

Maxillary sinus mucocoeles – 10 cases – 8 treated endoscopically*

Marc Makeieff, Quentin Gardiner, Michel Mondain, Louis Crampette

Service ORL A, Hôpital Saint-Charles, CHU de Montpellier, France.

SUMMARY

Maxillary sinus mucocoeles are rare, but may cause significant problems outside the sinus including diplopia and cheek swelling. We present 9 patients suffering from maxillary sinus mucocoele, including one patient with bilateral involvement, representing 10 involved sinuses. Eight sinuses were treated endoscopically and followed-up for 3 to more than 6 years; one of these developed a recurrence. Two sinuses were treated using a classical external approach (Caldwell-Luc), one at the beginning of our series, and one as the endoscopic technique was difficult to perform due to loss of anatomical landmarks. One of these two sinuses developed a recurrence and was then successfully treated endoscopically. We discuss the aetiology, pathogenesis and appropriate treatment of this unusual condition. Endoscopic treatment appears to be effective and leads to minimal recurrence. The aim is to perform a wide removal of the medial wall of the maxillary sinus including most of the inferior turbinate, immediately posterior to the lachrymal duct leaving intact the sinus mucosa. In some cases, however, the endoscopic technique can be difficult to perform due to loss of essential anatomical landmarks and in these cases recourse to an external approach may be necessary.

Key words: maxillary sinus, mucocoele, endoscopic surgery, middle meatal antrostomy

INTRODUCTION

A mucocoele of a paranasal sinus is an epithelial-lined cavity containing aseptic mucus (Natvig and Larsen, 1978). It can occur in the frontal, ethmoidal, sphenoidal and maxillary sinuses. In whichever paranasal sinus, they have a gradual growth which can produce bony destruction and may extend beyond the confines of the sinus leading to ophthalmic involvement and facial deformities (Hasegawa et al., 1979). We present our results in a series of ten mucocoeles involving the maxillary sinus either treated endonasally (8 sinuses) or by an open approach (2 sinuses) with three to more than six years of follow-up.

MATERIALS AND METHODS

In the Otolaryngology/Head and Neck Surgery Unit in Montpellier, France, 9 patients with 10 maxillary sinus mucocoeles were treated between 1988 and 1994. A complete endoscopic and CT scan examination was performed for each patient.

The age range was from 42 to 78 years (mean age 54). Five were female and four were male. One patient had a bilateral form and one had maxillary and frontal mucocoeles. All the other patients had isolated maxillary mucocoeles.

One patient had a long history of chronic sinusitis and one case had had facial trauma that was not treated. Seven had undergone previous open approach maxillary sinus surgery for chronic sinusitis or trauma. The mean time between performing a Caldwell-Luc procedure and the discovery of the mucocoele was 15 years (from 5 to 25 years).

Diplopia led to the discovery of the mucocoele in 5 of our cases. Another common symptom reported by the patients was nasal obstruction with purulent anterior nasal discharge in 5 cases, a swelling of the cheek below the orbit was present in three patients and a bulging of the buccal sulcus which caused difficulties with the correct fitting of dentures in 2 cases. Five had a sensory deficit in the territory of the infra-orbital nerve (though whether this was due to the development of the mucocoele or to the previous Caldwell-Luc procedure was difficult to assess), complained of a feeling of discomfort on the cheek, and unilateral epiphora was the only symptom reported in one patient (Table 1). In the majority of cases endonasal examination showed no abnormality, however bulging of the lateral wall of the nose was noted in 3 cases.

All patients had plain sinus views performed (showing opacification of the antrum) and CT scan evaluation showing a rounded homogeneous opacity of the entire maxillary sinus with clear

* Received for publication March 12, 1998; accepted April 27, 1998

Table 1. Principal symptoms of patients at presentation. (Nasal disch. - nasal discharge. Anaes. - anaesthesia in territory of infra-orbital nerve. Cheek sw. - swelling of cheek. Pain- pain in cheek or forehead. Bucc. bulge-swelling in buccal sulcus. Epiph.- unilateral epiphora)

| Patient | Nasal disch | Diplopia | Anaes. | Cheek sw. | Pain | Bucc. bulge | Epiph. |
|---------|-------------|----------|--------|-----------|------|-------------|--------|
| 1 | x | x | x | | x | | |
| 2 | x | x | x | | x | | |
| 3 | y | | x | | | | |
| 4 | | | | x | | | |
| 5 | x | | | | x | x | |
| 6 | | x | x | x | | x | |
| 7 | x | | x | x | | | |
| 8 | x | | | | | | |
| 9 | | | | | | | x |
| Total | 5 | 2 | 3 | 3 | 3 | 2 | 1 |

