

## Multidisciplinary orthopedic treatment of facial asymmetry in a pediatric patient: case report

### Tratamento ortopédico multidisciplinar da assimetria facial em paciente pediátrico: relato de caso

Sara Yrma Piccinin Villarroe<sup>ID\*</sup>, Juliana Marques Lindenberg<sup>ID</sup>, Aline Nardelli<sup>ID</sup>, Mauro Carlos Agner Busato<sup>ID</sup>, Carlos Augusto Nassar<sup>ID</sup>

#### ABSTRACT

Temporomandibular joint ankylosis (TMJA) may be fibrous or bony (resulting from the fusion of articular surfaces, glenoid fossa, and mandibular condyle). A multidisciplinary approach is necessary to successfully rehabilitate patients with this condition since it can cause problems related to psychosocial, nutritional, facial, and occlusal factors. Depending on the patient's age, orthotic intervention may be necessary in order to correct the malocclusion caused by TMJA. This article presents the case of a pediatric patient with idiopathic left TMJA who underwent multidisciplinary treatment at a reference center for craniofacial anomalies in western Parana. Balters Bionator orthopedic appliances were initially used, followed by rapid maxillary expansion using a Hyrax expander. This resulted in improved masticatory, esthetic, and speech functions, particularly mouth opening, as well as partial correction of mandibular asymmetry and the occlusal plane within the limits of the case. The use of functional and mechanical orthopedic appliances contributed to improved facial function and esthetics, and the multidisciplinary team's care was essential to improving the patient's quality of life during the rehabilitation process.

**Keywords:** Ankylosis. Orthotic devices. Temporomandibular joint.

#### RESUMO

A Anquilose da Articulação Temporomandibular (AATM) pode ser do tipo óssea pela fusão das superfícies articulares, cavidade glenoide e côndilo mandibular e do tipo fibrosa. Essa patologia pode gerar comprometimentos psicossociais, de nutrição, faciais e oclusais, sendo o tratamento multiprofissional a melhor terapêutica para reabilitação desses pacientes. A intervenção ortopédica é fundamental para correção de má oclusão causada pela AATM, dependendo da idade do paciente. O presente artigo objetiva relatar o caso de um paciente pediátrico com AATM esquerda idiopática que está realizando tratamento multidisciplinar em um centro de referência em tratamento de anomalias craniofaciais localizado no oeste do Paraná. Inicialmente, utilizou-se o aparelho ortopédico Bionator de Balters, seguido da expansão rápida da maxila com uso do Expansor Hyrax, permitindo ao paciente melhora na função mastigatória, estética, fonação e, principalmente, da abertura bucal com a correção parcial do plano oclusal e da assimetria mandibular dentro dos limites impostos pelo caso. O uso de dispositivos ortopédicos funcionais e mecânicos contribuíram para melhora da função e estética da face e o atendimento por uma equipe multiprofissional se mostrou essencial, disponibilizando oportunidade de obtenção de melhor qualidade de vida durante o processo de reabilitação.

**Palavras-chave:** Anquilose. Aparelhos ortopédicos. Articulação temporomandibular.



## INTRODUCTION

The temporomandibular joint (TMJ) is anatomically classified as a ginglymoarthroidal type, being bilateral and interdependent (Pereira, Carvalho, Garcia & Cavalcanti, 2011). It consists of a portion of the mandible (mandibular condyle) and the base of the skull (glenoid fossa of the temporal bone), both articular surfaces isolated by the articular disk, fibrocartilaginous structure that also divides the TMJ into a virtual superior and inferior space (Vieira & Rabelo, 2008; Pereira et al., 2011).

Temporomandibular joint ankylosis (TMJA) is characterized by the fusion of the articular surfaces of the temporomandibular joint and can be of the bony and fibrous type (Ataç, Çakir, Yücel, Gazioğlu & Akkaya, 2014). Its causes can be diverse, from the traumatic origin, which is the most frequent, systemic diseases, local and systemic infections, birth trauma due to the use of forceps during birth, idiopathic, and also congenital origin (Ajike et al., 2006; Vasconcelos, Porto & Nogueira, 2008; Vieira & Rabelo, 2008; Pereira et al., 2011; Santos, Araujo, Cavalieri, Vale & Canellas, 2011; Vibhute, Bhola & Borle, 2011; Ataç et al., 2014; Limongi, Manzi & Limongi, 2019).

