Journal of Cranio-Maxillo-Facial Surgery 50 (2022) 615–620

Contents lists available at ScienceDirect

Journal of Cranio-Maxillo-Facial Surgery

journal homepage: www.jcmfs.com

The effect of natural growth on chin point deviation in patients with unilateral craniofacial microsomia: A retrospective study

Ruben W. Renkema*, Irene van Beelen, Maarten J. Koudstaal, Cornelia J.J.M. Caron

The Dutch Craniofacial Center, Department of Oral and Maxillofacial Surgery, Erasmus University Medical Center, Sophia's Children's Hospital Rotterdam, the Netherlands

ARTICLE INFO

Article history:
Paper received 28 March 2020
Received in revised form
27 October 2021
Accepted 12 July 2022
Available online 19 July 2022

Keywords:
Craniofacial microsomia
Oculo-auriculo-vertebral syndrome
Hemifacial microsomia
Goldenhar syndrome
Chin point deviation
Growth

ABSTRACT

This study aimed to investigate the potential progressiveness of mandibular asymmetry and to study factors that influence chin point deviation in patients with unilateral craniofacial microsomia (CFM).

Paediatric patients with unilateral CFM with available radiologic imaging and medical photographs were included. Chin point deviation was measured on clinical photographs. A Jonckheere-Terpstra test and linear mixed model for repeated measurements assessed the relation of chin point deviation on natural growth, Pruzansky–Kaban score, and soft tissue score.

A total of 110 patients were included. The linear mixed model showed no statistically significant changes of chin point deviation during growth (effect estimate −0.006°, 95% CI −0.04° to −0.03°, $p = 0.74$). A statistical significant relation between both the Pruzansky-Kaban and soft tissue score on chin point deviation was found (effect estimate −5.10°, 95% CI −6.45° to −3.75°, $p \leq 0.001$ and effect estimate −3.42°, CI −5.86° to −0.98°, $p \leq 0.001$, respectively).

Within the limitations of the study it seems that craniofacial microsomia may be a non-progressive disorder, because chin point deviation did not change over time.

© 2022 The Author(s). Published by Elsevier Ltd on behalf of European Association for Cranio-Maxillo-Facial Surgery. This is an open access article under the CC BY license (http://creativecommons.org/licenses/by/4.0/).

1. Introduction

Craniofacial microsomia (CFM) is characterized by unilateral or bilateral hypoplasia of facial tissues. Various models have been developed to classify the degree of facial hypoplasia in patients with CFM (*Pruzansky 1969*; *Kaban* et al. *1988*; *Horgan* et al. *1995*; *Cousley and Calvert 1997*). Mandibular hypoplasia, which is seen in 89%–100% of the patients, is commonly described by the Pruzansky-Kaban classification (*Kaban et al. 1981*, *1986*; *Cousley and Calvert 1997*). This classification, which is based on radiographic evaluation, ranks the severity of mandibular hypoplasia from type I to types IIa, IIb and III. The degree of hypoplasia of all involved facial structures in patients with CFM is often assessed by the O.M.E.N.S. classification, which scores hypoplasia of the orbit, mandible, ears, facial nerve, and soft tissues (*Vento* et al. *1991*; *Birgfeld* et al. *2011*).

Functional problems associated with mandibular hypoplasia such as feeding or breathing or aesthetic difficulties may necessitate treatment (*Birgfeld and Heike 2012*; Caron et al. 2017, 2018). Timing of treatment depends on various aspects, including the natural growth of the mandible. The potential progressiveness of mandibular growth in CFM is debated in literature (*Converse* et al. *1973*; *Murray* et al. *1984*; *Kaban* et al. *1986*; *Polley* et al. *1997*; *Kusnoto* et al. *1999*; *Kearns* et al. *2000*; *Meazzini* et al. *2012*; *Ongkosuwito* et al. *2013*). Some authors advocate early treatment to prevent increasing facial asymmetry and to increase function, whereas others advise postponing treatment until adulthood to prevent tissue damage and unnecessary surgery, as it has been shown that early intervention increases the chances of needing additional surgery later in life, most likely due to the iatrogenic damage done (*Polley* et al. *1997*; *Nagy* et al. *2009*).

