J Oral Med Oral Surg 2024;30:16
https://doi.org/10.1051/mbcb/2024019

https://www.jomos.org

JOMOS
Journal of Oral Medicine and Oral Surgery

Case Report

Maxillary anomalies in choanal atresia: a case report

Marie Merheb*, Jean François Devoti, Etienne Simon, Muriel Brix

Maxillo-facial Department, Regional University Hospital, 29 av. Maréchal de Lattre de Tassigny, 54035 Nancy Cedex, France

(Received: 20 November 2023, accepted: 24 June 2024)

Keywords:
Choanal atresia / orthognathic surgery / solitary median maxillary central incisor syndrome / nasal obstruction / case report

Abstract – Choanal atresia is defined as the complete obstruction of the posterior nasal airway. The obstruction is the origin of maxillary impairment in the development. We describe a case of a man with unilateral choanal atresia who was referred to the maxilla-facial surgery department for his maxillary anomalies. The patient suffered from hypoplasia of the midface with class III malocclusion. and retromaxillia. He had also solitary median maxillary central incisor syndrome (SMMCI). Choanal atresia leads to nasal obstruction resulting in mouth breathing. The maxillary consequences are long face, contraction of the upper dental arch and high arched palate. The patients present class III malocclusion with posterior cross bite, anterior open bite. SMMCI may also be present. The maxillary anomalies would be only slightly due to the presence of chronic nasal obstruction. The mouth breathing, which results from the nasal obstruction, is an etiological factor at the origin of maxillary anomalies.

Introduction

Choanal atresia (CA) is a rare entity that affects one birth in eleven thousand, characterized by complete blockage between the nasal cavity and nasopharynx [1]. The atresia plates can be bone, membranous or mixed which is most frequent form [2].

The clinical presentation ranging from acute airway obstruction in bilateral forms to chronic nasal obstruction in unilateral cases [3]. Bilateral CA required emergency care because neonates are obligate nasal breathers the first months of life [3,4]. Then once the newborn is stabilized the etiologic treatment is surgical. In unilateral CA the surgery can be differed because often patients have no specific respiratory symptoms causing late diagnosis [3,4].

CA is a critical event in proper craniofacial development, is associated with maxillary anomalies of development with hypoplasia of the midface due to ventilation disorders [5].

Maxillary anomalies associated with CA have been little studied to date. Indeed, studies often focus on anomalies associated with tonsillar hypertrophy, which is much more prevalent.

We report the case of a patient with maxillary anomalies associated with unilateral CA.

Presentation of the case

A 19-year-old man was referred to the maxillo-facial surgery department for the medical care of consequences of unilateral CA with the solitary median maxillary central incisor syndrome (SMMCI). He also presented a history of operated pyelo-ureteral junction syndrome and allergic rhinitis. On medical examination, various anomalies were identified, firstly the patient had hypoplasia of the midface with retromaxillia. He had a class III malocclusion. Secondly, he had a median central maxillary incisor. And then he was suffering from inspiratory difficulties due to septo- turbinate synechiae.

The preoperative assessment included several photos, alginate dental impression of the maxilla and mandible, panoramic and cephalometric radiographs (Fig. 1).

The patient first needed orthodontic treatment to lateralize the single central incisor on the dental arch and to prepare the arches before the orthognathic surgery. The orthodontist lateralized the tooth in position 11 and added a prosthetic tooth to the arch to fill the space (Fig. 1b).

The surgical management was performed under general anesthesia. It included several steps to correct hypoplasia of the midface. First, maxillary intrasulcular incision was made and a flap is lifted to allow the elevation of nasal mucosa, the piriform apertures were exposed allowing an expansion plasty. In fact, the width of the two piriform apertures was one and half of one common size. The enlargement was made with a round bur. Reduction of the anterior nasal spine was performed at the same time.

Then, Lefort I osteotomy with a piezotome was performed, to allow a maxillary advancement of seven millimeters to correct the occlusion. Four osteosynthesis plates were placed: two close to the piriform apertures and two others on zygomatic archs.

