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ORIGINAL ARTICLE

Sinus mucocele: Natural history and long-term recurrence rate

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KEYWORDS

Sinus mucocele;
Nasal polyposis;
Endonasal sinus
surgery;
MRI

Summary

Objective: To define the natural history, clinical signs, treatment and the modalities of medium- and long-term follow-up of patients operated for sinus mucocele.

Patients and method: Retrospective study of all patients operated for sinus mucocele between January 1993 and December 2009 ($n = 68$). Demographic data, symptoms, medical imaging findings, surgical treatment and results were recorded.

Results: The mean age of patients in this series was 53 years (range: 27–82 years, sex ratio: 3/2). The most common site was fronto-ethmoidal. Fifty-one patients (75%) had a history of sinus surgery, essentially for nasal polyposis. Only 15% of mucoceles occurred spontaneously. Presenting symptoms, in decreasing order of frequency, were facial pain or headache (38%), ocular or orbital complications (28%), while 20% of patients were asymptomatic. Surgery was performed by endonasal endoscopic sinus surgery ($n = 57$, 84%) or via a combined, transfacial and endonasal approach, associated with navigation after January 2003. The mean follow-up was 7 years (range: 4 months–16 years). During this follow-up period, 23.5% of patients developed recurrence or a second mucocele after a mean interval of 4 years.

Conclusion: This study demonstrates the high recurrence rate of mucocele, particularly in multi-operated patients with chronic sinusitis. Long-term, regular, clinical and radiological follow-up is necessary to detect asymptomatic lesions prior to the onset of complications.

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Introduction

Sinus mucoceles are benign cystic tumours, arising at the expense of the paranasal sinus mucosa, lined by non-neoplastic epithelium, and containing usually sterile mucus [1]. The origin of sinus mucoceles remains controversial: they appear to be related to a chronic inflammatory process occurring in a closed space, related to a benign tumour,

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Table 1 Clinical characteristics of 51 patients with a history of surgery.

n	Type of operation	Indication				Free interval
		Polyposis	Infection	Tumour	Other	
33(64.7%)	Ethmoidectomy (n = 25)	18	2	5 (PI)	0	4 years 5 m
	Lemoine pin (n = 1)	0	1	0	0	7 months
	Albertini drain (n = 1)	0	1	0	0	5 years
	Meatotomy/ (n = 4)	2	2	0	0	3 years
	Draf 3 (n = 1)	1	0	0	<i>Pneumosinus dilatans</i>	2 years
	Transsphenoidal incision (n = 1)	0	0	0	Pituitary cyst	4 months
13(25.4%)	Bicoronal (n = 3)	0	0	0	Retroorbital cyst Craniostenosis <i>Pneumosinus dilatans</i>	8 years 6 m
	Caldwell-Luc (n = 10)	1	6	0	3	20 years
5 (9.8%)	PLN + endonasal (n = 2)	0	0	2 (IP)	0	5 years 8 m
	Bicoronal + endonasal (n = 1)	0	0	0	1	9 years
	Jacques + endonasal (n = 2)	1	1	0	0	8 years
Total		23	13	7	6	7 years
		45%	25.50%	13.70%		

PLN: paralateronasal incision; IP: inverted papilloma.

post-traumatic scarring or inflammation [2], causing ostial obstruction leading to accumulation of mucus secretions [3]. These space-occupying, expanding, and destructive formations remain asymptomatic for a long time and may present suddenly with potentially serious ocular or intracranial complications [4]. CT and MRI examination of the sinuses now allows assessment of their extension in relation to adjacent structures [5]. Surgery is the only curative treatment. Although external resection remained the reference technique for many years, endonasal endoscopic sinus surgery is now generally used, as it is more conservative and less aggressive [6]. In contrast, this endoscopic surgery constitutes one of the major causes of mucocoeles [7–9]; the incidence of mucocoeles has markedly increased since the 1990s, in parallel with the growth of endonasal endoscopic surgery [8].