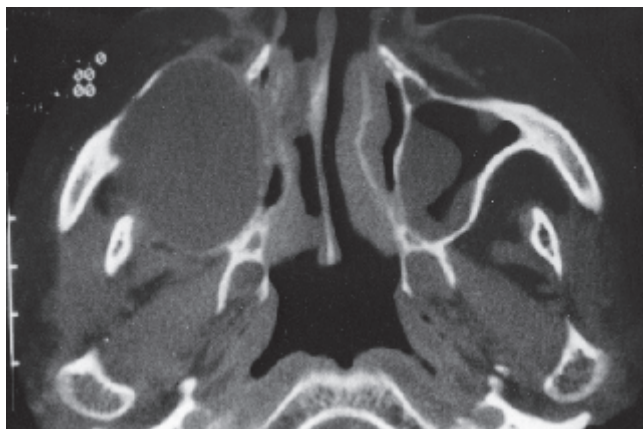


Figure 1. Axial CT of maxillary sinus mucocoele causing bulging of the lateral nasal wall and erosion of the anterior wall of the maxilla.

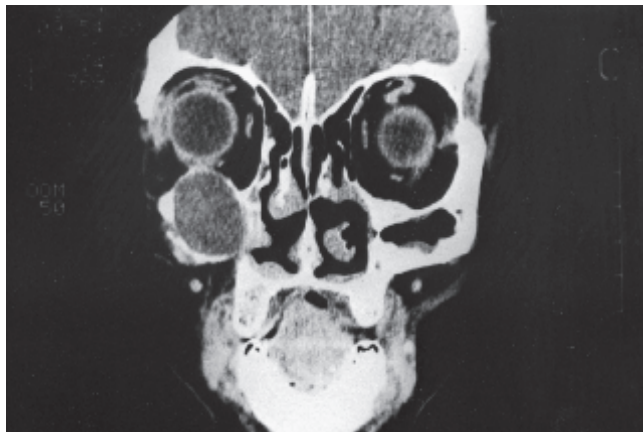


Figure 2. Coronal CT of maxillary sinus mucocoele involving the orbit.

limits and bone erosion in all cases (Figure 1). In 5 cases the mucocoele involved orbital structures (Figure 2). Magnetic resonance imaging was also performed in 3 patients which showed an increased signal intensity on the T2-weighted images (Figure 3). Ophthalmic examination, including the Lancaster test, was performed for every case with diplopia, confirming the diplopia but not showing any other visual problem. Surgery was performed under general anaesthesia. Eight sinuses were treated only by endoscopic surgery. Three sinuses were treated with a classical middle meatal antrostomy, and five had



Figure 3. Axial T2-weighted MR scan of maxillary mucocoele.

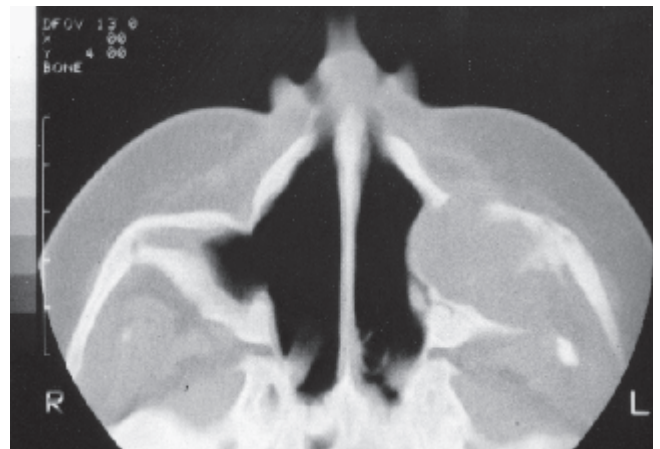


Figure 4. Axial CT scan of patient with bilateral mucocoeles showing recurrent left maxillary sinus mucocoele following open approach surgery (right side previously treated endoscopically).