The TMJA can occur at different ages, being more common in children under ten years, and may occur on one side of the face or bilaterally (Pereira et al., 2011). This condition can cause facial asymmetry, which, in unilateral TMJA, is a striking feature, along with deviation of the chin to the affected side, resulting in compromised facial harmony (Vieira & Rabelo, 2008).

Other possible adverse effects are psychosocial or nutritional disorders, and it is common for these patients to opt for a liquid-paste diet, in addition to a higher incidence of caries due to the difficulty in performing satisfactory oral hygiene (Vieira & Rabelo, 2008). Malocclusions are another problem resulting from this pathology and are caused by the atypical growth of the mandible (Vasconcelos et al., 2008; Vieira & Rabelo, 2008; Vibhute et al., 2011; Ataç et al., 2014).

The multi-professional work in rehabilitating these patients is the best possible therapy, with oral and maxillofacial surgeons, physical therapists, orthodontists, psychologists, and speech and hearing therapists (Vibhute et al., 2011), among others. Due to the high relapse rate of this pathology, there is still no consensus about the best surgical technique; however, immediate postoperative physiotherapy seems to bring positive results (Maia, Ravali, Almeida & Gallo, 2008; Vieira & Rabelo, 2008; Vibhute et al., 2011; Marcolino, Gomes & Silva, 2014).

The orthopedic treatment is essential for correcting malocclusion and depends on the patient's age and commitment to the treatment, which are fundamental for the success of the result (Marcolino et al., 2014). The Balters Bionator functional orthopedic appliance is indicated for correcting Angle Class II division 1 malocclusion in the active growth phase (Matos, Vieira,

Tomé & Carvalho, 2016). With the same indication of age range due to its orthopedic nature, in cases in which the maxilla is atrophic, it is indicated to perform the maxillary expansion through circuit-breakers that will separate the midpalatal suture (Fabrini, Gonçalves & Dalmagro, 2006; Scanavini, Reis, Simões & Gonçalves, 2006).

This manuscript aims to report the case of a patient with left TMJA with Angle Class II and maxillary atresia. The patient is undergoing multidisciplinary treatment in a reference center for craniofacial anomalies in western Parana, emphasizing orthodontic treatment.

## CASE REPORT

### Diagnosis

This study complies with the criteria of resolution 466/12 of the National Health Council, with authorization requested through the Free and Informed Consent Form (TCLE), approved by the Ethics and Research Committee under opinion n.º 4.250.143.

The patient is a 14-year-old male subject, leucoderma, under treatment at the Center for Attention and Research in Craniofacial Anomalies (CEAPAC) of the University Hospital of Western Parana (HUOP) in the city of Cascavel, Parana, diagnosed with left TMJA of idiopathic origin. He came to the HUOP service at six years of age, presenting with asymmetrical face, micrognathia, limited movement to the contralateral side, deviation of the chin to the left, and reduced mouth opening. The patient's parent reported that he was diagnosed with "impaired TMJ function" at one-year-old, and since then has been on a liquid diet due to chewing difficulties. There were no reports of forceps injury during delivery, no reports of trauma during childhood, or local or systemic infections, suggesting that the origin of TMJA is idiopathic.

In a genetic evaluation of the patient, besides the left TMJA, low and hyperdeveloped left ear implantation, reduction of the index finger phalanx (right and left) and middle finger (left) due to agenesis and hypogenesis of the distal phalanges, hypogenesis of the middle phalanx of the right index finger, and word articulation disorder (dyslalia) were observed. These minor malformations keep the patient under follow-up and investigation for syndromic features.