In this study, we report a series of 68 cases of operated mucocoele. The objective of this study was to define the presenting complaints and analyse the medium-term and long-term results in order to define the recurrence rate and propose the optimal postoperative follow-up.

Table 2 Sites of sinus mucocoeles.

Anatomical site	n
Anterior ethmoid	36
Frontal	32
Maxilla	11
Posterior ethmoid	11
Sphenoid	5

The number of mucocoeles exceeds the number of patients as some patients presented multiple sites.

Patients and methods

This retrospective study was based on a review of the medical charts of all patients operated for sinus mucocoele between January 1993 and December 2009: 68 patients with a mean age of 53 years (range: 27 to 82 years) with a sex ratio of 3/2. The number of mucocoeles operated has increased since January 2000: 18 cases were reported before 2000, 50 cases were reported after January 2000. The patient's history, presenting symptoms, clinical and radiological signs and treatment were analysed.

Fifty-one (75%) of these 68 patients had a history of sinus surgery. The main indications, type of operation and interval between surgery and the diagnosis of mucocoele are shown in Table 1. Among these 51 patients, 23 (45%) had been operated for nasal polyposis, 13 for sinus infection, and seven for inverted papilloma. Medical history included a history of head injury in four patients and non-operated chronic sinusitis in five patients. Among the patients, 15% had no medical or surgical history. The mean interval between surgery and the diagnosis of mucocoele was 7 years (Table 1). When mucocoeles of traumatic origin were added to this subgroup, the mean interval between head injury or surgery and the diagnosis of mucocoele was 9 years (n = 55, standard error: 1.4). The interval between surgery and mucocoele according to the type of surgical procedure is shown in Table 1.

The circumstances of discovery of mucocoele were variable: an incidental finding in 14 asymptomatic patients (20.6% of patients) during radiological or fibroscopic examinations performed for surveillance sinus disease; facial pain or headache in 26 patients (38%); and finally, mucocoele was revealed by a complication in 28 patients (41%); these complications were orbital (palpebral swelling, orbital cellulitis, diplopia) or ocular (loss of visual acuity) (28% of patients). An ophthalmological consultation was requested

in patients with ocular symptoms. Malar or palpebral swelling ($n=7$, 10.3% of patients) or sinus infection ($n=2$) were reported more rarely.

All patients were assessed by CT and MRI of the facial bones in order to confirm the diagnosis and define the anatomical relations with adjacent organs (Figs. 1 and 2). The anatomical sites of mucoceles are presented in Table 2. Seven patients presented multiple unilateral or bilateral mucoceles. On MRI, the mucocele presented a low-intensity signal on T1-weighted sequences with fine peripheral enhancement and homogeneous and intense high-intensity signal on T2-weighted sequences. The MR signal varied according to the age of the lesion. The high-intensity signal on T2-weighted sequences consequently decreased and became more heterogeneous with time, reflecting the higher protein content of the lesion [10] (Fig. 2).

All patients were treated by endonasal endoscopic surgery, systematically associated with computer-assisted navigation after January 2003. Mucoceles detected in a context of acute infection were treated by antibiotics and corticosteroids for 48 to 72 h prior to surgery. Bacteriological examination of the mucocele contents was not performed. Fifty-seven patients (83.8%) were treated by exclusive endonasal endoscopic surgery under general anaesthesia. Treatment consisted of marsupialization of the mucocele while preserving the mucocele mucosa. When opening of the mucocele was limited for anatomical reasons (narrow frontal recess region, for example), the orifice was maintained by a piece of rolled silastic left in place for 3 weeks. A combined surgical approach was necessary in 11 patients (16.2%): nine endonasal approaches completed by a superciliary incision for lateral or very large frontal lesions, and two bicoronal incisions in patients with a history of craniostenosis.

