

Hypoplastic Maxillary Antra

Hesham Yousif Ali Hasan, MD, FRCS (Ed)* Shilpa Prabhu, MBBS, DLO, MRCS (ENT)**
Mohammed Ahmed Mohammed, MB ChB, FRCS***

Background: Maxillary sinus hypoplasia is an uncommon pathology of the paranasal sinuses, which might be encountered in the clinical practice. Computed tomography scan confirms its existence and any associated anatomical anomaly and variations that might coexist.

Objective: To present three cases of maxillary sinus hypoplasia which were diagnosed and managed surgically.

Design: A Retrospective Study.

Setting: Otorhinolaryngology Department, Bahrain Defence Force Hospital, Bahrain.

Method: Three cases of maxillary sinus hypoplasia were managed from January 2012 to December 2015. The clinical presentation, radiological findings and surgical management of these patients were documented and found to be distinct from other cases.

Result: The first case was a fourteen-year-old male with long-standing nasal symptoms, which were not relieved by medical treatment. Sinus CT scan revealed an ill-defined infundibular passage and complete opacified small right maxillary sinus; CT was classified as hypoplasia type 2 according to Bolger et al and was successfully treated surgically.

The second case was a twenty-seven-year-old male with persistent left sided facial heaviness and left-sided headache. Sinus CT scan revealed left side normal uncinat process with well-defined infundibular passage indicating type 1 maxillary sinus hypoplasia.

The third case was a thirty-four-year-old pregnant female with diplopia and left-sided headache. MRI revealed left maxillary sinus and inferiorly displaced left orbital floor with non-homogenous fluid-like signal intensity suggesting marked left long-standing sinusitis. The patient was managed by an antral lavage under local anesthesia. After delivery, she was reassessed by CT scan of the sinuses, which revealed left side small sized partial opacified type 2 maxillary sinus hypoplasia.

Conclusion: Symptomatic maxillary sinus hypoplasia patients with a diseased blocked ostiomeatal complex have special diagnostic considerations and therapeutic challenges.

Bahrain Med Bull 2017; 39(3): 150 - 153

Maxillary sinus hypoplasia (MSH) is the underdevelopment of maxillary sinus antrum and it is usually associated with other anomalies, such as uncinat process and infundibular passage. Hypoplasia is classified into three: Type I is mild hypoplasia, normal uncinat and infundibulum; Type II is significant hypoplasia with hypoplastic or absent uncinat and pathological infundibular passage; Type III is cleft-like sinus and absent uncinat¹.

MSH is an abnormality that is not commonly encountered. In the literature, it ranges from 0.73% to 10.4% in patients with sinus symptoms¹. Most cases are asymptomatic and discovered incidentally on routine conventional radiology.

CT of the paranasal sinuses confirms the diagnosis and differentiates it from infections or neoplasia². In addition, it provides a guide to the surgeon to plan the surgery and avoid unforeseen surgical complications³.

The aim of this report is to present a series of three cases of maxillary sinus hypoplasia which were managed surgically.

Case Report 1

A fourteen-year-old male patient presented with long-standing history of bilateral nasal blockage, nasal allergy, postnasal discharge and bilateral ear blockage.

* Consultant ENT Surgeon and Head of Department
Department of ENT
** Senior Resident
Department of ENT
*** Consultant Radiologist and Head of Department
Department of Radiology
Bahrain Defence Force Hospital
Kingdom of Bahrain
E-mail: drshilent@gmail.com; heshambdf@hotmail.com

Examination revealed retracted tympanic membranes, serous watery rhinorrhea from both nasal cavities and right side inferior turbinate engorgement. Flexible endoscopy was suggestive of allergic rhinitis. The patient was prescribed anti-allergic (Aerius 5 mg OD and Nasonex Spray) course but failed to improve. CT scan revealed right side ill-defined infundibular passage and complete opacified small right maxillary sinus, see figure 1. The CT was classified as hypoplasia type 2 according to Bolger et al⁶. In addition, adenoid hypertrophy was noted.



Figure 1: Case 1 CT Scan Showing Opacified Hypoplastic Right Maxillary Antrum – White Dotted Lines

The nature of the disease was discussed with the parents; adenoidectomy, radiofrequency turbinoplasty, right sided functional endoscopic sinus surgery and middle meatal antrostomy were performed. Postoperative follow-up one week and every three months for one year showed total remission.

Case Report 2

A twenty-seven-year-old male presented with nasal blockage on the left side, left sided facial heaviness and persistent left sided headache for the past two years. On examination, the turbinates were hypertrophied bilaterally on anterior rhinoscopy and endoscopic evaluation revealed bilateral ostiomeatal complex engorgement. CT scan of the sinuses revealed left side normal uncinete process and well-defined infundibular passage indicating type 1 maxillary sinus hypoplasia, see figure 2.



