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Case Report

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Incidence of Oblique Traction Vector of the Pterigomasseteric Sling in Relapse of Mandibular Distraction Osteogenesis after Proportional Overcorrection in Goldenhar Syndrome: Case Report

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ABSTRACT

Distraction osteogenesis in the facial territory is a very useful tool in patients with dysmorphosis and syndromes. It allows histiogenesis after progressive traction of the intervened bone tissue, which increases in size according to the planned vector and the range of distraction.

The existence of post-distraction osteogenic relapse depends on multiple factors. In the maxillary region, it corresponds to approximately 10% of the total number of millimetres (mm) distracted. Anatomical and biomechanical factors play a fundamental role in the variability of these results.

The purpose of our work is to present the results obtained after vertical distraction of the mandibular ramus in a pediatric patient with Goldenhar syndrome. Following a proportional overcorrection, an asymmetric relapse was evident in the postoperative 3D measurements, with a greater relapse at the posterior border of the mandibular ramus. Additionally, we provide a literature review regarding the possible role of the pterygomasseteric sling vector of forces in the postoperative outcomes of these patients.

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Introduction

Hemifacial Microsomia (HFM) can be caused by various congenital and developmental anomalies, among which is Goldenhar syndrome. HFM is a congenital craniofacial syndrome that affects the various structures derived from the first and second pharyngeal arches: mandible, mandibular condyle, articular cavity, maxilla, orbit, auditory canal, soft tissues, and muscles innervated by the trigeminal and facial nerves. It was first described by Carl F. Von Arlt, but it was also known as Goldenhar syndrome, named after Maurice Goldenhar, being a variant of HFM. The severity of the pathology depends on the degree of penetrance of the syndrome and the number of affected structures. In most cases, it is unilateral, and facial asymmetry is due to the lack of development on the affected side, presenting vertical alterations and mandibular transverse changes derived from inadequate development of the temporomandibular joint (TMJ) components and ipsilateral mandibular ramus, causing maxillary development impairment, deformity, and occlusal alterations [1]. Patients with HFM may have auditory, respiratory, and swallowing difficulties due to underdevelopment of cervicofacial structures, and may also have diagnoses of obstructive sleep apnea syndrome, cleft lip and palate, musculoskeletal, cutaneous system malformations, and other systemic conditions [2].

The most commonly used classification for HFM is Pruzansky's classification, which categorizes the pathology into three grades: Grade I: minimal mandibular hypoplasia with normal structures; Grade II: small and variably shaped condyle, ramus, and sigmoid notch; Grade III: absence of mandibular ramus, including the TMJ. This classification was modified by Kaban, dividing Grade II into Type IIA (hypoplasia and inappropriate mandibular position, but allowing functional mandibular movement) and IIB (hypoplastic mandibular ramus, with abnormal shape and location, causing mandibular dysfunction). Currently, there are other classifications, such as the "OMENS" classification proposed by Vento in 1999, with its latest update in 2011, or the "craniofacial deformity score" introduced in 2001. While these classifications are more laborious to perform, they allow for a more precise diagnosis that helps extrapolate data in a more comparable manner for scientific research [1,3].

The traditional approach to treating craniofacial skeletal deformities has been through osteotomies and/or bone grafts. However, postoperative aesthetic, functional, or relapse problems have mainly been attributed to the surrounding soft tissues not adapting to the new position of the bones after treatment [4]. Currently, the management of HFM requires a multidisciplinary approach. Although there are no established protocols for its management, combined surgical and orthopedic therapies are suggested, depending on the type of affected structures and the severity of the pathology. Among the surgical procedures described are distraction osteogenesis (DO) and orthognathic surgery to correct facial asymmetry due to the lack of development of bone structures [1]. Within the options for bone reconstruction are ATM reconstruction with autologous graft, total alloplastic joint replacement, and microvascularized flaps [3]. Other surgical alternatives described for soft tissue reconstruction include pedicled flaps, microvascular free tissue transfer, structural fat grafting, alloplastic implants, auricular reconstruction, and functional facial reconstruction; the latter includes sural nerve grafting or direct coaptation with the contralateral facial nerve [5].