Deviation of the chin point on clinical photographs is a simple technique that can be used to estimate the severity of facial asymmetry (*Gursoy* et al. *2008*; *Ohtani* et al. *2012*; *Fattah* et al. *2014*; *Ko* et al. *2017*). Previous studies on three-dimensional (3D) analysis of the mandible in CFM showed that mandibular hypoplasia in CFM leads to a rotation to the affected side, which is greater in patients

* Corresponding author. Department of Oral and Maxillofacial Surgery, Erasmus University Medical Center, Sophia's Children's Hospital, 's Gravendijkwal 230, 3015 CE, Rotterdam, the Netherlands.
E-mail address: r.renkema@erasmusmc.nl (R.W. Renkema).

https://doi.org/10.1016/j.jcms.2022.07.006
1010-5182/© 2022 The Author(s). Published by Elsevier Ltd on behalf of European Association for Cranio-Maxillo-Facial Surgery. This is an open access article under the CC BY license (http://creativecommons.org/licenses/by/4.0/).

with more severe mandibular hypoplasia (Kim et al., 2018; Kaya et al., 2019). Although these studies based on radiographic data are essential to elucidate the extent of CFM, the effect of hypoplasia and growth on facial asymmetry in patients with CFM has not been examined. This study aims to investigate the influence of mandibular and facial soft tissue hypoplasia on chin point deviation in patients with unilateral CFM.

2. Materials and methods

2.1. Patients and procedures

This was a retrospective study performed at the Craniofacial Unit of the Erasmus University Medical Center, Rotterdam, the Netherlands. The use of clinical data was approved by the Institutional Review Board (MEC-2013-575). Patients with unilateral CFM were included if facial clinical photographs, radiologic images and medical history were available. Patients with bilateral CFM or other craniofacial syndromes were excluded. All patients with craniofacial anomalies were regularly and structurally seen at the outpatient clinic, after first presentation, at the ages of 4, 6, 9, 12 15, 18 and 21 years. At these visits, clinical photographs were taken. All available photos were assessed for analysis. The chin point deviation was measured if the photograph was taken right in front of the patient. Photos of patients smiling, crying or with a fully open mouth were excluded, as the chin point could not be measured reliably. If a patient had craniofacial surgery that affected the mandible and/or chin, only the preoperative photographs were used. Photographs were also excluded if there was any uncertainty about the type of surgery that was performed.

The severity of mandibular hypoplasia was classified by the Pruzansky–Kaban classification, based on computed tomography (CT) scans or panoramic radiographs *(Pruzansky 1969*; Kaban et al. 1981, 1986*)*. Type I mandibles are small but have normal morphology. Type II is divided into types IIa and IIb. In type IIa, the mandibular ramus is abnormal in size and morphology; in type IIb the mandibular ramus is abnormal in size and morphology and the TMJ is abnormally placed. Type III contains mandibles with an absent ramus, condyle and temporomandibular joint. Potential involvement of soft tissue deficiency on chin point deviation was assessed by using the O.M.E.N.S. classification. In this classification the soft tissue is scored from 0 to 3, ranging from no soft tissue deficiency to a severe soft tissue deficiency *(Vento* et al. *1991*; *Birgfeld* et al. *2016)*.

Chin point deviation (CPD) was measured on frontal facial medical photographs, using Adobe Illustrator CS6. Frontal view photographs were taken with the nose pointing towards the lens and showing equal amounts of both sides of the face. To establish reproducible and reliable measurements, chin point deviation was measured according to standardized reference points suitable for two-dimensional analysis in patients with facial asymmetry, as described by *Berlin* et al. *(2014)*. A sagittal line crossing the nasion and subnasal point was defined as the midline. A second line from the nasion through the gnathion was made. The angle between this line and the midline was defined as the chin point deviation *(Fattah* et al. *2014)*. Fig. 1 shows the measurements obtained in a patient over a decade.

Two observers R.W.R. and I.V.B. measured the chin point deviation in all photographs, to measure the interrater reliability. One observer (R.W.R.) measured the deviation twice in a 3-month time interval, to calculate the intrarater reliability.