Figure 2: Case 2 Showing Left Maxillary Hypoplasia – White Dotted Lines

The patient failed to improve on Aerius and Nasonex spray. Therefore, limited functional endoscopic sinus surgery

including uncinectomy, middle meatal antrostomy with limited ethmoidectomy) and submucosal diathermy under general anesthesia was performed. The orbital fat was prolapsed in the nasal cavity after uncinectomy and was repaired using cartilage graft from the septum. Postoperatively, the patient was reviewed periodically with no subsequent relapse.

Case Report 3

A thirty-four-year-old pregnant female presented with diplopia and left-sided headache of gradual onset and recent worsening for which an MRI was performed revealing moderately thickened mucosal lining with a collapsed left maxillary sinus and inferiorly displaced left orbital floor with non-homogenous fluid-like signal intensity suggesting marked left long-standing sinusitis, see figure 3.

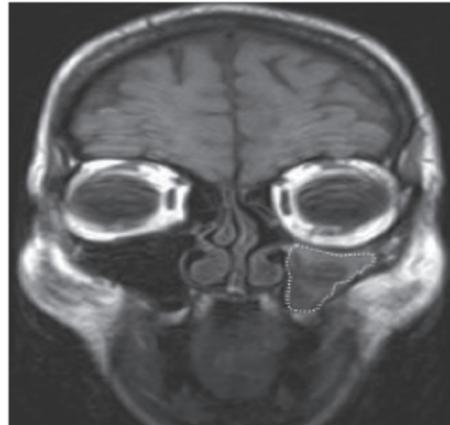


Figure 3: Case 3 MRI Scan Showing Hypoplastic Left Maxillary Antrum with Non-Homogenous Fluid-Like Signal Intensity – White Dotted Lines

On examination, moderate degree left enophthalmos, orbital asymmetry and an associated diplopia on the left lateral side. Nasal examination revealed mildly deviated septum with hypertrophy of the inferior turbinates. An antral lavage under local anesthesia was performed revealing resistance to irrigation due to blocked ostium and scanty mucopus. Post-partum, CT scan revealed an absent uncinete process on the left side, an ill-defined infundibular passage with inferior bowing of the orbital floor, lateral bowing of medial wall of maxillary sinus, posterior bowing of anterior wall of maxillary sinus and small partially opacified type 2 maxillary sinus hypoplasia, see figure 4.



Figure 4: Case 3 CT Scan Showing Hypoplastic Left Maxillary Antrum (White Dotted Lines) with Soft Tissue Thickening at the Left Infraorbital Margin

Left functional endoscopic sinus surgery was performed; laterally displaced medial wall of the maxillary sinus was found. The procedure included left sided middle meatal antrostomy with turbinate radiofrequency submucosal diathermy.

Postoperative follow-up for six months showed no relapse and widely patent middle meatal antrostomy.

DISCUSSION

Hypoplasia of the maxillary sinus incidence ranges from 1.73% to 10.4% of symptomatic patients; the maxillary sinus is rarely involved compared to the other sinuses^{1,4}.

The maxillary sinus starts developing by the fourth month of gestation as a mucosal evagination within the middle meatus. Any interference or arrest of this process could result into aplasia. The maxillary sinus continues to grow throughout life with progressive pneumatization laterally to attain its final adult size by mid-teens⁵.

Bolger et al were the first to propose an association between hypoplasia/aplasia of uncinate process and hypoplasia of maxillary sinus⁶. They classified the condition according to its severity, into three types: Type I - mild sinus hypoplasia, the uncinate process and the infundibulum are well formed with varying degrees of associated sinus mucosal thickening. Type II - a significant maxillary sinus hypoplasia is encountered with a hypoplastic or even an absent uncinate process with a poorly defined or absent infundibulum. The hypoplastic sinus might show total opacification. Type III - the maxillary sinus is profoundly hypoplastic with total absence of the uncinate process. In our study, cases 1 and 3 were Bolger type II and case 2 was Bolger type I. Failure to identify hypoplasia could result in inadvertent intraoperative breaching of the adjacent medial orbital wall with subsequent orbital complications⁶.

Bassiouny et al classified maxillary sinus hypoplasia into three, according to the position of the orbital floor in relation to the lateral nasal wall: grade I, II and III⁷. In our study, the first two cases were grade I and the third was grade II.

Further anatomical anomalies were highlighted by Erdem et al¹. They showed that such anomaly might lead to inadvertent orbital complications. None of our cases showed the presence of ethmoidmaxillary sinus. However, the uncinate was found to be absent or hypoplastic in all the three cases.

Weed et al found a vertical dystopia of the orbit along with a clinically absent natural maxillary ostium and an associated poorly developed uncinate process and infundibular passage⁸. In our study, vertical dystopia was found in case 3.

Sirikci et al found that there might be an association between a low-lying roof of the ethmoid and maxillary sinus hypoplasia, which might lead to intracranial complications during surgery⁹. In our case series we found that only case 3 had a low lying roof of ethmoid.