The last surgical step was the treatment of the SMMCI, an autologous bone graft across the 21 was performed. The bone was elevated from the vestibular surface of the right

* Correspondence: marie.merheb@gmail.com

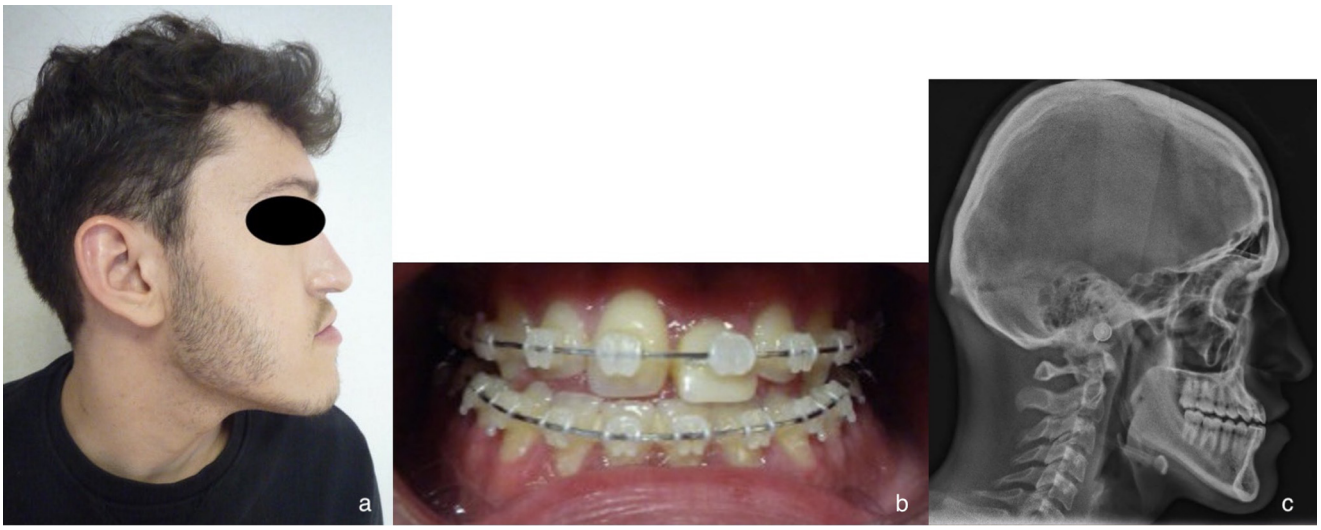

Fig. 1. Pre-surgery images of the patient with class III malocclusion and SMMCI: (a) Profile photo of the patient; (b) Intra oral photo; (c) Lateral cephalometric radiograph of the patient before surgery.

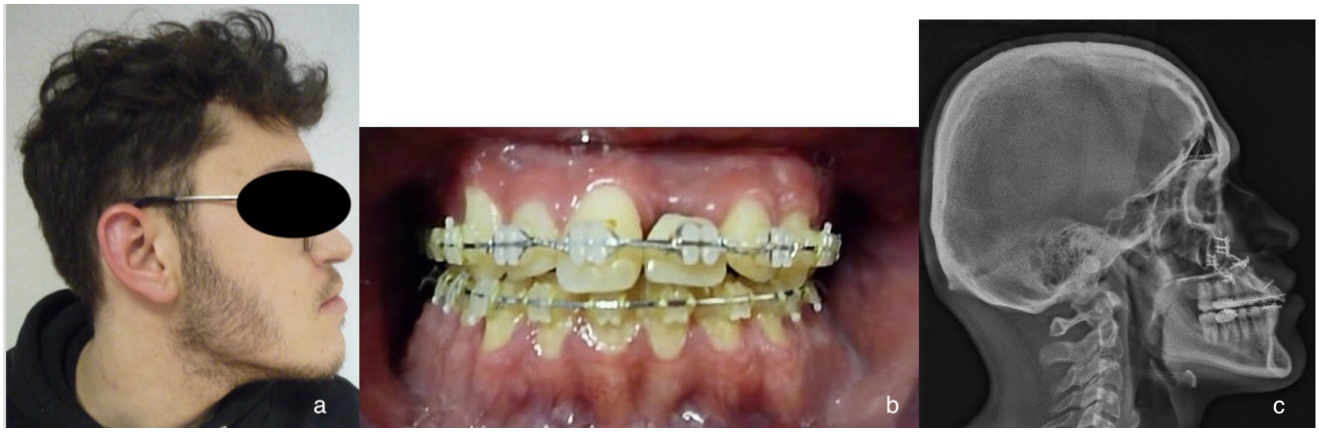

Fig. 2. Post-surgery images: the patient presents class I occlusion. (a) Profile photo of the patient; (b) Intra oral photo; (c) Lateral cephalometric radiograph at 1.5 months after the surgery.

mandibular ramus with a piezotome. The graft was completed in periphery by all the bone fragments taken during the surgery.

The prosthetic tooth couldn't be put back in place because the graft was low in crestal position.