2.2. Statistical analysis

Descriptive statistics were used. The intra- and interrater reliability was calculated using the intraclass correlation (ICC) coefficient based on a two-way random model with an absolute agreement definition, reporting single measures. The values of the ICC range from 0 to 1; values of 0.8 and higher were interpreted as excellent agreement. A Jonckheere–Terpstra test was used to assess the association between the Pruzansky–Kaban classification and the soft tissue score and the first measured CPD. This test was used because it determines whether there is a statistically significant trend between an ordinal independent variable and a continuous variable in an *a priori* ordering. We used a linear mixed model to see how the CPD changes with respect to age. The independent variables in this model were age, sex, the Pruzansky–Kaban classification, and the soft tissue score of the O.M.E.N.S. classification. The association of the Pruzansky–Kaban classification and soft tissue score on the CPD were assessed in separate models, as the variables are not dependent of each other, taking into consideration that patients with a severe phenotype of CFM show hypoplasia of multiple facial tissues. A random intercept and a random slope of age were used to account for the within-subject correlations. Statistical analysis was performed using IBM SPSS, version 24. All statistical tests used a two-sided significance level of 0.05.

3. Results

In total, 218 patients with unilateral CFM were evaluated during the study period. Of the 218 patients, 110 were included in the study; these patients were 3 and 56 years of age. A total of 108 patients were excluded because of the presence of other facial anomalies, surgeries of the mandible or chin, or insufficient imaging data. Slightly more male ($n = 56$) than female ($n = 54$) patients were included; the affected side was equally distributed in the studied cohort (55 left and 55 right). Table 1 shows the patient characteristics, Pruzansky–Kaban classification and the soft tissue score of the O.M.E.N.S. score in the studied cohort.

The total number of measurements per patient varied, as did the age at which patients underwent measurement (Table 2). All 110 included patients had one measurement, 69 patients had two measurements, 49 patients had three, and 23 patients had four or more. The mean chin point deviation of all patients at first measurement was 3.8° (SD = 3.2°). Subdivided in Pruzansky–Kaban type I, IIa, IIb and III, the mean chin point deviations were 2.3°, 2.9°, 4.2°, and 7.4°, respectively (Table 3). The ICC coefficient was 0.96 (95% CI = 0.94–0.97) for the interrater reliability and 0.88 (95% CI = 0.82–0.92) for the intrarater reliability. They were both interpreted as indicating excellent agreement.

A linear mixed model for repeated measurements showed no significant association between age and chin point deviation ($p = 0.74$), as for sex and chin point deviation ($p = 0.41$). The Pruzansky–Kaban score was significantly associated with chin point deviation ($p \leq 0.001$). Patients with a Pruzansky–Kaban type III mandible had a 5.1° larger chin point deviation to the affected side compared to patients with a Pruzansky–Kaban type I mandible (effect estimate = −5.10°, 95% CI = −6.45° to −3.75°). The soft tissue score was also significantly associated with chin point