Silent sinus syndrome was seen in our third case. It is a relatively rare clinical condition among maxillary sinus hypoplasia characterized by spontaneous and progressive

enophthalmos and hypoglobus with an associated atelectasis of the maxillary sinus with resultant alteration of the orbital floor¹⁰. The syndrome was first identified by Montgomery in 1964, but it was not until 1994 when the name was coined by Soparkar et al¹². The treatment consists of re-ventilation of the atelectatic sinus by endoscopic sinus surgery, similar to our management in our third case¹³.

Relieve of negative sinus pressure and re-expansion of the collapsed cavity by limited antrostomy could reduce the enophthalmos, but wide antrostomy prevents future reobstructions^{14,15}. Orbital floor management remains a gray area; it depends on the degree of the diplopia, the cosmetic alterations and the postsurgical evaluation¹⁶.

CONCLUSION

Maxillary sinus hypoplasia is a rare and asymptomatic condition; it is usually discovered incidentally during CT imaging. Otolaryngologist should be aware of it to avoid complications and breach of lamina papyracea during FESS.

Author Contribution: All authors share equal effort contribution towards (1) substantial contributions to conception and design, acquisition, analysis and interpretation of data; (2) drafting the article and revising it critically for important intellectual content; and (3) final approval of the manuscript version to be published. Yes.

Potential Conflicts of Interest: None.

Competing Interest: None.

Sponsorship: None.

Acceptance Date: 17 May 2017.

Ethical Approval: Approved by Research Ethic Committee of Bahrain Defense Force Hospital, Kingdom of Bahrain.

REFERENCES

1. Erdem T, Aktas D, Erdem G, et al. Maxillary Sinus Hypoplasia. *Rhinology* 2002; 40(3):150-3.
2. Jafari-Pozve N, Sheikhi M, Ataie-Khorasgani M, et al. Aplasia and Hypoplasia of the Maxillary Sinus: A Case Series. *Dent Res J (Isfahan)* 2014; 11(5):615-7.
3. Khanduri S, Agrawal S, Chabra S, et al. Bilateral Maxillary Sinus Hypoplasia. *Case Reports in Radiology* 2014; 2014: Article ID 148940, 3 pages.
4. Thiagarajan B, Narashiman NS. Hypoplasia of All Paranasal Sinuses: A Case Series and Literature Review. *Otolaryngology Online Journal* 2012; 2(2).
5. R Dwivedi, N Samanta, K Kishore, et al. Bilateral Maxillary Sinus Hypoplasia: A Rare Cause of Chronic Facial Pain. *The Internet Journal of Radiology* 2006; 7(2).
6. Bolger WE, Woodruff WW Jr, Morehead J, et al. Maxillary Sinus Hypoplasia: Classification and Description of Associated Uncinate Process Hypoplasia. *Otolaryngol Head Neck Surg* 1990; 103(5(Pt 1)):759-65.

7. Bassiouny A, Newlands WJ, Ali H, et al. Maxillary Sinus Hypoplasia and Superior Orbital Fissure Asymmetry. *Laryngoscope* 1982; 92(4):441-8.
8. Weed DT, Cole RR. Maxillary Sinus Hypoplasia and Vertical Dystopia of the Orbit. *Laryngoscope* 1994; 104(6 Pt 1):758-62.
9. Sirikçi A, Bayazit Y, Gümüşburun E, et al. A New Approach to the Classification of Maxillary Sinus Hypoplasia with Relevant Clinical Implications. *Surg Radiol Anat* 2000; 22(5-6):243-7.
10. Guillen DE, Pinargote PM, Guarderas JC. The Silent Sinus Syndrome: Protean Manifestations of a Rare Upper Respiratory Disorder Revisited. *Clin Mol Allergy* 2013; 11(1):5.
11. Montgomery WW. Mucocele of the Maxillary Sinus Causing Enophthalmos. *Eye Ear Nose Throat Mon* 1964; 43:41-4.
12. Soparkar CN, Patrinely JR, Cuaycong MJ, et al. The Silent Sinus Syndrome. A Cause of Spontaneous Enophthalmos. *Ophthalmology* 1994; 101(4):772-8.
13. Sciarretta V, Pasquini E, Tesi F, et al. Endoscopic Sinus Surgery for the Treatment of Maxillary Sinus Atelectasis and Silent Sinus Syndrome. *J Otolaryngol* 2006; 35(1):60-4.
14. Nkenke E, Alexiou C, Iro H, et al. Management of Spontaneous Enophthalmos Due to Silent Sinus Syndrome: A Case Report. *Int J Oral Maxillofac Surg* 2005; 34(7):809-11.
15. Numa WA, Desai U, Gold DR, et al. Silent Sinus Syndrome: A Case Presentation and Comprehensive Review of all 84 Reported Cases. *Ann Otol Rhinol Laryngol* 2005; 114(9):688-94.
16. Cardesín A, Escamilla Y, Romera M, et al. Single Surgical Step for Endoscopic Surgery and Orbital Reconstruction of a Silent Sinus Syndrome. *Acta Otorrinolaringol Esp* 2013; 64(4):297-9.