The patient was leaving the hospital on second postoperative day with analgesics and antibiotherapy with AMOXICILLIN-CLAVULANIC ACID during eight days. Nasal washes and mouth washes were recommended during fifteen days.

The patient was met at one-week follow up and showed favorable evolution.

At one month, although the bone healing was in progress and the plates and screws were in place (Fig. 2), we noticed a slight exposure of the graft in front of the sulcus of 21 of millimetric size (Fig. 3). A light milling of the area and care with mouthwash allowed mucosal healing.

The prosthetic tooth could only be put back in place left out of the occlusion two months after the surgery to protect the mucosa and the bone graft.

At three months after the surgery, the osteotomy was consolidated, and the bone stability was favorable. He was referred to his dentist to place the dental implant of 21.

Discussion

CA is responsible of chronic upper airway obstruction. Breathing is the first function used by newborn; in addition to oxygenation function, it contributes to develop and maintain the volume of upper air cavities. However one study with seven cases of CA had noticed that six cases of CA had symmetry or more developed ipsilateral maxillary sinuses on the atresia side [6]. This suggested that nasal airflow had no influence on

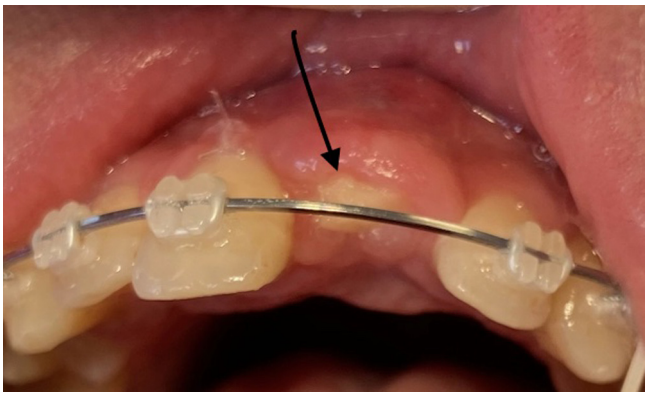

Fig. 3. Intra oral photography of the bone graft exposition.

maxillary sinus growth. A more recently study with eleven cases of unilateral CA had the same conclusion, it showed also mucosal thickness in the sinuses was like that in the control group [7]. But CA is often associated with others malformations as CHARGE syndrome (Coloboma of the eye, Heart disease, Atresia of the choana, Retarded growth, development or CNS anomalies, Genital hypoplasia, Ear anomalies and/or deafness) that may affect facial and sinuses' development [6].

In functional matrices theory by Moss and Salentijn, the nasal breathing is necessary for balanced nasal growth. When nasal obstruction is not removed quickly, oral breathing is established, this has negative effects on the development of the dentofacial complex [8]. In fact, oral breathing results in oral muscle imbalance, the children have a significant decrease in tongue pressure on the palate. This tends to have a descending position of the lingual muscles which contributes to compression of the upper dentition and narrowing of the maxillary dental arch, as well as crossbite of the posterior teeth [9].

Harvold *et al.* realized an experiment on monkeys to show the link between oral breathing and dental malocclusions. He built mouth breathing models in rhesus monkeys by obstructing the nasal passages with silicon nasal plugs. After comparing the facial appearance and occlusion of experimental and control animals, morphologic changes on the facial development have been identified some as increased face height, steeper mandibular plane, and larger gonial angle. This study highlighted that is the increased tonic activity in certain muscles and the change in jaw positioning may cause bone remodeling at the roots of maxillofacial defects [10].

In humans, the patients with oral breathing have usually a long face by excess anterior verticality [11]. Some of studies observed that once the obstacle is removed, patients maintained oral breathing by habit. The breathing mode may not always be related to airway patency. People with permeable airways breathed orally, out of habit rather than necessity [11]. Some long face subjects have been identified in early childhood while in others this feature develops during adolescence. This could be explained by adaptation for previous airway deficiencies. The adaptive posture may have resulted in altered muscle forces that can have an impact on dental and skeletal structures [11].

Regarding occlusal anomalies, children with mouth breathing can have class II or less frequently class III malocclusion. In the class II malocclusion, patients have posterior and inferior rotation of the mandible. The buccinator exerts vestibular pressure on the maxillary bicuspids and molars which do not receive sufficient support from the tongue, resulting in a defect in maxillary development and a narrowing of the palatal arch and upper dental arch [11,12]. We also find posterior crossbite, anterior open bite and increased overjet with incompetent lip seal [12].

For class III malocclusion, Rokasi and Schilli explain this by the fact of children have open jaw and a low posture of the tongue with excessive mandibular growth, with constant distraction of the mandibular condyle from the fossa which may be a growth stimulus [12].