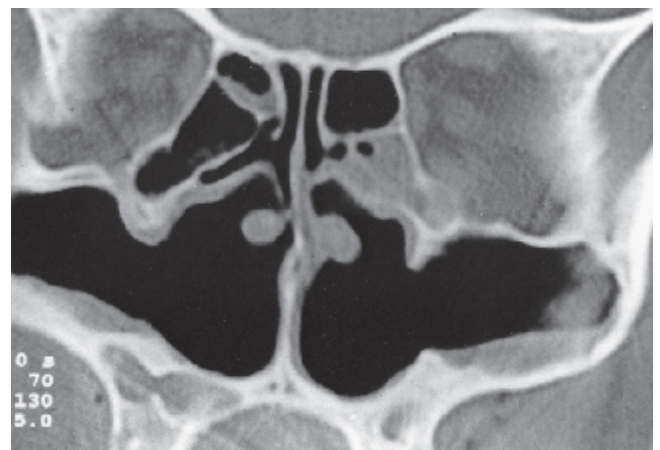


Figure 5. Coronal CT of patient with bilateral mucocoeles following endoscopic procedure for recurrence in left maxillary sinus.

an extended antrostomy including the middle and inferior meatuses with removal of the inferior turbinate creating a large window into the nose. The two other cases had a transoral procedure (Caldwell-Luc approach) with opening of the inferior meatus, which was followed either because of difficulties in gaining access endonasally or because the procedure was performed before endonasal surgery was available in our department. Patients remained in hospital for two days after the operation, until removal of the nasal packing.

The follow-up is more than 6 years for 4 patients (the 2 patients treated by an open approach and 2 treated endonasally) and more than 3 years for the others. None of the patients treated had a postoperative complication, and the diplopia resolved in all cases. Of the 10 sinuses treated, good results were obtained in 8. Recurrences were found in two patients. One of these had bilateral maxillary mucocoeles- the right sided mucocoele was treated endonasally, the left side treated by an open approach because of difficulty in gaining access endonasally. The left-sided mucocoele recurred 2 years later (Figure 4). Then it was found that an endonasal approach was possible due to bulging of the lateral nasal wall which enabled an easier access into the sinus. This mucocoele has not recurred since (Figure 5). The other patient with a recurrence was the one who failed to attend for post-operative care soon after a middle meatal antrostomy. He presented with a recurrence of the disease two years after operation and was then treated by transoral surgery associated with a large inferior meatal antrostomy.

DISCUSSION

Epidemiology and Pathogenesis

A mucocoele is an expansive pseudo-cystic lesion lined with a modified mucous membrane involving the entire sinus. Mucocoeles may appear in any paranasal sinus. In the literature, involvement of the frontal sinus (65 percent of the cases) and the anterior part of the ethmoid sinus (30 percent of the cases) is more frequent. The maxillary sinus is a less common site of development, with only 3 to 10 percent (Natvig and Larsen, 1978). They have a progressive growth and the ability to cause bony erosion that may lead to signs on the outside of the sinus (East, 1985).

Maxillary sinus mucocoeles must be differentiated from other benign types of cystic lesion partially involving the floor of the sinus, sometimes called muroid cysts or pseudocysts; even if some muroid cysts involving the floor of the sinus can expand to fill the entire sinus, the main pathophysiological mechanism leading to the development of a mucocoele involves obstruction of the sinus ostium (East, 1985). The definition of a mucocoele is important when comparing different series as there sometimes appears to be some confusion in the terminology for these lesions (Le Guillou et al., 1985).

All reports about maxillary sinus mucocoeles from European and North American centres involve a small number of patients and very few cases reported have been treated endoscopically. One recent publication reports on 9 patients, 3 of whom were treated endoscopically (Marks et al., 1997). In Japan, however, the series are much larger, Hasegawa reporting 132 cases (Hasegawa et al., 1979), defining clearly that these were all true mucocoeles. In this report, 99% of the patients had had previous sinus surgery. Maxillary mucocoele was thought to be more frequent in Japan, perhaps because of the more frequent surgical removal of the sinus mucosa in chronic sinusitis. The relationship between the development of a maxillary sinus mucocoele and previous transoral surgery of the sinus (a Caldwell-Luc procedure) is emphasised in most reports (Hasegawa et al., 1979; Demaldent and Groboi, 1983; Mendelsohn et al., 1984). Seven of the nine patients that were treated in our institution had pre-

