

Studies on mucocoeles of the ethmoid and sphenoid sinuses: Analysis of 47 cases

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Abstract

Forty-seven mucocoeles of the ethmoid and/or sphenoid sinuses (33 males, 14 females) were operated on during the 10-years period from 1980 through 1989. Thirty-seven cases were post-operative mucocoele, while the remaining 10 were so-called primary mucocoeles. None of the cases had a history of facial trauma. In the majority of post-operative cases, the mucocoele develops 15–24 years after initial surgery. Paranasal sinus surgery in young patients (teenagers) may lead to a mucocoele due to post-operative scarring in the surgical wound. The principal symptoms include globe displacement, double vision, headache, deep orbital pain, a mass in the supero-medial quadrant of the orbit, visual disturbance, *etc.* Sufficient opening of the mucocoele wall by the endonasal approach is recommended for surgical treatment of ethmoidal and sphenoidal mucocoeles.

Introduction

Mucocoeles of the ethmoid and sphenoid sinuses, which cause globe displacement, headache, ophthalmalgia, double vision, visual impairment and other symptoms, are often encountered in everyday clinical practice. The incidence of post-operative mucocoele is higher in Japan than that of so-called primary mucocoele which develops in patients with no history of nasal surgery. Because the primary symptoms of ethmoidal/sphenoidal mucocoeles are ophthalmological, most patients first visit ophthalmologists and are referred to otorhinolaryngologists after diagnosis of the disease. However, due to the recent progress in imaging, even mucocoeles which are difficult to diagnose by simple/tomographic X-rays have become easier to diagnose by CT scan in the early stages of the disease. Thus, it has become possible to initiate treatment of this disease before it becomes serious.

Recent advances in surgical techniques mean that, it is currently possible to achieve cure by sufficient opening of the wall of the mucocoele by an endonasal approach. In the past, the optic canal in the mucocoele and cranial base, could not be adequately observed with the naked eye. However, such structures can now be accurately observed using an endoscope, and it is thus possible to marsupialize the lesion safely and accurately.

There are differences between Japan and other countries in the method and number of paranasal operations. Accordingly, it is thought that there are pathological features in ethmoidal/sphenoidal mucocoeles, unique to the Japanese. We investigated in particular the aetiology, of post-operative mucocoeles on the basis of the clinical findings for ethmoidal/sphenoidal mucocoeles which have been treated in the past decade.

Method and materials

There were 47 patients (49 lesions) with mucocoeles of the ethmoid and/or sphenoid sinuses who were operated on during the 10-year period from 1980 to 1989. Thirty-seven of them had previously been operated for sinusitis, while the remaining 10 had so-called primary mucocoeles. None of the cases had a history of facial trauma. The cases were reviewed in relation to the sex, age, surgical method used for past sinusitis, time from the past operation to the development of symptoms of mucocoele, intranasal findings, location of the mucocoele, clinical symptoms and the surgical method of treatment. In addition, the mechanism of development of post-operative mucocoeles was investigated.

In two cases, the mucocoele had expanded, destroyed the nasal septum and invaded the contralateral ethmoid sinus. Only one case had undergone more than one operation before the mucocoele development. Mucocoeles were discovered incidentally in three cases by CT scan performed in search for the cause of dizziness or as part of a physical examination.

Results and discussion

1. Sex and age of subjects

The subjects consisted of 33 males and 14 females. The post-operative mucocoele cases included a larger number of male patients (30) than female patients (7). This is because the incidence of paranasal sinus surgery is generally higher in males than females in Japan. Nugent *et al.* (1970) and Natvig and Larsen (1978), on the other hand, did not find any sex difference in the incidence of mucocoeles of the paranasal sinuses in their

studies from the USA and Scandinavia, perhaps reflecting a larger number of so-called primary mucocoeles compared with our study population.

The age of the 46 patients (one patient who had undergone more than one operation in the past was excluded) ranged from 20 to 69 years. The mean age was 46.2 years (44.4 years for the post-operative mucocoele cases, and 52.4 years for the primary mucocoele cases), with the peak in the fifth decade, and two-thirds of the 46 cases consisted of patients in their fourth and fifth decades. This is related, to the time from the age at the first nasal operation to the onset of symptoms of the mucocoele. In this regard, Nugent *et al.* (1970) reported that the age range of their cases was 13–75 years, with a mean of 42 years, while Natvig and Larsen (1978) reported that 75 per cent of their cases were between 40 and 70 years.

