lieu JE, Cohen MS. Speech and language consequences of unilateral hearing loss: a systematic review. *Otolaryngol Head Neck Surg* 2017;**157**:572–579.
24. Sangen A, Royackers L, Desloovere C, et al. Single-sided deafness affects language and auditory development - a case-control study. *Clin Otolaryngol* 2017;**42**:979–987.
25. Collett BR, Wallace ER, Kapp-Simon KA, et al. Cognitive, motor, and language development of preschool children with craniofacial microsomia. *Cleft Palate Craniofac J* 2021;**58**:1169–1177.
26. Cohen MS, Samango-Sprouse CA, Stern HJ, et al. Neurodevelopmental profile of infants and toddlers with oculo-auriculo- vertebral spectrum and the correlation of prognosis with physical findings. *Am J Med Genet* 1995;**60**:535–540.
27. Collett BR, Speltz ML, Cloonan YK, et al. Neurodevelopmental outcomes in children with hemifacial microsomia. *Arch Pediatr Adolesc Med* 2011;**165**:134–140.
28. Dellon AL, Claybaugh GJ, Hoopes JE. Hemipalatal palsy and microtia. *Ann Plast Surg* 1983;**10**:475–479.
29. Dentino KM, Valstar A, Padwa BL. Cleft characteristics and treatment outcomes in hemifacial microsomia compared to non-syndromic cleft lip/palate. *Int J Oral Maxillofac Surg* 2016;**45**:679–682.

Please cite this article as: Tio, Pauline A.E., Rooijers, Wietse, de Gier, Henriëtte H.W. et al. Velopharyngeal insufficiency, speech, and language impairment in craniofacial microsomia: a scoping review, *British Journal of Oral and Maxillofacial Surgery*, https://doi.org/10.1016/j.bjoms.2023.09.008

30. Goetze TB, Sleifer P, Machada Rosa RF, et al. Hearing characterization in oculoauriculovertebral spectrum: a prospective study with 10 patients. *Am J Med Genet A* 2017;**173**:309–314.
31. Shprintzen RJ, Croft CB, Berkman MD, et al. Velopharyngeal insufficiency in the facio-auriculo-vertebral malformation complex. *Cleft Palate J* 1980;**17**:132–137.
32. Johns AL, Luquetti DV, Heike CL. Parental reports of intervention services and prevalence of teasing in a multinational craniofacial microsomia pediatric study. *J Craniofac Surg* 2021;**32**:2687–2691.
33. van Hövell Tot Westerflier C, Bracamontes IC, Tahiri Y, et al. Soft palate dysfunction in children with microtia. *J Craniofac Surg* 2019;**30**:188–192.
34. Wallace ER, Collett BR, Heike CL, et al. Behavioral-social adjustment of adolescents with craniofacial microsomia. *Cleft Palate Craniofac J* 2018;**55**:664–675.
35. Lancaster HS, Lien KM, Chow JC, et al. Early speech and language development in children with nonsyndromic cleft lip and/or palate: a meta-analysis. *J Speech Lang Hear Res* 2019;**63**:14–31.

Please cite this article as: Tio, Pauline A.E., Rooijers, Wietse, de Gier, Henriëtte H.W. et al. Velopharyngeal insufficiency, speech, and language impairment in craniofacial microsomia: a scoping review, *British Journal of Oral and Maxillofacial Surgery*, https://doi.org/10.1016/j.bjoms.2023.09.008