At six years of age, the patient underwent arthroplasty of the left mandibular condyle to reconstruct the mandibular condyle. Six months after the procedure, he continued to have difficulty in opening his mouth, and new imaging exams (Figure 1) showed a neoformation of an ankylotic mass in the left TMJ region. The patient continued to be monitored by the team that performed the surgery and the multi-professional team with physical therapy, speech therapy, psychology, social work, dentistry, nursing, genetics, medicine, and nutrition, with maintenance of hypersalivation and feeding difficulties.

**Figure 1**

Initial panoramic radiograph exposing bone degeneration in the left mandibular condyle region.



Source: The authors.

At eight years of age, he presented a maximum interincisive distance of ten mm in the mouth opening, and the oral maxillofacial surgery team decided to plan a new surgical procedure for resection of the left condyle and reconstruction of with a costochondral graft from the right rib, obtaining 15 mm of maximum interincisive distance in the 11-month postoperative period. After the surgery, the physiotherapy team frequently attended to the patient.

Following the second surgery, the intraoral evaluation performed by the orthodontist found that the patient had an inclined occlusal plane (cant), Angle Class II, Division 1, subdivision left, as well as right posterior deep overbite and complete transposition of tooth 32 concerning tooth 33 (Figures 2 and 3). In the lateral cephalometric radiograph, lower incisors were proclined buccally and protruded (Figure 4 and Table 1).

**Figure 2**

Initial extraoral photographs.



Source: The authors.

**Figure 3**

Initial intraoral photographs.



Source: The authors.

**Table 1**

Comparison of initial and normal standards cephalometric measurements of orthodontic treatment.

Cephalometric variables	Initial	Normal Standards
SNA	74.95	82.00
SNB	64.60	80.00
ANB	10.35	2.00
WITS (MM)	2.27	0-1
SN.GOGN	51.63	32.00
FMA	42.83	25.00
1.NA	21.00	22.00
1-NA (MM)	1.86	4.00
1.NB	32.78	25.00
1-NB (MM)	6.91	4.00

Source: The authors.

**Figure 4**

Initial profile telerradiography.



Source: The authors.

**Treatment objectives**

The aim was to begin treatment with the correction of mandibular asymmetry, followed by rapid maxillary expansion.

**Treatment planning**

The starting treatment proposed was with the Balters Bionator orthopedic appliance for correction of the vertical asymmetry of the lower third of the face for rapid expansion of the maxilla with the use of the Hyrax Expander and extraction of tooth 32.

**Treatment progress**

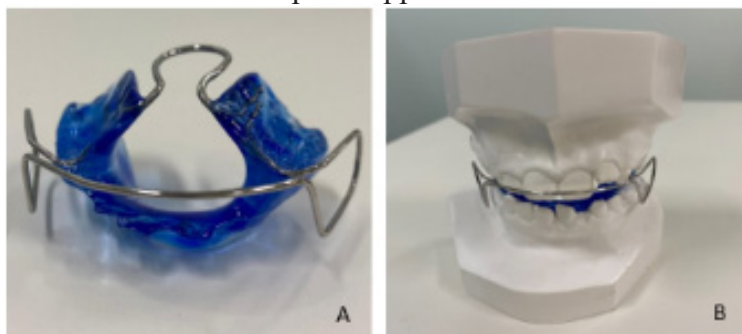
The Balters Bionator orthopedic appliance (Figure 5, A and B) was chosen due to the right overbite resulting from abnormal growth of the mandibular condyle on the opposite side of the face at the patient's tenth birthday years old age. In the follow-up visits, the Balters Bionator was worn on the acrylic in the upper left occlusal region in order to stimulate the vertical growth of the upper left alveolar process intending to correct the transverse occlusal plane unevenness.

With excellent cooperation, the patient wore the Balters Bionator appliance for nine months. It is noteworthy

that the choice for starting orthodontic treatment with this device was only to anticipate mandibular growth (growth mortgage), improving asymmetry, even aware that treatment with this appliance in the pre-surge period of growth could be less efficient (Saad, 2012).

### Figure 5

Balters Bionator orthopedic appliance.



Source: The authors.

Note. A: in frontal view. B: in occlusion with the patient's study model.