Ilizarov demonstrated the scientific basis and clinical effectiveness of bone distraction or lengthening in long bones in 1988. Based on his work, other authors conducted a series of laboratory studies to test the feasibility of distracting the membranous bones of the craniofacial skeleton [6]. Thus, the first reported case at this level was by Snyder, performed on the mandible of a dog. Later, MacCarthy performed mandibular lengthening with extraoral appliances in humans with congenital and acquired mandibular hypoplasias, becoming the main proponent of facial distraction [7]. Subsequently, in 1994, Monasterio and Molina published an important series of such cases, demonstrating the viability of the procedure [8].

There are several factors to consider to ensure the success of osteodistraction. Among them, planning and choosing the distraction vector are crucial, considering the biological forces of the maxillofacial region that will influence the morphology of the newly formed bone during the active period of distraction osteogenesis and eventual relapse, determined by muscular activity and soft tissue [9,10]. Significant relapse has been described in the literature in DO and orthognathic surgery procedures, especially in patients with mandibular deficiency, due to the inability of the musculature to adapt to the new anatomical relationships achieved in surgery. Thus, it is important to understand the biomechanical behavior of the masticatory muscles when planning dentofacial morphological changes to achieve long-term stability. The internal pterygoid and masseter muscles work synergistically, moving the mandible upward and forward, acting as powerful elevators of the mandible, while the temporal muscle has a mandibular elevating and retrusive action, depending on the contracted muscle fibers [11]. Thus, the temporal muscle force vector is generated through a line that intersects the highest point of the coronoid process and runs tangentially to the anterior border of the ascending ramus, while for the pterigomasseteric sling, it is generated by a line connecting the gonion and intersecting the point where the frontal bone joins the zygomatic process of the temporal bone, forming a posterosuperior oblique vector that follows a direction from front to back, from bottom to top, and from inside to outside [12]. The desired mandibular change in shape and function can be achieved by selecting and controlling these force vectors operating during active distraction [9].

The Wolff's law and Moss's functional matrix theory established the relationship between bone morphology and the functional forces of the soft tissues acting upon it. According to the functional matrix theory, any alteration in the size, shape, and growth of the skeletal unit is secondary and compensatory to changes in its related functional matrix. Thus, a direct relationship has been reported between mandibular deformity and the state of the masticatory muscles in patients with HFM. There are several authors, such as Marsh and Marquez, who propose that the mandibular ramus dysmorphology in HFM could result from the interaction between primary bone and soft tissue deficiencies [13,14]. Thus, muscular movement during mandibular growth can generate a high variability in mandibular growth [11].

It should be considered that during the activation phase of osteodistraction to elongate the mandibular ramus, if the device was placed vertically, a change in its orientation can occur guided by the neuromuscular effect determined by the direction of the previously described vectors, generating a counterclockwise mandibular rotation [10].

Case Report

The evaluation of the patient, who was treated with distraction osteogenesis, was conducted in a private practice in Santiago, Chile. The patient was a 7-year-old male diagnosed with right-sided hemifacial microsomia (HFM), classified according to the modified Pruzansky classification by Kaban as Grade I. Upon directed clinical examination, he presents right facial asymmetry.

