Maxillary Sinus Asymmetry in Children: Report of Two Cases and Literature Review

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Abstract: Asymmetry of the Maxillary Sinus in children can be caused by various benign and malignant pathologies. Facial asymmetry is part of the clinical picture of these patients and sufficient investigation is necessary. We present a case of idiopathic asymmetry of the maxillary sinus and a case of hypoplasia of the maxillary sinus as well as a description of the main differential diagnoses.

Keywords: Facial asymmetry, maxillary sinus, silent sinus syndrome, children, differential diagnosis, maxillary hypoplasia.

INTRODUCTION

Facial asymmetry in children secondary to pathological processes involving the sinuses and surrounding structures is a challenge for pediatricians and otolaryngologists. In the evaluation of patients with maxillary asymmetry, it is important to differentiate between pathologies of the sinuses themselves such as silent sinus syndrome and pathologies of adjacent structures, such as dentition and the orbit.

CASE REPORT 1

A seven year old male was treated in the outpatient Pediatric ENT clinic of a tertiary university hospital for intermittent bilateral nasal obstruction, worse on the right, which was first noted two years ago, accompanied by progressive bulging of the right maxillary region. Sporadic purulent rhinorrhea accompanied by facial pain, snoring and nasal allergy symptoms were also reported. The patient had also been diagnosed with asthma controlled with inhaled corticosteroid treatment.

On physical examination, the patient had significant facial asymmetry due to bulging of the right maxillary region that was painful on palpation (Figures 1 and 2).

On anterior rhinoscopy, the hypertrophied inferior turbinates were grade III, with abundant purulent rhinorrhea in the right nasal cavity. Oroscopy showed a slightly ogival palate, with grade III tonsil hypertrophy. Nasofibroscopy showed purulent secretions coming from the right middle meatus and pharyngeal tonsil hypertrophy approximately 70%.



Figure 1: Facial asymmetry due to bulging of the right maxillary region.

Computed tomography of the sinuses showed mucosal thickening of the right maxillary sinus with slight bulging of the anterior wall and a densification of the soft tissues of the right malar region (Figure 3). There were no expansive processes or evidence of bone erosion. The other paranasal sinuses did not present any abnormalities.

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Figure 2: Facial asymmetry due to bulging of the right maxillary region.

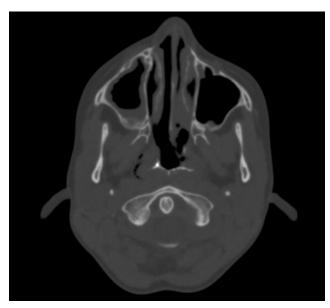


Figure 3: Thickening of the right maxillary sinus with slight bulging of the anterior wall and a densification of the soft tissues of the right malar region.

The patient was treated with topical nasal corticosteroids and nasal rinse with saline solution, and ten days of oral prednisone and amoxicillin-clavulanate. After three months of treatment, the patient had partial improvement of symptoms. He underwent adenotonsillectomy with improvement of the obstructive symptoms and rhinorrhea. A right middle antrostomy of the asymmetry of the maxillary sinus was tentatively planned for clinical control if symptomatic deterioration of the sinus dilation was detected.

CASE REPORT 2

A 13 year old male presented to the outpatient Pediatric ENT clinic of a tertiary university hospital with complaints of bilateral nasal obstruction since the age of three. This obstruction was worse on the right and was associated with the sensation of post-nasal drip,

hyposmia, nasal allergy symptoms and snoring. The patient reported typical symptoms of rhinosinusitis and was on regular antibiotics.

On physical examination, the patient had facial asymmetry with lowering of the right eyesocket (hipoglobus) and enophthalmos (Figure 4). Oroscopy presented ogival palate, dental malocclusion and grade II palatine tonsils. Rhinoscopy showed a posterior non-obstructive septal deviation to the right and signs of allergic rhinitis with the absence of secretion or lesions. Nasofibroscopy revealed a non-obstructive slight deviation of the septum to the right in area IV of Cottle, mild hypertrophy of the inferior nasal turbinate and moderate hypertrophy of the middle nasal turbinate, normotrophic mucosa, bulging of the lateral nasal wall and hypertrophy of the right pharyngeal tonsil occupying approximately 60% of the nasopharynx. The remaining ENT examination was within normal limits.



Figure 4: Facial asymmetry with lowering of the right eyesocket (hipoglobus) and enophthalmos.