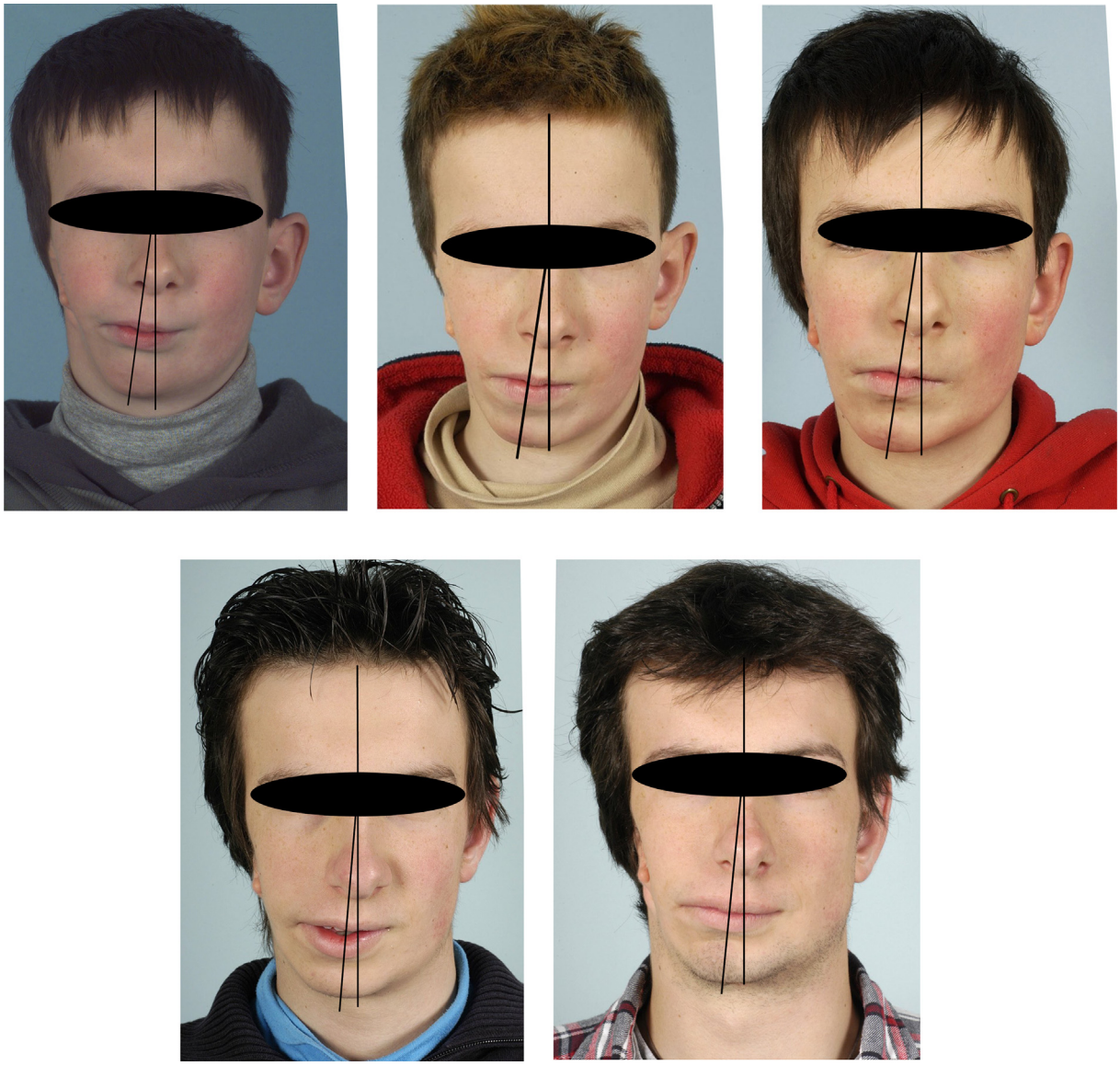

Fig. 1. Example of measurement.

Table 1
Patient characteristics.

Sample characteristics	
Sample, n	110
Age (year), range	3–56
Gender, n	
Male	56
Female	54
Affected side, n	
Left	55
Right	55
Pruzansky-Kaban classification, n (%)	
Type I	41 (37)
Type IIa	29 (26)
Type IIb	18 (17)
Type III	22 (20)
Soft Tissue score of O.M.E.N.S., n (%)	
0	19 (17%)
I	45 (41%)
II	38 (35%)
III	8 (7%)

deviation ($p \leq 0.001$). The chin point deviation to the affected side was 3.4° larger in patients with a soft tissue score III compared to patients with a soft tissue score I (effect estimate = −3.42°, CI = −5.86° to −0.98°). All measurements were taken into account in this analysis, except for one patient who was considered an outlier because of her age of 56 years at first measurement and was therefore excluded from this analysis (median age at first measurement, 7 years; 90th percentile of all first measurements, 18 years). Measurements of patients more than 18 years of age were excluded in the analysis on the relation between chin point deviation and growth. The results of the models is shown in Table 4.

The Jockheere–Terpstra test, which only used only the first CPD measurement of all 110 patients, also showed that patients with a higher Pruzansky–Kaban score had a significantly more deviated chin point ($p \leq 0.001$), as was for patients with a higher soft tissue score ($p \leq 0.001$).

Table 2
Numbers and age of measurements.

Total number of measurements per patient	Number of patients with measurement	Median age at time of measurement *(in years)*	Age range at time of measurement *(in years)*
1	111	7	3–56
2	69	10	5–29
3	49	12	9–22
4	23	15	12–19
5	11	17	14–18
6	5	19	18–21
7	1	20	20

Table 3
Pruzanksy-Kaban score and chin point deviation.