Another critical factor in the decision to use this orthodontic appliance in this period was that cited by Álvarez et al. (2011), since the use of removable functional orthopedic appliances in patients presenting mandibular retrognathism and predominantly horizontal growth patterns showed great results when used before the growth spurt (Álvarez et al., 2011). Moreover, the patient's guardian was informed that the treatment performed before the ideal period would have limitations related to stability when compared to this same therapy performed during the pubertal growth spurt (Saad, 2012; Cheong, Kassam, Eccles & Hensher, 2016).

Following the discontinuation of the Balters Bionator, the Hyrax expander appliance was installed for maxillary expansion. This Rapid Maxillary expansion was performed two times: initially, with banding of the deciduous maxillary second molars and, after a one-year interval, with banding of the permanent maxillary first molars. The interval between expansions was precisely to wait for permanent teeth eruption for the appliance's anchorage. During this period, tooth 32 was also extracted, as it was transposed in relation to tooth 33 and had no space in the lower arch.

At the age of 14, when the patient was undergoing corrective orthodontic treatment, pain in the left TMJ region was reported during a follow-up consultation with the oral and maxillofacial team. After clinical examination, a decrease in mouth opening was observed, and a new imaging exam was requested for evaluation and planning of a new approach. An essential factor that aggravated the limited mouth-opening treatment in this period was the SARS-CoV-2 pandemic, which forced services to be suspended for a few months, and the patient did not continue with physical therapy and orthodontic care.

The updated imaging exam showed resorption of the costochondral graft in the left TMJ and the absence

of the coronary process on the same side of the face, besides noting the prominence of the coronoid process on the right side (Figure 6). Given this, at the time this paper was written, a new surgery was being planned to maintain the patient's condition, so the need to approach the contralateral coronoid process and study a possible pseudoarthrosis in the left condylar region was discussed by the surgical team.

### Figure 6

Updated magnetic resonance.



Source: The authors.

Following what was reported, the orthodontic team decided to interrupt the patient's orthodontic treatment due to the likely need for a new surgical intervention to be planned. Another decisive factor that led to the pause in corrective orthodontic treatment after eight months was the patient's severe pain during appointments, even though he maintained a minimal mouth opening to perform maintenance procedures, in addition to the fact that he was clenching his teeth day and night. Therefore, this approach was chosen to improve the patient's quality of life until the new surgical intervention (Figures 7, 8, 9, 10 and Table 2).

### Figure 7

Final panoramic radiograph.



Source: The authors.

**Figure 8**

Extraoral photographs after removal of the conventional fixed appliance.



Source: The authors.

**Figure 9**

Intraoral photographs after removal of conventional fixed appliance.



Source: The authors.

**Figure 10**

Final lateral telerradiography.



Source: The authors.

**Table 2**

Comparison of initial and final cephalometric measurements of orthodontic treatment.

Cephalometric variables	Initial	Final
SNA	74.95	76.59
SNB	64.60	71.77
ANB	10.35	4.82
WITS (MM)	2.27	-1.95
SN.GOGN	51.63	44.10
FMA	42.83	36.05
1.NA	21.00	18.69
1-NA (MM)	1.86	3.01
1.NB	32.78	23.23
1-NB (MM)	6.91	5.76

Source: The authors.

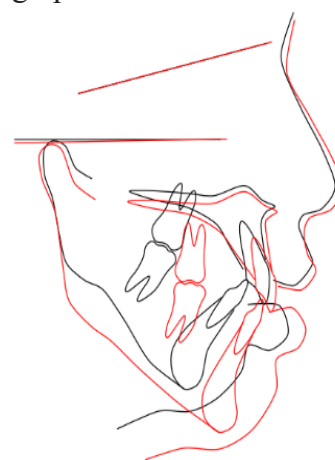
**DISCUSSION**

The treatment of TMJA is challenging due to the high possibility of relapse and non-consensual surgical protocols in the literature (Mendoza, Costa & Freitas, 2011; Genaro, Passos, Felix & Trindade, 2013). The multidisciplinary treatment is essential, in accordance with what Vibhute et al. (2011) described in their study, in which the work of immediate postoperative physiotherapy is unique in an attempt to maintain the mouth opening obtained with surgery, avoiding the loss and consequent relapse of the case (Vibhute et al., 2011).