viously undergone a Caldwell-Luc procedure. The mechanism involved may be a fibrous reaction involving the sinus ostium and therefore obstructing it, or the formation of fibrotic bands between the anterior and posterior antral walls obstructing the normal drainage of the maxillary sinus (Som, 1992). Other, less common, causes may be involved; an obstruction may be caused by bony trauma with alteration of the mucosa around the sinus ostium (one case in our series) or from previous sinus infections (East, 1985). Histological examination of the wall of a mucocoele shows a mucosal lining of modified pseudo-stratified columnar epithelium (Natvig and Larsen, 1978). Areas of squamous metaplasia are frequent. The submucosa is thick and fibrous with an infiltration of inflammatory cells. Because of this chronic inflammation, the epithelium may secrete inflammatory factors including prostaglandins, some of which (such as PGE2) can lead to osseous resorption. In a study of fronto-ethmoidal sinus mucocoeles, however, the osteolytic cytokine interleukin-1 was found in all cases of mucocoele, but in very few cases of chronic sinusitis (even when the sinus ostium was obstructed). The underlying cause for this increase in levels of IL-1 remains unclear (Lund et al., 1993). However, ventilation of the sinus and marsupialisation of the mucocoele often promotes the spontaneous reformation of the lost bone whatever the underlying mechanism.

Symptoms and signs

The clinical presentation may vary greatly depending on the development of the mucocoele within or outside the bony walls of the sinus. No signs of the development of the mucocoele may be noted for a long period of time (Demaldent and Groboi, 1983; Mendelsohn et al., 1984). During the intrasinus phase there are often no signs suggestive of an abnormality – sometimes a vague feeling of discomfort on the cheek has been complained of, but often this is only mentioned after the diagnosis has been made and the symptoms are reviewed retrospectively. The more usual symptom complained of first is a purulent nasal discharge, but whether this is due to the mucocoele or to an underlying rhinosinusitis is unclear. Anaesthesia in the territory of the infra-orbital nerve and epiphora may also be complained of, but it can be difficult to determine whether in fact this is due to the mucocoele or to the previous surgery. It may develop completely silently until it reaches a size when it begins to cause bulging of one of the walls of the maxilla and bone erosion (Sadoff and Rubin, 1991). The more serious signs noted are when the mass starts to involve the orbit. Diplopia due to pressure on the inferior rectus muscle is the most frequent sign of orbital involvement. The muscle is pushed upwards by the bulging of the floor of the orbit caused by the expanding mass (Demaldent and Groboi, 1983; Crain et al., 1990).

Radiological signs

Plain films show complete opacity of the antrum but the information gained from plain sinus views is minimal, and CT scan evaluation is now the investigation of choice. CT reveals a diffuse opacity with thinning of the walls of the sinus (Som, 1990). The mass has a uniform and homogeneous density. The mar-

gins of the mass appear convex even if there has been erosion of the walls of the sinus and there is no infiltration of the surrounding tissues (Figure 1). There appears to be a clear division between the mucocoele and the surrounding tissues which is due to a thickening of the mucosa and the periosteum. When the lesion pushes through the floor of the orbit, the inferior rectus muscle is elevated (Figure 2). The upper border of the mass is convex, because even if there is bone erosion the periosteum is never breached, in distinction to malignant tumours of the sinus. MR imaging is probably not necessary in all cases if the diagnosis is clear from CT results. MRI does not clearly define the bony walls of the sinus, but may be useful particularly with fronto-ethmoidal and sphenoidal mucocoeles when there is doubt as to the exact pathology and where the mucocoele must be distinguished from tumour (Lanzieri et al., 1991).

Treatment

The usual treatment until recently was a Caldwell-Luc procedure (Hasegawa et al., 1979; Marks et al., 1997). This was done using a transoral approach with the creation of a bony window on the anterior face of the maxilla. The mucosa was then entirely removed, and an inferior meatal antrostomy fashioned with the aim of achieving drainage and ventilation of the sinus. Over the last few years, with the development of endoscopic surgery, our preferred surgical technique has changed. The endoscopic treatment of mucocoeles was perfected initially for the treatment of frontal and ethmoidal mucocoeles, as the concept of marsupialising the mucocoele into the nasal cavity rather than completely removing the mucosa was recommended as the preferable method, the aim being to ensure adequate ventilation of the sinus cavity (Kennedy et al., 1989; Serrano et al., 1992).