Given the reported recurrent rhinosinusitis, a CT scan of the paranasal sinuses was performed which showed the following: involuted right maxillary sinus, atelectasia with infundibular obstruction, contraction of the walls of the ipsilateral sinus and considerable inferior dislocation of the orbital floor (Figures 5 and 6).

From the radiological and physicial findings as well as medical history, the patient was diagnosed with hypoplasia of the right maxillary sinus and silent sinus syndrome, in addition to mouth breathing syndrome, allergic rhinitis, and non-obstructive septal deviation.

The patient remains in regular follow-up. We await the development of symptoms and the end of the facial development to define the need for surgical intervention. Upon the initiation of therapy with topical,

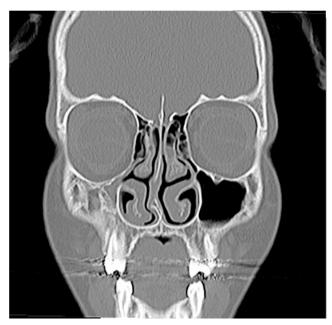


Figure 5: Atelectasia with infundibular obstruction and inferior dislocation of the orbital floor.



Figure 6: Involuted right maxillary sinus.

oral and nasal, corticosteroids, the patient reported significant improvement insymptoms.

DISCUSSION

Facial symmetry and harmony of the cervicofacial structures contribute to a nice appearance and influence the development of personality. Several structures contribute to facial harmony including dentition, the pneumatization of the nasal sinuses, and hereditary characteristics. In the field of Pediatrics,

these aspects are very important, since imperfections in facial structures may compromise the aesthetic and functional outcomes in adulthood.

Subclinical facial asymmetry is very common. In this case, skeletal disharmony can be masked by the soft tissue cover and thus a discrepancy between skeletal measurements and facial appearance may exist [1]. However, when facial asymmetry is clinically detected, either by the presence of symptoms or aesthetic nuisance, it warrants pediatric and ENT diagnostic investigation.

A systematic approach is essential in the evaluation of these patients in order to differentiate pathologies originating from the sinuses or adjacent structures such as the parotid gland, dental, and orbital structures. History and physical examination and radiographic study via computed tomography or magnetic resonance imaging should be performed in order to rule out all differential diagnoses and establish specific modalities of treatment.

Several differential diagnoses must be considered in children with facial asymmetry secondary to alterations of the maxillary sinuses.

Silent Sinus Syndrome (SSS)

SSS is a rare disorder characterized by facial asymmetry, spontaneous enophthalmos, and hipoglobus. Patients with SSS usually seek medical intervention due to facial asymmetry. The syndrome is characterized by involution of the maxillary sinus after infundibular occlusion. It has been suggested that SSS is due to hypoventilation secondary to sinus ostial obstruction and sinus atelectasis with chronic negative pressure inside the sinus [2]. Nasal endoscopy may show a characteristic pattern of unilateral collapse of the middle turbinate and uncinate process of the ethmoid [3]. Treatment is controversial. Functional endoscopic surgery of the sinuses can be used in order to establish functional drainage through the antrostomy of the compromised maxillary sinus. One should always consider conservative nasal-sinus surgical procedures in children. For aesthetic purposes, surgical reconstruction of the orbital floor for the correction of hipoglobus is also considered [4].

Maxillary Sinus Hypoplasia (MSH)

Hypoplasia of the maxillary sinus is a clinical condition found in up to 9% of patients with sino-nasal symptoms and quite uncommon in children [5]. The origin of hypoplasia is not well-known, but some hypotheses are proposed, such as abnormalities in intrauterine development, chronic infections, facial trauma, obstruction of sinus drainage, and systemic diseases such as Thallasemia and Wegener's granulomatosis [6]. Patients may be asymptomatic and present with only facial asymmetry. The diagnosis is established *via* radiological examination; computed tomography of the sinuses may show opacification of the affected sinus and lateral dislocation of the wall between the nasal cavity and the hypoplastic sinus.

Pneumosinus Dilatans (PS)

The pneumosinus dilatans corresponds to an abnormal dilation of the paranasal sinuses, which contain only air [7]. The wall of the affected sinus and its mucosa typically remain intact. Clinically, pain and swelling of the unilateral face unilateral, exofltalmia and nasal obstruction are common, however, patients may be asymptomatic [8]. The frontal sinus is the most commonly affected, followed by the ethmoid and sphenoid. The maxillary sinus is rarely affected and there are only 11 cases described in the literature. The cause of PS remains unknown, but raises the possibility of the formation of a valve mechanism that allows the trapping of air within the sinus and, therefore, positive pressure and insufflation [8]. Endoscopic sinus surgery may be a treatment option.