Oral breathing can have repercussions on dental health. When air flows through the mouth, saliva evaporates and causes drying out of oral mucosa. Many studies have indicated that children with chronic mouth breathing have higher levels of streptococcus mutans and plaque responsible for higher risk of caries. Moreover the accumulation of plaque and dehydration of the gingival surface lead to gingivitis and others periodontal diseases [9].

As in our patient, the link between CA and SMMCI has only recently been studied. In fact, CA are managed surgically in neonatal period, months before the eruption of the maxillary incisor tooth. SMMCI can be isolated or be a predictive factor for holoprosencephaly. All these diseases are expressions of the same delay in, or failure of induction of, cell division in the midline on or about day 38 in utero [13].

However, it is important to note that there are no study recruiting patients with CA that have investigated the resulting maxillary anomalies. As the meta-analysis by Zhao *et al.* which included ten studies or 1353 children, shows most of studies are based on patients with adenoid hypertrophy. However, the level of obstruction may have different impact on maxillary development [14].

Conclusion

The maxillary anomalies frequently found in CA are class III malocclusion with hypodevelopment of the maxilla, narrowing of the palatal arch and upper dental arch. However maxillary growth is multifactorial. Mouth breathing, which results from the nasal obstruction present in CA, is also an etiological factor at the origin of some of the maxillary anomalies.

Funding

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

Conflicts of interest

The authors declare no conflicts of interest in regards to this article.

Data availability statement

This article has no associated data generated or analyzed.

Informed consent

Written consent was obtained.

References

1. Lesciotto KM, Heuzé Y, Jabs EW, Bernstein JM, Richtsmeier JT. Choanal atresia and craniosynostosis: development and disease. Plast Reconstr Surg 2018; 141:156–168.
2. Rajan R, Tunkel DE. Choanal atresia and other neonatal nasal anomalies. Clin Perinatol 2018; 45:751–767.
3. Kwong KM. Current Updates on Choanal Atresia. Front Pediatr. 2015. Disponible sur: http://journal.frontiersin.org/Article/10.3389/fped.2015.00052/abstract [cité 1 déc 2022]
4. Syed I, Jephson C. Choanal atresia. Br J Hosp Med Lond Engl 2005 2012; 73:687–690.
5. Kurosaka H. Choanal atresia and stenosis: Development and diseases of the nasal cavity. WIREs Dev Biol [Internet]. 2019; 8(1). Disponible sur: https://onlinelibrary.wiley.com/doi/10.1002/wdev.336 [cité 17 nov 2022].
6. dos Santos Guimarães RE, Gonçalves Becker C, Gonçalves Becker HM, Tormin Borges Crosara PF, Pena Galvão C, dos Anjos GC. Absence of nasal air flow and maxillary sinus development. Braz J Otorhinolaryngol 2007;73:161–164.
7. Terzi S, Dursun E, Çeliker FB, Çeliker M, Beyazal M, Özgür A, *et al.* The effects of choanal atresia on development of the paranasal sinuses and turbinates. Surg Radiol Anat 2017;39:1143–1147.
8. Moss ML, Salentijn L. The primary role of functional matrices in facial growth. Am J Orthod 1969;55:566–577.
9. Lin L, Zhao T, Qin D, Hua F, He H. The impact of mouth breathing on dentofacial development: a concise review. Front Public Health 2022;10:929165.
10. Harvold EP, Tomer BS, Vargervik K, Chierici G. Primate experiments on oral respiration. Am J Orthod 1981;79:359- 372.
11. Fields HW, Warren DW, Black K, Phillips CL. Relationship between vertical dentofacial morphology and respiration in adolescents. Am J Orthod Dentofacial Orthop 1991;99:147–154.
12. Grippaudo C, Paolantonio EG, Antonini G, Saulle R, La Torre G, Deli R. Association between oral habits, mouth breathing and malocclusion. Acta Otorhinolaryngol Ital 2016;36:386–394.
13. Hall RK, Bankier A, Aldred MJ, Kan K, Lucas JO, Perks AGB. Solitary median maxillary central incisor, short stature, choanal atresia/midnasal stenosis (SMMCI) syndrome. Oral Surg Oral Med Oral Pathol Oral Radiol Endodontology 1997;84:651–662.
14. Zhao Z, Zheng L, Huang X, Li C, Liu J, Hu Y. Effects of mouth breathing on facial skeletal development in children: a systematic review and meta-analysis. BMC Oral Health 2021;21:108.