	N	Mean *(in degrees) (including 95% CI)*	Minimum and maximum measurement *(in degrees)*
P-K type I	41	2.30 (1.81–2.79)	0.0–6.9
P-K type IIa	29	2.91 (1.86–3.97)	0.0–9.9
P-K type IIb	18	4.18 (2.74–5.62)	0.0–10.8
P-K-type III	22	7.37 (5.78–8.96)	2.6–14.0
Total	110	3.21 (3.18–4.39)	0.0–14.0

*P-K: Pruzansky-Kaban; CI: Confidence Interval

Table 4
Estimates of Fixed Effects on chin point deviation.

PARAMETER	**EFFECT ESTIMATES** (IN DEGREES)	**CONFIDENCE INTERVAL** (IN DEGREES)
AGE*	−0.006	−0.04 – 0.03
MALE	−0.41	−1.39 – 0.58
FEMALE	0 (redundant)	.
PRUZANSKY-KABAN I	−5.1	−6.45–−3.75
PRUZANSKY-KABAN IIA	−4.5	−5.97–−3.07
PRUZANSKY-KABAN IIB	−3.2	−4.83–−1.57
PRUZANSKY-KABAN III	0 (redundant)	.
SOFT TISSUE 0	−3.42	−5.86–−0.98
SOFT TISSUE I	−2.15	−4.38 – 0.08
SOFT TISSUE II	0.02	−2.24 – 2.27
SOFT TISSUE III	0 (redundant)	.

*Patients above 18 years of age were excluded in this analysis

4. Discussion

This study aimed to research the potential progressiveness of mandibular asymmetry in unilateral CFM and to examine factors that influence mandibular asymmetry. A total of 110 patients were included. More patients with a Pruzansky–Kaban type I or IIa were included than patients with type IIb or III, which is in line with the literature. Both sex and the affected side were equally divided in our population. Other studies did find differences in sex and affected side predominance in CFM, although the meta-analysis by Xu et al. showed no differences in male–female and left–right ratio *(Xu et al. 2015*; Caron et al. 2017*)*.

No significant changes in chin point deviation occurred during growth. It can therefore be assumed that growth of both the affected and unaffected side of the mandible is similar in patients with unilateral CFM. This was also shown by Ongkosuwito et al., who studied panoramic x-rays, found that patients with CFM experience similar growth compared to a normal population, but start and end with a smaller mandible *(Ongkosuwito et al. 2013)*. Polley et al. studied longitudinal records of 26 patients with unilateral CFM and assessed posterior–anterior cephalometric radiographs. They concluded that the mandibular asymmetry is not progressive and that the affected and unaffected sides show parallel growth *(Polley et al. 1997)*. Newer methods such as 3D CT scans can be used to describe the mandibular deformity in more detail. Kaya et al. showed, by using principal component analysis, that the mandible rotates to the affected side due to lateral rotation and shortening of the condyle–gonial height with outward bending of the mandibular angle *(Kaya et al. 2019)*. The unaffected mandibular side in patients with unilateral CFM is often bending inwards because of compensatory remodeling. No differences were observed during growth, as both younger and older patients showed inward bending of the unaffected side *(Kaya et al. 2019)*. Kim et al. studied 3D reconstructed mandibles from CT scans of patients with CFM to investigate growth of the anatomical regions of the mandible separately *(Kim et al. 2018)*. They found that the angulation in patients with milder type (Pruzansky–Kaban type II), but not in those with severely hypoplastic mandibles (Pruzansky–Kaban type III), may decrease with age, although the type II mandibles still show more than 6° angulation compared to those in healthy controls *(Kim et al. 2018)*. This study was based on cross-sectional analysis of 28 patients with CFM categorized according to various age groups, which could explain the different outcomes compared to those of our study, which assessed 110 patients with CFM in a longitudinal analysis.

Deviation of the chin point is influenced by the Pruzansky–Kaban score and the soft tissue score on the O.M.E.N.S. scale. This study shows a strong association between the Pruzansky score and soft tissue score with the chin point deviation. However, the variation between patients, especially in the effect of the soft tissue score on the chin point deviation, is considerable, as is displayed by the wide confidence interval. This individual variation was also seen in a recent study by Apostolopous et al., who showed, by using 3D mapping in eight patients, that the bony and soft tissue hypoplasia were correlated at the gonion and malar region, whereas other facial areas showed a poor correlation (Apostolopoulos et al., 2021).