The surgical treatment was performed in the operating room under general anesthesia, with nasotracheal intubation. A right mandibular vestibulotomy access was made to expose the ipsilateral mandibular ramus, angle, and body. A horizontal osteotomy was performed at the mandibular ramus level using a piezoelectric device to achieve vertical distraction of the mandibular ramus, according to virtual planning (Figure 1). A mandibular internal fixation distractor (CIBEY X0101-20 model), distributed by Orthomax Limited Chile, was used, allowing for a distraction length of up to 20 mm. To achieve proportional overcorrection, considering the difference in length between the affected ramus and the healthy ramus was 18.69 mm. Taking into account that literature describes a 10% relapse of the maxilla, 1.86 mm would be required, totaling 20.5 mm. to distract, thus, this distractor meets the planning requirements. The distractor was fixed with five 7 mm screws each from the 2.0 system, with the support of a transjugal system for the placement of distal screws (Figure 2). Closure of the surgical wound was performed with vicryl 4-0 in the mucosal plane and nylon 7-0 for the cutaneous plane, leaving the distractor stem with submandibular level emergence. Immediate postoperative control was performed with computed tomography. The latency period was 5 days and the activation period included a protocol of 0.4 mm turns of the screw every 12 hours for 25 days. The consolidation period was 6 months. The distractor was removed at 6 months postoperatively, in the operating room under general anesthesia.

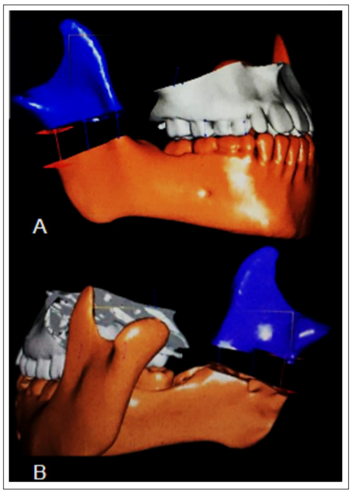

Figure 1: Image of Virtual Planning with Horizontal Osteotomy of the Right Mandibular Ramus in Lateral View, showing its external face (A) and Internal Face (B)

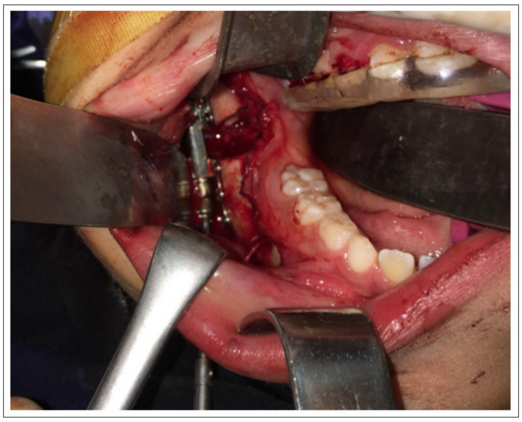

Figure 2: Intraoperative Image of the Horizontal Right Bundle Branch Osteotomy. Frontal View

In the immediate postoperative computed tomography (CT) scan (Figure 3), a discrepancy of 18.69 mm is observed in the area of the condyle affected by HFM. This value was measured as a straight line between the highest point of the mandibular condyle intersecting with the mandibular angle ("C - AM"). These points were arbitrarily defined and described in the methodology to represent the posterior border of the mandibular ramus (Figure 4).

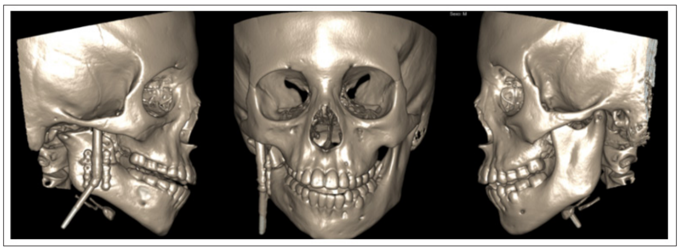

Figure 3: 3D Reconstruction of Immediate Postoperative Status following Installation of the Mandibular Ramus Distractor