2. Age at first operation and surgical method

For 36 post-operative mucocoele cases (excluding one case which had undergone more than one operation), the mean age at the time of the first nasal operation was 21.7 years, with the peak at 15–19 years; most patients underwent the first nasal operation before 25 years of age and 26 cases (72.2 per cent) underwent the first nasal operation when they were teenagers (Fig. 1). This suggests that performance of a nasal operation at a young age is a factor in the development of a mucocoele.

The previous surgery consisted of a transmaxillary paranasal operation combined with an endonasal operation were performed in 33 cases (91.7 per cent). However, in one-third of them, unresected cells were clearly seen in the anterior ethmoidal region, suggesting inadequate endonasal treatment. Three cases (8.3 per cent) had been treated only by endonasal surgery.

3. Time from first nasal operation to presentation with mucocoele

This analysis covered the same 36 post-operative mucocoele cases, excluding the one multiple-operation case. This ranged from 11 to 49 years (mean time 22.7 years). The cases in which the interval was greater than 15 years but less than 24 years accounted for 58 per cent of the 36 cases, indicating that the symptoms of the

mucocoele developed quite a long time after the first operation (Fig. 2). From this it would appear that, in Japan, most patients with a post-operative mucocoele were operated on for sinusitis between, approximately, 10 to 25 years of age and manifested symptoms of a mucocoele 15 or more years after that operation. Also, for fronto-ethmoidal mucocoeles, Lund (1987) reported that the time interval was 14–57 years and the average time was 29 years in the case of surgery for infection.

4. Endonasal findings

The endonasal findings were reviewed for all 47 patients (49 lesions). Using anterior rhinoscopy, polyps were observable in the middle meatus in nine cases (18.4 per cent), adhesions in the middle meatus in 15 cases (30.6 per cent), and the anterior wall of the mucocoele in nine cases (18.4 per cent). In 16 patients (32.7 per cent), however, the endonasal findings were normal or showed only slight pathological changes, such as oedema of the middle meatus, and, on the whole, only a few cases had severe sinusitis (Table I). In contrast, half of the patients with a primary mucocoele had polyps. Natvig and Larsen (1978) also stated that chronic sinusitis was found in approximately half of the patients they treated. Similarly, Canalis *et al.* (1978) reported that polyps were found in more than half of 20 patients with an ethmoidal mucocoele. In both the patient groups treated by Natvig and Larsen and Canalis *et al.*, the majority had a primary mucocoele.

The relationship between deviation of the nasal septum and the mucocoele was studied in 39 of the 47 patients, excluding two patients in whom the mucocoele extended over the right and left sinuses and six patients in whom two mucocoeles were present bilaterally. In only four patients, clear nasal septum deviation, including deviation of the superior region, was found on the ipsilateral side. In the remaining 35 cases, either no clear deviation was seen as a result of past surgery on the nasal septum, or the nasal septum curved to the contralateral side. Accordingly, it can be said that there is no relationship between the development of a mucocoele and nasal septal curvature.

5. Location of mucocoele

Most mucocoeles were unilocular, but multilocular

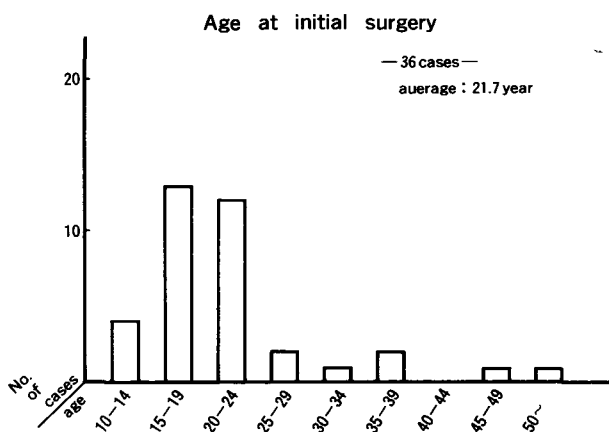


FIG. 1
Age at initial sinus surgery.