Postoperative care was limited to irrigation of the nasal cavities with physiological saline several times a day for three to four weeks. When paranasal sinus inflammation was observed intraoperatively, mainly in patients with nasal polyposis, a short course of oral corticosteroids was administered (prednisone: 1 mg/kg/day for 7 days), followed by long-term local corticosteroids. Follow-up comprised regular and prolonged clinical endoscopic examinations (D8, D15, M1, M3, then every 6 months for 2 years then annually thereafter). Follow-up CT was systematically performed every 2 to 3 years or when there was a clinical suspicion of recurrence. The mean postoperative follow-up was 7 years (range: 4 months-16 years).

Results

Three patients developed immediate postoperative complications: an intraorbital haematoma, a frontal abscess (*Pneumosinus dilatans*) and cerebro-spinal fluid (CSF) leak (craniostenosis). During postoperative follow-up (mean follow-up: 7 years), 16 cases (23.5%) of recurrences or second mucoceles were detected, with a mean interval of 4 years after resection of the first mucocele (range: 4 months-41 years). Ten of the 16 patients with recurrent mucocele had a history of nasal polyposis (62.5% of recurrences). In eight of these patients, nasal polyposis was poorly controlled (severe symptoms, frequent need for oral corticosteroids, numerous episodes of acute

superinfection). This difficult control was related to either an aggressive form of nasal polyposis despite well-conducted medical treatment and good compliance, or poor compliance with treatment. Six of these recurrences were detected incidentally during follow-up in the absence of any symptoms suggestive of mucocele.

Discussion

This study was based on retrospective review of the medical charts of 68 patients operated for sinus mucocele between 1993 and 2009, which represents one of the largest single-centre series published in the literature [8-12] (Table 3). More than 80% of patients were operated exclusively by endonasal endoscopic surgery. As a result of progress in endoscopic techniques and intraoperative navigation, most lesions are now accessible via a minimally invasive approach, which is both more conservative and less aggressive than conventional external surgery. Treatment of mucocele is based on marsupialization, enlarging the usual drainage pathways, while sparing the non-neoplastic mucosa [3]. The periosteum left in place allows osteogenesis and bone remodelling [1]. The development of intraoperative navigation has significantly extended the indications for endoscopic surgery, allowing both detection and opening of certain mucoceles with difficult anatomical access [13-14]. The present series confirms the limitations of an exclusively endoscopic approach for lateral or extensive frontal lesions, which required a combined approach (16.2%). These results are concordant with those of several authors who consider that open surgery is preferable in these anatomical sites. The main subject of debate at the present time concerns fronto-ethmoidal mucoceles, which are both the most frequent sites and also those with the highest morbidity and recurrence rates [15]. This predominance of fronto-ethmoidal mucoceles was confirmed in the present series, although the reasons for this predominance cannot be explained at the present time. Nevertheless, although our epidemiological, clinical and radiological data are globally comparable to those of the literature (Table 3) with more than 70% of fronto-ethmoidal mucoceles, the recurrence rate (23%) was higher in the present series. What are the possible explanations for this difference?

The first explanation is related to the duration and modalities of follow-up, as the mean time to onset of recurrence was 4 years. Detection of recurrence therefore requires a long follow-up, and the mean follow-up in this series was 7 years, about twice that reported in the other series, which could explain the higher recurrence rate in our series. One half of these recurrences were also discovered incidentally during systematic follow-up. These asymptomatic lesions would not have been detected in the absence of this regular clinical and radiological follow-up. The present series also reveals that the time to onset of recurrences can be as long as 41 years, indicating the need for very long-term follow-up of these patients. This long follow-up explains the higher recurrence rate observed in this series compared to other series with a much shorter mean follow-up [16].

The high percentage of patients with nasal polyposis in this series could also help to explain this high