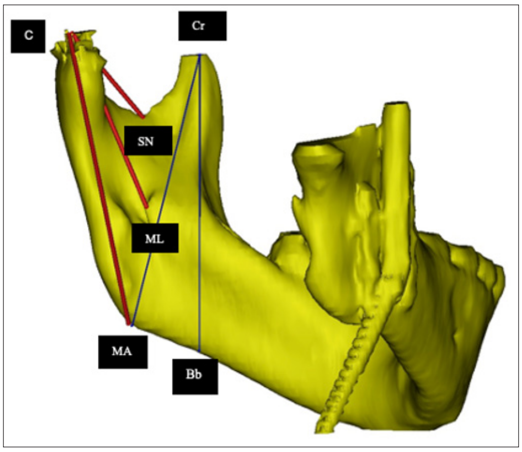

Figure 4: 3D Reconstruction of Immediate Postoperative Status. Over these Images, Measurements of Posterior Ramus Height (Distance from point C to AM), Condylar Height from SN (Distance from line C to SN), Condylar Height from ML (Distance from C to ML) and Anterior Ramus Height (Distance from Cr to Bb) are Observed

After the removal of the distractor, the patient underwent serial clinical and imaging controls; first at 7, 14, and 21 days, then at the first, third, sixth, and eighth postoperative months. The patient progressed favorably from a surgical standpoint, without complications, achieving adequate facial symmetry, functional mandibular dynamics, and stable occlusion. As mentioned, the distractor was removed at 6 months postoperatively, and a new control CT scan was performed at 8 months postoperatively (Figure 5).

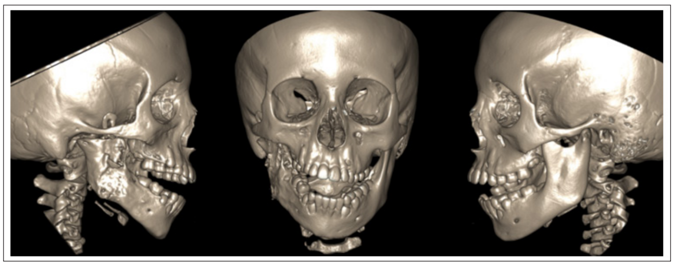

Figure 5: 3D Reconstruction after 8 Months of Mandibular Distraction and Distractor Removal

Methods

A table was designed to present the results of the measurements through the 3D images of the CT scan, obtained in the immediate postoperative period prior to the initiation of distraction and at 8 months postoperative, along with a comparative table highlighting the differences in measurements.

The 3D images were obtained using helical CT scans with minimum slice thickness of 0.1 mm, and the software used for analysis was RadiAnt DICOM Viewer 2022.1 (64-bit).

To perform the measurements of the mandibular ramus, the protocol proposed by Fariña et al. was followed [15]. The cephalometric points and measurements obtained were as follows:

- **Point 1: Mandibular Condyle (C):** The highest point of the convexity of the mandibular condyle.
- **Point 2: Sigmoid Notch (SN):** The lowest point of the concavity of the sigmoid notch.

- **Point 3: Mandibular Lingula (ML):** The base of the mandibular lingula in relation to the mandibular foramen.
- **Point 4: Mandibular Angle (AM):** Bisector formed by an angle of the tangent to the parotid edge and the tangent to the basilar edge.

With these four identified points, a connecting plane is drawn from point 1 to point 4, determining the length of the mandibular ramus. Then, three lines perpendicular to this plane are drawn, passing through points 1, 2, and 3, which are lines A, B, and C, respectively, for both the healthy and affected sides [15].

To this protocol, the following measurement is added: from Coronoid to Basilar Border, defined as "Cr" and "Bb" respectively; this will allow us to measure the anterior border of both mandibular ramus and thus compare them with the measurements obtained at the posterior border (Figure 3).

The measurement from the immediate postoperative period was used to obtain more reliable data, as preoperative measurements may undergo slight changes due to growth and remodeling phenomena between the preoperative period and surgery.

Results

When comparing measurements between the different cephalometric points mentioned bilaterally, it becomes evident that there is a vertical increase in the affected mandibular ramus, with a gain of 12.55 mm in (C - AM), corresponding to the posterior border of the mandibular ramus, while a discrepancy of 5.17 mm persists compared to the contralateral side.