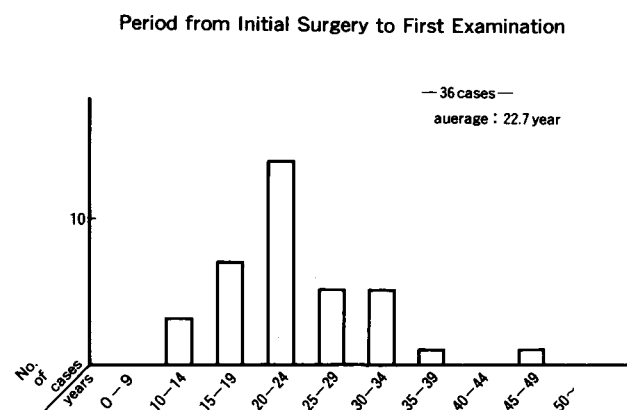


FIG. 2
Period from initial surgery to first examination.

TABLE I
NASAL FINDINGS (49 EXAMPLES)

Almost normal and slight change	: 16 examples
Polyp in middle meatus	: 9 examples
Adhesion of middle meatus	: 15 examples
Visible cyst wall	: 9 examples

mucocoeles (existence of two or three mucocoeles separated by mucosa or lamella) were seen in four cases. In addition, in two cases, the ethmoidal mucocoele had destroyed the nasal septum and extended over both sides of the sinus. A mucocoele (including frontal sinus mucocoele) was present bilaterally in six cases (Fig. 3).

The incidence of mucocoeles extending from the anterior and posterior ethmoidal sinus is generally high, but mucocoeles in the posterior sinus or sphenoidal sinus are not rare (Fig. 4). In the literature, mucocoeles are said to be more common in the anterior ethmoidal sinus. However, in our present study, half of the mucocoeles were present in the posterior ethmoidal sinus or sphenoidal sinus (Table II). A mucocoele of the anterior ethmoidal sinus affects the frontonasal, duct, or recess and, sooner or later, it causes an obstructive lesion in the frontal sinus. A mucocoele in the posterior ethmoidal sinus also affects the natural ostium of the sphenoidal sinus if it grows larger, and it eventually causes a mucocoele of the sphenoidal sinus. Most bicyclic mucocoeles are present in those regions, *i.e.*, the posterior ethmoidal and the sphenoidal sinuses.

6. Clinical symptoms

Clinical symptoms of a mucocoele occur due to compression of the adjacent organs, such as the orbit and base of the skull, by the enlarged mucocoele itself or due to spread of the inflammation. The principal symptoms include globe displacement, double vision, headache, deep orbital pain, a mass in the supero-medial quadrant of the orbit, visual disturbance (dysopsia), *etc.* (Fig. 5). In our present study, 15 cases (31.9 per cent) complained of pain, such as headache and ophthalmalgia. McCarthy *et al.*, reported that 53 of 72 cases of sphenoidal mucocoeles (73 per cent) had pain (circumocular pain: 40 per cent; headache: 33 per cent).

Nasal symptoms if any are generally absent or mild. Some patients complain of nasal obstruction and/or

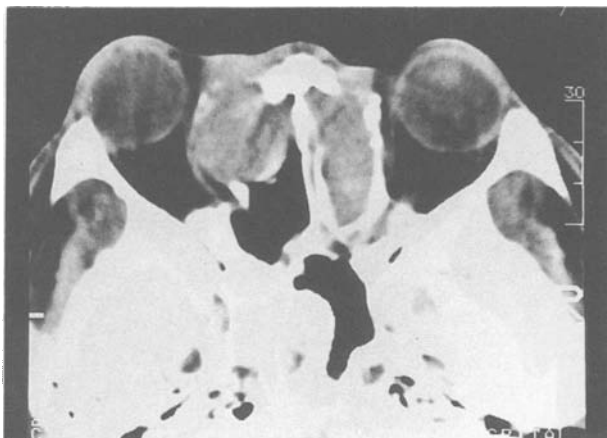


FIG. 3

A mucocoele was present in each of the right and left sinuses.

olfactory disturbance. Canalis *et al.* (1978) pointed out that ocular symptoms occurred most frequently. Double vision: Nugent *et al.* (1970) stated that double vision was due to impairment of the oculomotor and abducens nerves in 12/19 cases, and to impairment of the abducens nerve in the remaining seven. McCarthy *et al.* (1972) reported that eye movement disorders were due to impairment of the oculomotor nerve in the remaining eight cases. In the present study, diplopia occurred in five cases, and in all these it was attributable to ocular deviation due to globe displacement, rather than nerve impairment. Post-operative diplopia never occurred in our series.