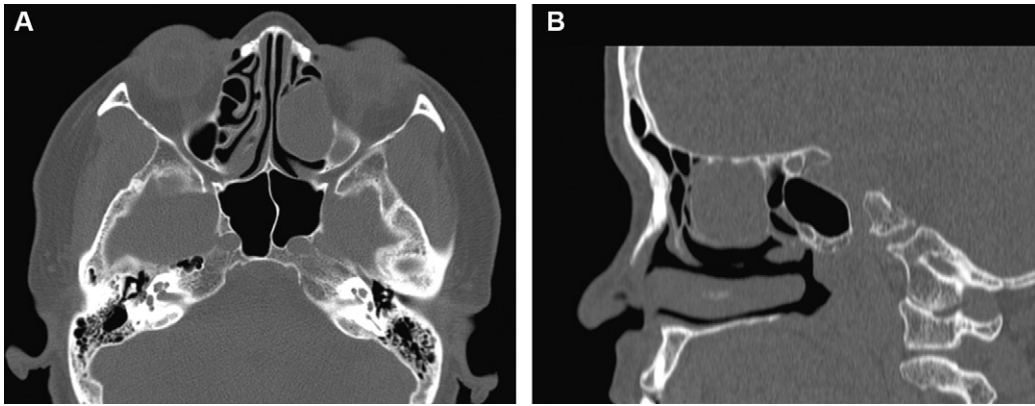


Figure 1 CT scan of facial bones (A. Axial, B. Sagittal). Typical radiological appearance of ethmoidal mucocele visualized as a convex space-occupying opacity with thinned and eroded bone walls displacing adjacent structures with a homogeneous content.

recurrence rate, as these patients more frequently require surgery than patients with other paranasal sinus diseases. Among patients of this series, 33.8% had nasal polyposis and 45% of the patients with a history of surgery had been

operated for nasal polyposis. Furthermore, two-thirds of recurrences (i.e. 10 out of 16 patients) concerned patients with particularly aggressive and/or previously operated nasal polyposis. These findings support the hypothesis of