The anterior border of the ramus, measured from "Cr-Bb" at 8 months postoperatively, was 57.39 mm, with a gain of 15.7 mm compared to the initial situation of the ramus with HFM. When compared to the contralateral ramus, which measures 65.15 mm, there is a difference of 7.76 mm.

Finally, comparing the vertical gain between the anterior and posterior borders of the ramus with HFM undergoing distraction, a difference of 3.15 mm is observed in favor of "Cr-Bb," corresponding to the anterior border of the mandibular ramus.

Table 1: Comparison of Measurements Obtained in the Immediate Postoperative Period, in the Length of the Osteotomized Mandibular Ramus Prior to Distraction Movements, Compared to its Healthy Contralateral Side

Distance	Right Ramus with osteotomy without activation of the DO (mm),	Left/Healthy Ramus (mm)	Initial Difference between both ramus (mm)
C - AM	44.06	62.75	18.69
C - SN	12.51	21.74	9.23
C - ML	27.91	37.99	10.08
Cr - AM	40.44	61.16	20.72
Cr - Bb	41.69	63.94	22.25

C: Condyle, AM: Mandibular Angle, SN: Sigmoid Notch, ML: Mandibular Lingula, Cr: Coronoid Process, Bb: Basilar border. C - AM: Posterior Ramus Height, C - SN: Condylar Height from SN, C - ML: Condylar Height from ML, Cr - AM: Anterior Ramus Height, Cr - Bb: Anterior Ramus Height.

C: Condyle, AM: Mandibular Angle, SN: Sigmoid Notch, ML: Mandibular Lingula, Cr: Coronoid Process, Bb: Basilar border.

Table 2: Measurements Obtained in the Right Mandibular Ramus after a 20 mm Distraction

Distance	Right Ramus/Distracted
C - AM	64.00
C - SN	31.30
C - ML	46.85
Cr - AM	59.40
Cr - Bb	61.60

C: Condyle, AM: Mandibular Angle, SN: Sigmoid Notch, ML: Mandibular Lingula, Cr: Coronoid Process, Bb: Basilar border.

Table 3: Comparison of Measurements Obtained at 8 Months Postoperative: in the Length of the Distracted Mandibular Ramus Compared to its Healthy Contralateral Side

Distance	Right Ramus/Distracted (mm)	
	Increment of mm post distraction	*Relapse*
C - AM	+ 12.55	- 07.39
C - SN	+ 11.02	- 08.77
C - ML	+ 07.60	- 12.34
Cr - AM	+ 14.64	- 05,32
Cr - Bb	+ 15,70	- 04,21

Discussion

Biological forces influencing bone morphology arise from genetic information and functional stimuli. Mechanical forces generated during DO originate from the activation of distraction devices in the osteotomized bone, their specific orientation with respect to skeletal anatomy. Therefore, during the gradual traction process in mandibular distraction, it is imperative to consider the powerful impact of both biological and mechanical force systems to anticipate their resulting effects on bone consolidation, paying special attention to the action of premature external forces, such as the use of elastics [9].

In the study published by Marquez et al., where they conducted post-osteogenic distraction follow-up, they indicated that after achieving a prior overcorrection of the mandibular dental midline by 3 mm, after two years of follow-up, the midline had deviated towards the affected side. Additionally, the height of the mandibular ramus had decreased by 13 mm compared to the length achieved during distraction, progressively shortening. The authors attribute this long-term effect on mandibular ramus height to the action of the masticatory muscles. Thus, they suggest that early facial skeleton reconstruction will fail or recur due to the inability to reconstruct the muscle-periosteal component of growth force, following the functional matrix theory. In the article, they also cite a study by Harvold who reports that the success of any treatment to elongate the jaw in patients with MHF will depend on the development of the masticatory musculature, especially the masseters and internal pterygoids, and dental occlusion [14]. Therefore, they suggest early treatment during growth to achieve adequate and stable soft tissue expansion, explaining that the mandibular ramus would decrease in size to adapt to the existing soft tissue matrix, such that distraction osteogenesis might not be sufficient to improve the soft tissue deficiency needed to guide growth, with the eventual need for a subsequent second distraction.