Identifying reliable landmarks and thus measuring chin point deviation in patients with CFM is difficult, as facial anatomy varies among patients. A horizontal line through the lateral canthi with a perpendicular sagittal line can be used to determine the midline of the face. However, this is questionable in patients with CFM, as orbital dystopia, a common feature in these patients, may influence placement of the landmarks *(Fattah et al. 2014)*. In this study, frontal view photographs were taken in a straight direction

showing equal amounts of both sides of the face. The midline of the face was determined by placing a sagittal line through the nasion and subnasal point. Additionally, the chin point, defined as the gnathion, can be difficult to determine on medical photographs, as it is not always as visible in these as compared to radiologic images. Although these difficulties with landmark placement in studying patients with CFM are inevitable, excellent intra- and interobserver agreement was reached because of the strict methodology used.

Facial asymmetry in this study was determined by measuring chin point deviation on two-dimensional photographs. By using this method, the complex nature of mandibular hypoplasia in CFM might not have been fully represented. Hard conclusions on the potential progressiveness on CFM could therefore not be made. Nonetheless, this study showed that if any progressiveness was present, it did not have a visible effect on facial asymmetry.

In recent decades, conflicting results on the progressiveness of CFM have been published. Early treatment could stimulate mid-facial growth and lead to better facial symmetry *(Kaban et al. 1986, 1988; Kearns* et al. *2000; Shetye* et al. *2006; Weichman* et al. *2017; Chen* et al. *2020; Qiu* et al. *2021)*. Especially in patients with mild mandibular hypoplasia, additional surgery was not always needed *(Kaban et al. 1986, 1988)*. However, recent systematic reviews by Nagy et al. and Pluijmers et al. showed no evidence for long-term stability of early treatment in patients with CFM *(Nagy* et al. *2009; Pluijmers* et al. *2014)*. The earlier correction of mandibular asymmetry is performed, the more surgical procedures are needed to correct the asymmetry later in life *(Pluijmers* et al. *2019)*. The findings in this study support informing parents and children that the condition will not get worse over time and should also support avoiding early surgery for the sake of preventing increasing facial asymmetry.

5. Conclusion

Within the limitations of the study it seems that craniofacial microsomia may be a non-progressive disorder, because chin point deviation did not change over time.

Author contributions

All authors made substantial contributions to the conception and design, acquisition of data, or analysis and interpretation of data. All authors were involved in drafting the article or critically revising it for important intellectual content. Finally, all authors approved the version to be published.

Funding statement

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Declaration of competing interest

There are no conflicts of interest in the materials or subject matter dealt with in the manuscript.

Acknowledgements

The authors would like to thank J.M. van Rosmalen, biostatistician, for his help with reviewing the data and for advice on the statistical analysis.

References

Apostolopoulos, K., Bous, R.M., ElNaghy, R., Kumar, A.R., Valiathan, M., 2021. Examining the variability of bone and soft tissue morphology in hemifacial microsomia: a case series of 8 patients. J. Cranio-Maxillo-Fac. Surg. 49 (5), 352–357.

Berlin, N.F., Berssenbrugge, P., Runte, C., Wermker, K., Jung, S., Kleinheinz, J., Dirksen, D., 2014. Quantification of facial asymmetry by 2D analysis–a comparison of recent approaches. J. Cranio-Maxillo-Fac. Surg. 42 (3), 265–271.

Birgfeld, C.B., Heike, C., 2012. Craniofacial microsomia. Semin. Plast. Surg. 26 (2), 91–104.

Birgfeld, C.B., Heike, C.L., Saltzman, B.S., Leroux, B.G., Evans, K.N., Luquetti, D.V., 2016. Reliable classification of facial phenotypic variation in craniofacial microsomia: a comparison of physical exam and photographs. Head Face Med. 12 (1), 14.

Birgfeld, C.B., Luquetti, D.V., Gougoutas, A.J., Bartlett, S.P., Low, D.W., Sie, K.C., Evans, K.N., Heike, C.L., 2011. A phenotypic assessment tool for craniofacial microsomia. Plast. Reconstr. Surg. 127 (1), 313–320.

Caron, C., Pluijmers, B.I., Joosten, K.F.M., Dunaway, D., Padwa, B.L., Wolvius, E.B., Koudstaal, M.J., 2018. Feeding difficulties in craniofacial microsomia: a multicenter retrospective analysis of 755 patients. J. Cranio-Maxillo-Fac. Surg. 46 (10), 1777–1782.