The aim of endoscopic treatment is to open into the maxillary sinus by performing a middle meatal antrostomy which includes the natural ostium to recreate normal physiological drainage. When fashioning the antrostomy the surgeon must bear in mind two points:

- 1) that the localisation of the natural ostium can be difficult
- 2) that there is a tendency in this condition of the antrostomy to spontaneous stenosis

Therefore, it is important to make the middle meatal antrostomy as large as possible and when necessary the classical middle meatal antrostomy can be extended further than the usual limits so that the medial wall of the maxillary sinus is widely fenestrated. The limits of this extended antrostomy are (Figures 4 and 5):

- superiorly, the floor of the orbit
- anteriorly, the lachrymal duct and
- inferiorly, the floor of the nose (with consequent removal of the inferior turbinate)

In difficult cases when there may have been previous trauma or surgery distorting or destroying anatomical landmarks, an open approach may be required. The use of the open approach neither implies the removal of the sinus mucosa nor the fashioning of an inferior meatal antrostomy as was the case in the past (a Caldwell-Luc procedure), but allows the safe creation of a large combined middle and inferior meatal antrostomy as described

above. The open approach is therefore used as an adjunct to the endonasal procedure rather than as its replacement.

Whichever surgical procedure is chosen, post-operative care is of great importance. All patients who have undergone such surgery require careful follow-up for a few weeks to ensure that the healing of the antrostomy is proceeding normally. Secretions and crusts must be removed, and adhesions within the sinus or between the ethmoid wall and the middle turbinate divided. The one patient who required a second procedure after endoscopic surgery had not complied with our recommended post-operative care and when he returned two years later his antrostomy had closed up.

CONCLUSION

Isolated maxillary sinus mucocoeles are rare and are often diagnosed late when orbital complications or facial deformities have already occurred. Despite their size and their sometimes impressive clinical appearances, endoscopic surgery, in this small series, seems to be an effective and rapid treatment which causes minimal morbidity. Two of our patients treated endoscopically have been followed-up for more than six years without recurrence, and all patients treated endoscopically who returned for regular post-operative care have had no recurrence at three years.

REFERENCES

1. Crain MR, Dolan KD, Maves MD (1990) Maxillary sinus mucocoele. *Ann Otol Rhinol Laryngol* 99: 321-322.
2. Demaldent JE, Groboi J (1983) Mucocèle géante du sinus maxillaire. *Ann Otolaryngol (Paris)* 100: 523-525.
3. East D (1985) Mucocoeles of the maxillary antrum; description, case report and review of the literature. *J Laryngol Otol* 99: 49-56
4. Hasegawa M, Saito Y, Watanabe I (1979) Postoperative mucocoeles of the maxillary sinus. *Rhinology* 17: 253-256.
5. Kennedy DW, Josephson JS, Zinreich J, Mattox DE, Goldsmith MM (1989) Endoscopic sinus surgery for mucocoeles: a viable alternative. *Laryngoscope* 99: 885-895.
6. Lanzieri CF, Shah M, Krauss D, Laveru P (1991) Use of Gadolinium-enhanced MR imaging for differentiating mucocoeles from neoplasms in the paranasal sinuses. *Radiology* 178:425-428
7. Le Guillou C, Brette MD, Klap P, Negrier B, Sicot H, Freyss G (1985) Mucocèle du sinus maxillaire. Problèmes diagnostiques et thérapeutiques. A propos de 5 cas. *Ann Otolaryngol (Paris)* 102: 531-535
8. Lund VJ, Henderson B, Song Y (1993) Involvement of cytokines and vascular adhesion receptors in the pathology of fronto-ethmoidal mucocoeles. *Acta Otolaryngol (Stockh)* 113: 540-546
9. Marks SC, Latom JD, Mathog RH (1997) Mucocoeles of the maxillary sinus. *J Otolaryngol Head Neck Surg* 117:18-21.
10. Mendelsohn DB, Glas RBJ, Hertzanu Y (1984) Giant maxillary antral mucocoele. *J Laryngol Otol* 98: 305-310.
11. Natvig K, Larsen TE (1978) Mucocoele of the paranasal sinus. *J Laryngol Otol* 92: 1075-1082
12. Sadoff RS, Rubin M (1991) Bilateral antral mucocoeles. A report of a case. *J Oral Maxillofac Surg* 49:193-196
13. Serrano E, Pessey JJ, Lacomme Y (1992) Les mucocèles sinusiennes: aspects diagnostiques et chirurgicaux. (A propos de 8 cas traités par rhino-chirurgie endoscopique) *Acta Otorhinolaryngol Belg* 46: 287-292.
14. Som PM (1992) Postsurgical mucocoele. *Am J Otolaryngol* 13:237-238

Dr M Makeieff MD
 Service ORL A,
 Centre Hospitalo-Universitaire St Charles
 300, rue Auguste Broussonnet
 34295 Montpellier Cedex 5, France
 Fax +33 (0)4 67 54 50 15