They also consider the relevance of serial cephalometric controls to determine and document skeletal changes associated with relapse early on. This is consistent with the findings observed in our clinical case, where rapid relapse and loss of gained height were observed, but with greater gain in the bony areas with less force from the masticatory musculature.

In a study published by Meazzini et al., where they evaluated a 5-year follow-up in 8 patients with MHF treated with unilateral extraoral distraction osteogenesis (Pruzansky I and II), they observed a gradual relapse to asymmetry in the vertical direction. There was an average loss of 77% of the vertical correction obtained at the end of the follow-up period, but these values could vary depending on the patient's age and stage of growth [16]. One of the theories that could explain the relapse, referred to by Simpsons et al., is that osteogenesis requires a greater elongation rate than myofibrillogenesis, and thus ideal muscular adaptation may not be achieved during mandibular distraction [11]. The article also mentions that, although the short-term changes achieved with unilateral mandibular ramus distraction osteogenesis are satisfactory in these patients, there may be a genetic component associated with both neuromuscular and bony aspects, which could cause changes in the distracted bone, leading it back to its previous configuration. Additionally, the healthy mandibular ramus continues to grow, guiding the maxilla in the process, thus exacerbating the condition of the affected contralateral side due to MHF even further. In the presented case, the initial gain in bone height is greater at the anterior border of the ramus than at the posterior, which could be explained by the direction of muscle fiber traction and its biomechanical vectors in an anterosuperior oblique direction, but eventually showed relapse. In this sense, we could consider the existing clinical limitations to define the level of overcorrection to use in each patient and the stage of growth they are in, which could be associated with rapid relapse, as was the case with our patient.

In a study published by Chow et al., where they evaluated the stability over 5 years of distraction osteogenesis in terms of maxillary width/height, occlusal height, and mandibular ramus height in four patients with MHF, aged 7 to 11 years, treated with unilateral intraoral mandibular distraction, they concluded that due to the greater growth potential of the healthy side, significant overcorrection is needed to compensate for persistent asymmetry in growing patients and relapse. Their conclusions were based on the following findings: The distracted side mandibular growth remained stable and at the same level as the control side up to two years post-distraction and reached its greatest discrepancy at 5 years of follow-up, which coincides with the conclusions of Tehranchi and Behnia referred to in the same article, and this could be explained by mandibular rotation around the healthy side during unilateral distraction and the eventual lower growth potential of the affected side), is that osteogenesis requires a greater elongation rate than myofibrillogenesis [13]. However, they mention that it is not possible to quantify the amount of overcorrection to be indicated in this study, so further research with larger samples is needed, which should be compared according to age and severity of the condition. Although in our case, proportional overcorrection was performed, there may be additional factors not fully understood that influence the higher or lower relapse rate.

Another factor that we could consider associated with relapse after DO is mentioned by Datarkar et al., where they evaluated the newly formed bone using 3D computed tomography using Hounsfield units, determining that the bone formed by distraction osteogenesis was satisfactory but less mineralized, with a less dense trabecular pattern compared to undistracted bone [17]. This could be a useful, simple, and non-invasive tool to use to help decide on consolidation times and thus prevent relapses in these cases that have a less predictable evolution. However, the cost/benefit associated with serial monitoring would need to be considered.

The placement of the device can be described as vertical, horizontal, or oblique. It is important to note that the position of the device is best described in relation to the longitudinal axis of the mandibular body. Vertical placement of the device results in an increase in the vertical dimension of the mandibular ramus. During activation, a change in the orientation of the apparatus occurs, which appears to be caused by the nonlinear molding effect of the neuromuscular system on the regenerated bone as it forms. The mandible automatically rotates clockwise; thus, an anterior and posterior open bite can occur on the side that has undergone vertical distraction in the mandibular ramus, but this can be managed with molar stops that gradually wear down to allow the maxillary dentoalveolar segment to follow the mechanical growth achieved with the mandible.