Tumors of the Maxillary Sinus

Few studies have reported the incidence of sinonasal tumors in children. Shrarnm et al. studied 376 children with sino-nasal masses with nasal polyp being the most frequent [9]. Less common benign lesions were hemangiomas, gliomas, dermoid and fibrous lesions. Malignant tumors are quite rare, with non-Hodakin lymphoma, chondrosarcoma rhabdomyosarcoma being the most frequent. Late diagnosis and anatomical complexity of the region contribute to poor prognosis for these neoplasias, especially malignant tumors. Sinonasal tumors remain silent until they infiltrate some cranial nerve, erode some ostial wall or obstruct ostial sinus drainage. Incipient neoplasia can cause nonspecific symptoms that simulate chronic rhinosinusitis such as nasal obstruction, anosmia, rhinorrhea and epistaxis.

The above conditions must always be considered as differential diagnoses in children with facial asymmetry. However, it is often difficult to define the

etiology of these diseases as standardized diagnostic criteria have not been established and these diseases are rare with few studies in the literature [10]. The cases presented demonstrate the difficulty of defining their etiology. In the first case, the etiologic diagnosis has not been established and in the second case, the patient has many diagnostic criteria for HSM, however, SSS cannot be ruled out. In these cases, we maintained clinical follow-up of the patients and, in case of deterioration of symptoms, a surgical approach will be considered.

CONCLUSION

Facial asymmetry in children may be associated with changes in the maxillary sinus. The clinical picture can be a challenge, presenting with non-specific nasal obstruction or change in facial aesthetics. Although the diagnosis is clinical, it is confirmed by imaging studies that show characteristic changes such as obstruction of the maxillary sinus and sinus volume loss caused by retraction of its walls.

Several diseases, benign and malignant, can affect the nasal cavity and the paranasal sinuses. Treatment must be individualized, considering the patient's complaints and the aesthetic facial deformities, being thus established the clinical and/or surgical treatment. We present two cases of asymmetrical maxillary sinus. After systematic investigation, it was concluded, for both, the diagnosis of idiopathic asymmetry of the maxillary sinus and hypoplasia of the maxillary sinus. Both cases showed improvement in their symptoms, with clinical measures and remain, to date, without the necessity of surgical approach.

REFERENCES

- [1] Allgayer S, Mezzomo FS, Polido WD, Rosenbach G, Tavares CAE. Orthodontic-surgical treatment of skeletal facial asymmetry: Case report. Dental Press J Orthod 2011; 16(6): 100-10. http://dx.doi.org/10.1590/S2176-94512011000600016
- [2] Davidson JK, Soparkar CN, Williams JB, Patrinely JR. Negative sinus pressure and normal predisease imaging in silent sinus syndrome. Arch Ophthalmol 1999; 117: 1653-4.
- [3] Cury AS, Manfrim A, Demeneghi P, Roithmann R. Silent sinus syndrome: case report and literature review. AMRIGS, Porto Alegre 2007; 51(1): 53-57.
- [4] Todd AL, Sang HH. The hypoplastic maxillary sinus and the orbital floor. Current Opinion in Otolaryngology & Head and Neck Surgery 2006; 14: 35-37. http://dx.doi.org/10.1097/01.moo.0000193167.39203.42
- [5] Karmody CS, Carter B, Vincent ME. Developmental anomalies of the maxillary sinus. Trans Ana Acad Ophthalmol Otolaryngol 1977; 84: 723-8.

- [6] Bassiouny A, Newlands WJ, Ali H, Zam Y. Maxillary sinus hypoplasia and superior orbital fissure asymmetry. Laryngoscope 1982; 92: 441-8. http://dx.doi.org/10.1288/00005537-198204000-00014
- [7] Benjamins CE. Pneumo Sinus frontalis dilatans. Acta Otolaryngologica (Stockholm) 1918; 1: 412-22. http://dx.doi.org/10.3109/00016481809122008
- [8] Juhl HJ, Buchwald C, Bollinger B. An extensive maxillary pneumosinus dilatans. Rhinology 2001; 39(4): 236-8.
- [9] Ngo H, Tewfik TL. Inverted papillomas of the nose and paranasal sinuses in children. J Otolaryngol 1987; 16(4): 244-6.
- [10] Hourany R, Aygun N, Santina CCD, Zinreich SJ. Silent Sinus Syndrome: An Acquired Condition. AJNR Am J Neuroradiol 2005; 26: 2390-92.

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