Caron, C., Pluijmers, B.I., Maas, B., Klazen, Y.P., Katz, E.S., Abel, F., van der Schroeff, M.P., Mathijssen, I.M.J., Dunaway, D.J., Mills, C., Gill, D.S., Bulstrode, N., Padwa, B.L., Wolvius, E.B., Joosten, K.F.M., Koudstaal, M.J., 2017a. Obstructive sleep apnoea in craniofacial microsomia: analysis of 755 patients. Int. J. Oral Maxillofac. Surg. 46 (10), 1330–1337.

Caron, C., Pluijmers, B.I., Wolvius, E.B., Looman, C.W.N., Bulstrode, N., Evans, R.D., Ayliffe, P., Mulliken, J.B., Dunaway, D., Padwa, B., Koudstaal, M.J., 2017b. Craniofacial and extracraniofacial anomalies in craniofacial microsomia: a multicenter study of 755 patients. J. Cranio-Maxillo-Fac. Surg. 45 (8), 1302–1310.

Chen, X., Yang, X., Gu, S., Li, H., Zin, M.A., Mooi, W.J., Han, W., Zhang, Y., Chai, G., 2020. Early hemi-mandibular lengthening by distraction osteogenesis contributes to compensatory maxillary growth. J. Cranio-Maxillo-Fac. Surg. 48 (4), 357–364.

Converse, J.M., Coccaro, P.J., Becker, M., Wood-Smith, D., 1973. On hemifacial microsomia. The first and second branchial arch syndrome. Plast. Reconstr. Surg. 51 (3), 268–279.

Cousley, R.R., Calvert, M.L., 1997. Current concepts in the understanding and management of hemifacial microsomia. Br. J. Plast. Surg. 50 (7), 536–551.

Fattah, A.Y., Caro, C., Khechoyan, D.Y., Tompson, B., Forrest, C.R., Phillips, J.H., 2014. Cephalometric outcomes of orthognathic surgery in hemifacial microsomia. J. Craniofac. Surg. 25 (5), 1734–1739.

Gursoy, S., Hukki, J., Hurmerinta, K., 2008. Five year follow-up of mandibular distraction osteogenesis on the dentofacial structures of syndromic children. Orthod. Craniofac. Res. 11 (1), 57–64.

Horgan, J.E., Padwa, B.L., LaBrie, R.A., Mulliken, J.B., 1995. OMENS-Plus: analysis of craniofacial and extracraniofacial anomalies in hemifacial microsomia. Cleft Palate-Craniofacial J. 32 (5), 405–412.

Kaban, L.B., Moses, M.H., Mulliken, J.B., 1986. Correction of hemifacial microsomia in the growing child: a follow-up study. Cleft Palate J. 23 (Suppl. 1), 50–52.

Kaban, L.B., Moses, M.H., Mulliken, J.B., 1988. Surgical correction of hemifacial microsomia in the growing child. Plast. Reconstr. Surg. 82 (1), 9–19.

Kaban, L.B., Mulliken, J.B., Murray, J.E., 1981. Three-dimensional approach to analysis and treatment of hemifacial microsomia. Cleft Palate J. 18 (2), 90–99.

Kaya, O., Pluijmers, B.I., Staal, F., Ruff, C., Padwa, B.L., Koudstaal, M.J., Dunaway, D.J., 2019. Describing the mandible in patients with craniofacial microsomia based on principal component analysis and thin plate spline video analysis. Int. J. Oral Maxillofac. Surg. 48 (3), 302–308.

Kearns, G.J., Padwa, B.L., Mulliken, J.B., Kaban, L.B., 2000. Progression of facial asymmetry in hemifacial microsomia. Plast. Reconstr. Surg. 105 (2), 492–498.

Kim, B.C., Bertin, H., Kim, H.J., Kang, S.H., Mercier, J., Perrin, J.P., Corre, P., Lee, S.H., 2018. Structural comparison of hemifacial microsomia mandible in different age groups by three-dimensional skeletal unit analysis. J. Cranio-Maxillo-Fac. Surg. 46 (11), 1875–1882.

Ko, E.W., Chen, P.K., Lo, L.J., 2017. Comparison of the adult three-dimensional craniofacial features of patients with unilateral craniofacial microsomia with and without early mandible distraction. Int. J. Oral Maxillofac. Surg. 46 (7), 811–818.