In the literature search, the following study was considered, in which the dimensions of bone and soft tissues, as well as volumetric changes in the chewing muscles, were measured after mandibular distraction osteogenesis in patients with MHF. In the 8 patients in the study, there was an increase in total muscle volume after distraction, with a greater increase in volume on the affected side of the jaw compared to the healthy side. However, one-year follow-up records showed relapse on the affected side of the jaw in five out of eight cases [4]. These results could be related to the lack of stability and activity achieved by the masticatory muscles after distraction osteogenesis, which could limit subsequent bone formation, suggesting a determinant role of the posterosuperior oblique vector in distraction cases, especially associated with the mandibular ramus. Although soft tissue images were not studied in our case, it is an interesting complementary alternative to consider for monitoring these patients. However, to establish conclusive protocols, serial electromyographic studies should be conducted to explain the actual muscular behavior.

Despite the histogenesis induced by distraction osteogenesis (DO), soft tissue containment around the distracted bone, along with scarring from previous procedures, can lead to relapse, with values varying depending on the studies and the type of osteodistraction performed, but can be close to 10%, depending on the amount of movement [9]. Some authors, such as Duocet et al., even suggest larger overcorrections, up to 20% in growing patients, based on their experience in a retrospective study with a 4-year follow-up in maxillary DO [18]. We agree with these statements and suggest conducting a detailed study of each patient to determine the level of overcorrection to apply, considering the vector of growth guided by distraction and its relationship with the vector of muscular forces, especially the pterygomasseteric sling.

Conclusion

In the literature, one of the possible causes of relapse after distraction osteogenesis is described to be the force vectors of the masticatory muscles. While many studies refer to this cause, few are backed with tangible data, and these may be insufficient to be conclusive or have insufficient sample sizes. Additionally, none of the mentioned studies refer to the quantified effect of the pterygomasseteric sling on bone gain and relapses in mandibular ramus height achieved after distraction osteogenesis. Therefore, it is suggested to conduct studies in which the role of the pterygomasseteric sling in vertical distraction osteogenesis of the mandibular ramus in patients with Goldenhar Syndrome can be quantified and compared through electromyographic records. This would establish working algorithms to determine the degree of overcorrection to be performed according to the type of distraction, the patient's growth stage, and the severity of the pathology, along with the standardization of follow-up protocols for each case.

The proportional overcorrection of mandibular ramus with MHF that we propose based on the results obtained could correspond to approximately 40%, rather than the 10% described as post-distraction osteogenic relapse in the maxilla. Despite the aforementioned conclusion, we must consider that there are multiple surrounding tissues, different genetic, anatomical, and functional characteristics in each patient, and an occlusion that can modify the results in all three spatial dimensions. Therefore, in many of these cases, a single surgery is insufficient, requiring multiple procedures with long-term follow-up.

The orientation of the pterygomasseteric sling in Angle Class I patients tends to be more vertical than the orientation of this muscular complex in Angle Class II patients with posterior rotational growth and marked pregonial notches. Malformations such as MHF are usually associated with skeletal Class II, so the results presented correspond to this maxillomandibular pattern and facial architecture. It would be interesting to draw conclusions about the changes achieved after DO in patients who do not have this maxillomandibular pattern.

According to the presented case, we believe that the oblique vector of forces generated by the muscular action of the pterygomasseteric sling plays a crucial role in the pattern of bone growth in vertical mandibular distraction osteogenesis (DO), considering a greater initial elongation of the height at the anterior border of the mandibular ramus compared to the posterior or parotid border, where the action of the mentioned musculature and its synergistic action with the temporal muscle is more predominant. However, it is also important to consider that there is a significant rate of relapse over time in the mandibular ramus and the factors that may be associated with it. Therefore, it is necessary to develop these types of studies to anticipate the growth pattern and potential complications or relapses in osteogenic distraction treatments.

Ethics: The use of the clinical case with their respective images is supported by the informed consent signed by the patient**.**

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