

Idiopathic maxillary antral mucocele in a child: a rare presentation

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Abstract

We report a case of a large maxillary sinus mucocele in a 14-year-old girl presenting with epiphora, proptosis and dental pain. This was marsupialized endoscopically, with complete resolution of symptoms over three months' follow up. The literature is reviewed.

Key words: Mucocele; Maxillary Sinus, Surgery; Endoscopes

Introduction

Mucoceles of the maxillary sinus are rare. They are mostly seen in adults who have undergone previous surgery, the reported incidence in adults varying from 3 to 10 per cent.¹ A higher prevalence has been reported in the Japanese population and has been ascribed to the fact that more Caldwell-Luc procedures are performed in that country.^{1,2} In children, paranasal sinus mucoceles are rarer and are thought to be related to cystic fibrosis, although some reports challenge this view.^{2,3} Idiopathic mucoceles of the maxillary antrum are rare in children^{3,4} and the authors could identify only one case in the literature.

Case report

A 14-year-old girl presented to the accident and emergency department with a three-month history of persistent, right-sided facial pain. On further questioning, she also reported retro-orbital pain, epiphora and toothache over the right upper molars. She had been treated by her general practitioner with three courses of oral antibiotics and fluticasone nasal spray, which had not relieved her symptoms. She had had no previous rhinological surgery or trauma and her past medical and developmental histories were unremarkable.

On examination, the patient had a diffuse swelling over her right cheek, causing facial asymmetry and mild right proptosis. Anterior rhinoscopy revealed complete obstruction of the right nasal cavity by a large, fleshy, smooth swelling inside the nasal cavity. A computed tomography (CT) scan of the paranasal sinuses showed a large, expansile, low attenuation lesion arising from the right maxillary sinus (Figure 1). This was seen extending into the right ethmoid and sphenoid sinuses and the nasal cavity, with deviation of the septum to the left (Figure 2). The lesion extended posteriorly to the infratemporal fossa (Figure 1b). The lesion had caused expansion and thinning of the bony margin of the right maxilla. There was minimal enhancement of the lining mucosa with intravenous contrast, but some mucosal enhancement was seen in the ethmoid sinus. A left maxillary cyst, 3 cm in diameter,

was also noted (Figure 1). From these appearances, the lesion was deemed likely to be a large right mucocele arising from the maxillary sinus.

The patient underwent nasal endoscopy under general anaesthesia, with endoscopic marsupialization of the mucocele. The diagnosis of a mucocele was confirmed histologically.

The patient was discharged 24 hours post-operatively on oral antibiotics and nasal decongestants. She was symptom free over the next six months, and evaluation in the out-patients department showed no evidence of recurrence.

Discussion

Idiopathic maxillary mucoceles in children are rare and their aetiology is poorly understood. Retention cysts are far more common in the maxillary antrum,⁴ but mucoceles have been described in association with neoplasia,^{5,6} trauma, surgery, inflammatory processes (e.g. cystic fibrosis) and congenital abnormalities.⁷ Idiopathic mucoceles are thought to be due to chronic inflammation causing obstruction of mucus-secreting glands.² Our patient had a three-month history, suggesting a chronic problem.

The most common presenting symptom of maxillary mucocele is facial swelling and, when extending to the orbit, displacement of the eye, with possible diplopia.¹ Epiphora is more common in ethmoid sinus mucoceles⁸ but has also been described in association with a maxillary sinus mucocele.² Facial pain and headache are common although toothache appears to be more unusual. What makes our case unique is that all the walls of the maxillary antrum were affected, causing all the above symptoms simultaneously.

The radiological findings of the CT scan were consistent with previous descriptions of mucoceles, with a uniformly expansile mass within the maxillary antrum, which was not enhanced by contrast, and thinning of the bone, mostly of the anterior wall.^{4,9} Bony thinning implies a chronic process. However, as the differential diagnosis includes carcinoma, a biopsy is still required to confirm the radiological diagnosis.

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FIG. 1

Axial post-contrast computed tomography scan of the sinuses: (a) soft tissue and (b) bony windows. The images show a soft tissue density that occupies the right maxillary sinus and extends to fill completely the right nasal cavity. The lateral bony wall is thinned, with extension into the infratemporal fossa.

- Idiopathic maxillary sinus mucoceles are rare in children
- The presenting symptoms vary according to the route of the mucocele's expansion
- This case shows that mucoceles may present with a wide range of symptoms in children and may erode through all the walls of the maxillary sinus
- Endoscopic surgery for marsupialization of idiopathic mucoceles can produce amelioration of symptoms, but long-term follow up is required to ensure that no relapse occurs