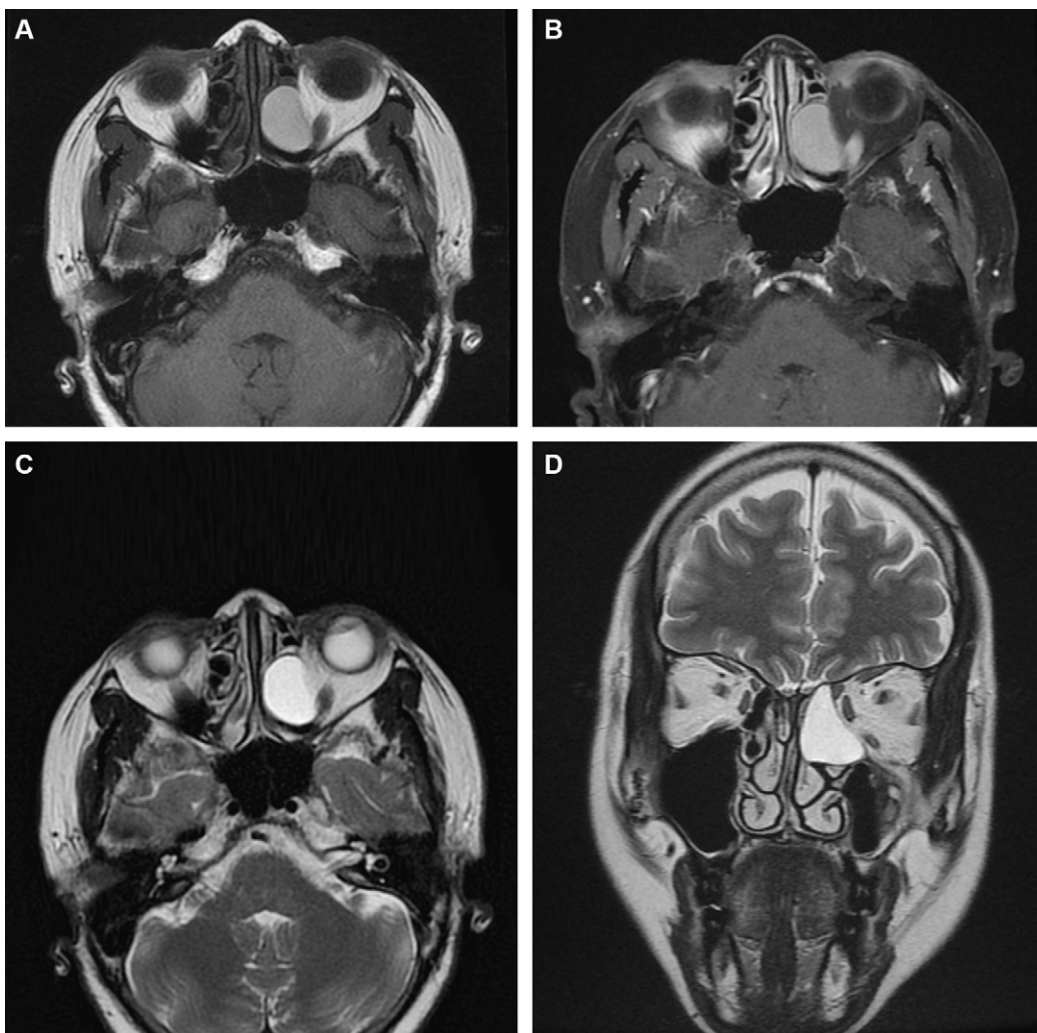


Figure 2 MRI of facial bones (A. Axial T1, B. Axial T1 gadolinium, C. Axial T2, D. Coronal T2). The mucocele (same as in Fig. 1) has a high-intensity signal on T1-weighted sequences and a homogeneous and intense high-intensity signal on T2-weighted sequences.

Table 3 Comparative review of the literature.

Author	Raynal et al.	Rombaix et al.	Har-El	Serrano et al.	Facon et al.	Present series
Reference	[8]	[10]	[11]	[6]	[9]	
Number	42	140	103	60	52	68
Mean age	45	46		53	49	53
Years	87–97		98–00	87–95	93–01	93–09
% of anterior fronto-ethmoidal lesions	69	64	66	66	54	73
Medical history						
Nasal polyposis (%)	17		22	15	36	34
Infection (%)	7			10	27	20
Tumour (%)	1			2	4	
Trauma (%)	4	2		1	4	4
Surgical history (%)	53	63		45	38	74
No surgical history (%)	21	35		45	38	15
Endonasal approach (%)	62	94		55	87	83
Transfacial approach (%)	12	6		23	10	16
Interval between surgery and diagnosis (months)	22	24		24		84
Post-op follow-up (years)	3.5	4.6	4.5	4	7	
Recurrence rate (%)		2.2	0.9	5	3.8	23

the role of inflammation and the role of trauma in the pathophysiology of mucoceles. Lund et al., in 1993, suggested this role of inflammation by demonstrating release of prostaglandin PGE2 and proinflammatory cytokines (IL-1, TNF-alpha) into the wall of the mucocele, inducing osteoclastic bone resorption [1]. Similarly, Jankowski et al. showed that the incidence of spontaneous mucocele was higher in a population of patients with nasal polyposis (0.6%) than in the general population and that the incidence of mucocele increased to 2.5% in patients with nasal polyposis after nasalisation [17,18]. Patients operated for nasal polyposis therefore constitute one of the populations at highest risk of mucocele, constituting a clinical model comprising both inflammatory and traumatic mechanisms.

Conclusion

This study demonstrates the high recurrence rate of mucocele, particularly in patients with multi-operated, chronic paranasal sinus inflammation. It also confirms the efficacy of endonasal surgery for the treatment of mucoceles. The indications for this surgery have been considerably extended, especially due to progress in intraoperative navigation.

The prevention and detection of recurrent mucocele constitute major challenges in the management of this disease, which is why we recommend regular and prolonged clinical follow-up in order to detect lesions while they are still asymptomatic, before the onset of complications. In the context of this follow-up, CT scan should be performed every 2 years for a minimum of 4 or 5 years, especially when the

ethmoidectomy cavity is difficult to examine (inflammation, synechiae). This clinical and radiological follow-up must be even more rigorous in patients with inflammatory sinus diseases, such as nasal polyposis.

Local control of postoperative inflammation plays a very important role in terms of prevention, as inflammation appears to be one of the major factors predisposing to the formation of mucocele.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

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