Kusnoto, B., Figueroa, A.A., Polley, J.W., 1999. A longitudinal three-dimensional evaluation of the growth pattern in hemifacial microsomia treated by mandibular distraction osteogenesis: a preliminary report. J. Craniofac. Surg. 10 (6), 480–486.

Meazzini, M.C., Mazzoleni, F., Bozzetti, A., Brusati, R., 2012. Comparison of mandibular vertical growth in hemifacial microsomia patients treated with early distraction or not treated: follow up till the completion of growth. J. Cranio-Maxillo-Fac. Surg. 40 (2), 105–111.

Murray, J.E., Kaban, L.B., Mulliken, J.B., 1984. Analysis and treatment of hemifacial microsomia. Plast. Reconstr. Surg. 74 (2), 186–199.

Nagy, K., Kuijpers-Jagtman, A.M., Mommaerts, M.Y., 2009. No evidence for long-term effectiveness of early osteodistraction in hemifacial microsomia. Plast. Reconstr. Surg. 124 (6), 2061–2071.

Ohtani, J., Hoffman, W.Y., Vargervik, K., Oberoi, S., 2012. Team management and treatment outcomes for patients with hemifacial microsomia. Am. J. Orthod. Dentofacial Orthop. 141 (4 Suppl. l), S74–S81.

Ongkosuwito, E.M., van Vooren, J., van Neck, J.W., Wattel, E., Wolvius, E.B., van Adrichem, L.N., Kuijpers-Jagtman, A.M., 2013. Changes of mandibular ramal height, during growth in unilateral hemifacial microsomia patients and unaffected controls. J. Cranio-Maxillo-Fac. Surg. 41 (2), 92–97.

Pluijmers, B.I., Caron, C., van de Lande, L.S., Schaal, S., Mathijssen, I.M., Wolvius, E.B., Bulstrode, N., Evans, R.D., Padwa, B.L., Koudstaal, M.J., Dunaway, D.J., 2019. Surgical correction of craniofacial microsomia: evaluation of interventions in 565 patients at three major craniofacial units. Plast. Reconstr. Surg. 143 (5), 1467–1476.

Pluijmers, B.I., Caron, C.J., Dunaway, D.J., Wolvius, E.B., Koudstaal, M.J., 2014. Mandibular reconstruction in the growing patient with unilateral craniofacial microsomia: a systematic review. Int. J. Oral Maxillofac. Surg. 43 (3), 286–295.

Polley, J.W., Figueroa, A.A., Liou, E.J., Cohen, M., 1997. Longitudinal analysis of mandibular asymmetry in hemifacial microsomia. Plast. Reconstr. Surg. 99 (2), 328–339.

Pruzansky, S., 1969. Not all dwarfed mandibles are alike. Birth Defects 5, 120–129.

Qiu, X., Sun, H., Zhu, M., Chen, X., Chai, G., Yang, X., Zhang, Y., 2021. Using orthodontic elastic traction during the active period of distraction osteogenesis to increase the effective vertical extension of hemifacial microsomia patients: a multi-center randomized clinical trial. J. Cranio-Maxillo-Fac. Surg. 49 (11), 1054–1063. https://doi.org/10.1016/j.jcms.2021.06.013.

Shetye, P.R., Grayson, B.H., Mackool, R.J., McCarthy, J.G., 2006. Long-term stability and growth following unilateral mandibular distraction in growing children with craniofacial microsomia. Plast. Reconstr. Surg. 118 (4), 985–995.

Vento, A.R., LaBrie, R.A., Mulliken, J.B., 1991. The O.M.E.N.S. classification of hemifacial microsomia. Cleft Palate-Craniofacial J. 28 (1), 68–76. ; discussion 77.

Weichman, K.E., Jacobs, J., Patel, P., Szpalski, C., Shetye, P., Grayson, B., McCarthy, J.G., 2017. Early distraction for mild to moderate unilateral craniofacial microsomia: long-term follow-up, outcomes, and recommendations. Plast. Reconstr. Surg. 139 (4), 941e–953e.

Xu, S., Zhang, Z., Tang, X., Yin, L., Liu, W., Shi, L., 2015. The influence of gender and laterality on the incidence of hemifacial microsomia. J. Craniofac. Surg. 26 (2), 384–387.