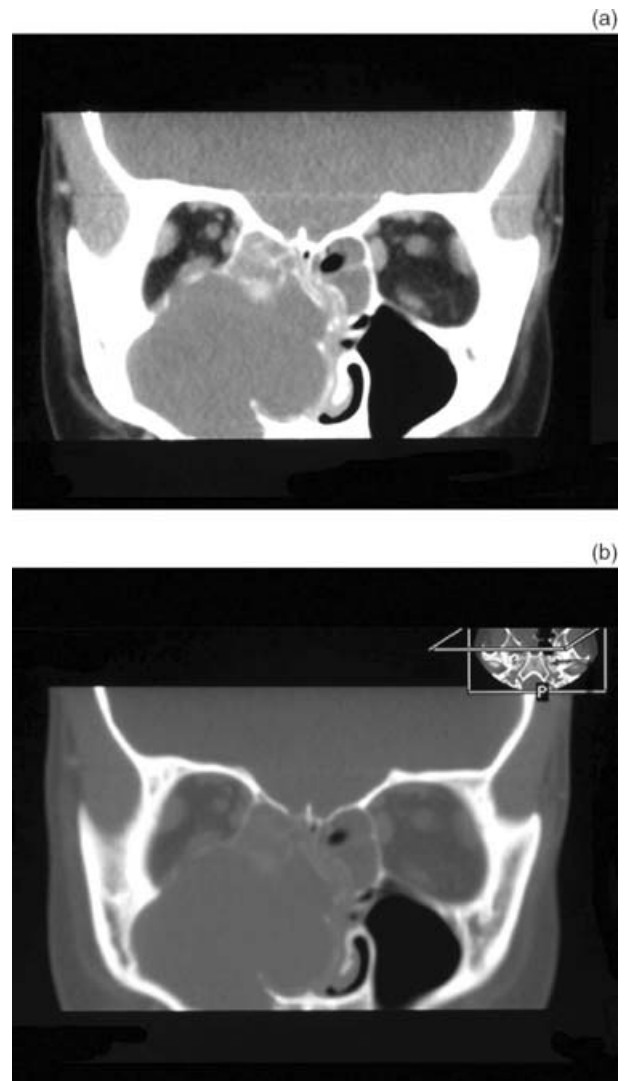


FIG. 2

Coronal post-contrast computed tomography scan of the sinuses: (a) soft tissue and (b) bony windows. The images show the maxillary mucocele causing expansion of the right maxillary sinus. The nasal septum is bowed to the left.

There is some evidence that mucoceles in children can be treated endoscopically, and there are reports suggesting that the results of endoscopic treatment are so satisfactory that this should now be the 'gold standard' for management of this condition.^{3,10-13} In our experience thus far, endoscopic marsupialization has produced good results, although we would stress that long-term follow up is needed.

Conclusion

We report a case of idiopathic maxillary sinus mucocele in a child, a rare occurrence. In addition, this case is unusual as the mucocele was affecting all the walls of the maxillary antrum, giving rise to corresponding signs and symptoms. This patient was treated surgically using endoscopic marsupialization, with a good result.

References

1 Hartley BEJ, Lund VJ. Endoscopic drainage of pediatric paranasal sinus mucoceles. *Int J Pediatr Otorhinolaryngol* 1999;**50**:109-11

- 2 Skoulakis CE, Velegrakis GA, Doxas PG, Papadakis CE, Bizakis JG, Helidonis ES. Mucocele of the maxillary antrum in an eight-year-old boy. *Int J Pediatr Otorhinolaryngol* 1999;**47**:283–7
- 3 East D. Mucocoeles of the maxillary antrum. Description, case reports and review of the literature. *J Laryngol Otol* 1985;**99**:49–56
- 4 Al-Dousary S, Al-Kharashi S. Maxillary sinus mucopyocele in children: a case report and review of the literature. *Int J Pediatr Otorhinolaryngol* 1996;**36**:53–60
- 5 Raj P, Mohanradja D, Siddiq MA, Stansbie JM. Nasal non-Hodgkin's lymphoma causing mucocele of the maxillary antrum. *J Laryngol Otol* 2000;**114**:158–9
- 6 Scott A, Raine M, Stansbie JM. Ethmoid metastasis of endometrial carcinoma causing mucocoele of maxillary antrum. *J Laryngol Otol* 1998;**112**:283–5
- 7 Yue V, Bleach NR, Van Hasselt CA. Double maxillary antrum as a cause of maxillary sinus mucocele. *Ear Nose Throat J* 1994;**73**:839–41
- 8 Ormerod LD, Weber AL, Rauch SD, Feldon SE. Ophthalmic manifestations of maxillary sinus mucoceles. *Ophthalmology* 1987;1994(8):1013–19
- 9 Han MH, Chang KH, Lee CH, Yeon KM, Han MC. Cystic expansile masses of the maxilla: differential diagnosis with CT and MR. *Am J Neuroradiol* 1995;**16**:333–8
- 10 Ikeda K, Takahashi C, Oshima T, Suzuki H, Satake M, Hikada H *et al.* Endonasal endoscopic marsupialisation of paranasal sinus mucoceles. *Am J Rhinol* 2000;**14**:107–11
- 11 Sciaretta V, Pasquini E, Farneti G, Ceroni AR. Endoscopic treatment of paranasal sinus mucoceles in children. *Int J Pediatr Otorhinolaryngol* 2004;**68**:955–60
- 12 Rombaux P, Bertrand B, Eloy P, Collet S, Daele J, Bachert C *et al.* Endoscopic endonasal surgery for paranasal sinus mucoceles. *Acta Otorhinolaryngol Belg* 2000;**54**:115–22
- 13 Har-El G. Endoscopic management of 108 mucoceles. *Laryngoscope* 2001;**111**:2131–4

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