Using the Bionator before expanding the maxilla allowed the patient to improve chewing function since the misalignment of the occlusal plane was considered the initial priority for resolution together with mandibular retrognathism to improve esthetics, speech, and, especially, mouth opening, since the patient had not yet reached pubertal growth, the age of indication for this device (Saad, 2012). The Balters Bionator causes the stimulation of bone growth in the mandibular body and ramus (Matos et al., 2016), which were crucial for the stage of treatment in which the patient found himself since the left mandibular ramus had delayed bone growth (Figure 11).

**Figure 11**

Superimposition of the initial and final telerradiographs.



Source: The authors.

Rapid maxillary expansion using the Hyrax expander resulted in the solution of transverse maxillary atresia. This tooth-supported orthodontic device opens the midpalatal suture through the activation of the expander screw (Scanavini et al., 2006). According to Fabrini et al. (2006), palatal expansion increases the arch's perimeter by restoring the maxilla's transverse dimensions (Fabrini et al., 2006).

Similar to the Balters Bionator, the Hyrax expander is an orthopedic appliance; appliance, thus there is an age restriction of the patient at which the orthodontist can use these orthodontic devices. In the case of the Hyrax expander, using this device after the active growth phase to perform the rupture of the midpalatal suture would

require a considerably high force, resulting in pain complaints and other harm to the patient (Scanavini et al., 2006). As previously described, the age at which the patient used the Hyrax expander was ideal (Scanavini et al., 2006), and the patient's cooperation was crucial to the result.

The literature shows that TMJA is more common in children under ten, but it can also occur at any age (Pereira et al., 2011), with the reported case being compatible with this more frequent average life stage. Still on AATM, the present case is supported by another recurrent issue reported in the literature, which is the high relapse rate (Shamia, James & Adekeye, 1977).

The autogenous costochondral graft is obtained from the patient's rib, thus being biologically compatible, and composed of cartilaginous and bone tissue (Vieira & Rabelo, 2008; Álvares et al., 2011; Vibhute et al., 2011). Another benefit of this type of graft is the ability to transfer a bone growth center, as Mendonça et al. (2011) elucidated, thus being the material of choice in children and youth patients (Freitas et al., 2011; Mendonça et al., 2011).

The pandemic period caused by SARS-CoV-2, when the patient was 13 years old, resulted in months of interrupted visits at CEAPAC because they were elective, and the patient remained without assistance from the physical therapy team with mandibular opening exercises that were essential to maintain the patient's condition, which could be a significant cause for the recent relapse and increase in pain complaints, as well as a decrease in mouth opening. According to Shamia et al. (1977), patients with congenital ankylosis who did not

perform jaw-opening exercises had higher relapse rates.

## CONCLUSION

This case report described a patient with left TMJA and also with facial asymmetry who needed to undergo two surgical procedures due to relapse, being arthroplasty of the left mandibular condyle and resection of the left condyle with costochondral graft. In combination of orthodontic treatment with the use of orthopedic appliances (Balters Bionator and Hyrax expander) in order to improve the functional condition, esthetic, and mouth-opening. Finally, multidisciplinary care was essential, providing the opportunity for obtain better quality of life.

## COMPETING INTERESTS

The authors declare that there are no conflicts of interest.

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## AUTHOR CONTRIBUTIONS

*Conceptualization:* S. Y. P. V. e M. C. A. B. *Data curation:* M. C. A. B. e C. A. N. *Formal analysis:* S. Y. P. V. e M. C. A. B. *Investigation:* S. Y. P. V., J. M. L. e A. N. *Methodology:* S. Y. P. V. e M. C. A. B. *Project administration:* S. Y. P. V. e M. C. A. B. *Resources:* S. Y. P. V. *Supervision:* M. C. A. B. e C. A. N. *Validation:* S. Y. P. V. e M. C. A. B. *Visualization:* S. Y. P. V. *Writing - original draft preparation:* S. Y. P. V. *Writing - review and editing:* S. Y. P. V., M. C. A. B. e C. A. N.

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