Naturally, the clinical symptoms differ with anterior ethmoidal mucocoeles (Fig. 6), globe displacement (48.7 per cent in the present study), diplopia, ophthalmalgia, *etc.* are the major symptoms. In the case of a mucocoele located between the posterior ethmoidal and sphenoidal sinuses (Fig. 7), the mucocoele may directly destroy the optic canal and cause visual disturbance; this may lead to loss of eyesight in severe cases. In our present study, the optic canal was destroyed, causing exposure of the optic nerve, in 13/14 cases with visual disturbance. In one case, protrusion of the optic nerve could not be confirmed in the mucocoele. In that case, we speculate that, rather than direct compression of the optic nerve by the mucocoele, mild secondary visual disturbance occurred due to a circulatory disorder around the optic nerve in the orbit as a result of expansion of the mucocoele.

7. Medical department first visited by patients

Because ophthalmological symptoms were the first and primary symptoms, 33 patients first visited a department of ophthalmology, nine patients first visited a department of otorhinolaryngology, and one first visited a department of neurosurgery due to severe headache. Of the 10 cases with a primary mucocoele, nine first visited a department of ophthalmology. In the case of a

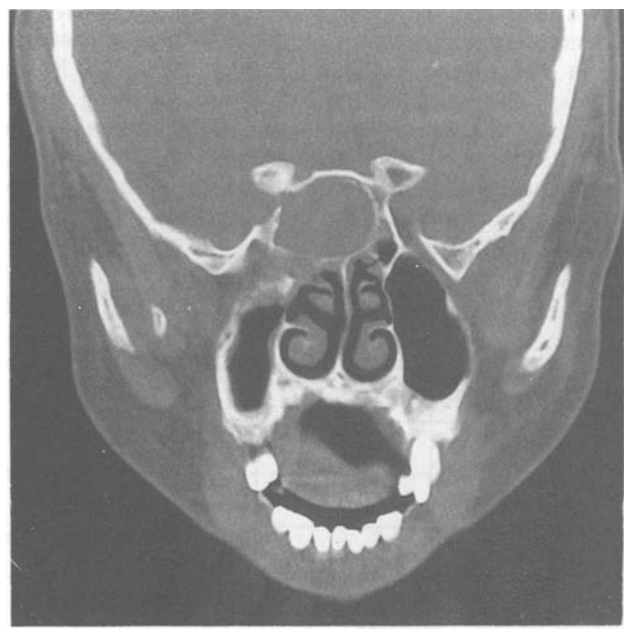


FIG. 4

Coronal CT scan of a patient with sphenoidal mucocoele.

TABLE II
LOCATION OF CYST (47 CASES, 49 EXAMPLES)

	Post-operative	Primary
AE :	9	0
AE + PE :	8	9
PE :	7	1
PE + S :	7	0
S :	5	3

post-operative mucocoele, on the other hand, even when the symptoms were ophthalmological, there was a tendency for the patients to first visit a department of otorhinolaryngology, probably as they had undergone surgery for sinusitis in the past.

8. Mechanism of development of post-operative mucocoele

According to studies reported in Europe and the USA, the incidence of post-operative mucocoeles developing in patients who had undergone surgery for sinusitis is low, while the incidence of primary mucocoeles is high. Natvig and Larsen (1978) reported that only 11 (10 per cent) of 112 cases with a mucocoele had undergone previous nasal surgery. In Japan, the incidence of mucocoele patients undergoing paranasal surgery is high (Moriyama *et al.*, 1981) probably due to the large number of patients with chronic sinusitis. We speculate that this fact is the reason for the higher incidence of post-operative mucocoeles in Japan than in Europe and the USA. In this connection, the mechanism of post-operative mucocoele development is speculated upon below.

The present study revealed that a large percentage of post-operative mucocoele cases had undergone their first nasal operation when they were young, especially in their teens; this is a finding worth of special note. The post-operative changes, such as narrowing due to cicatrization, adhesion and bone hyperplasia, are strongly related to mucocoele formation. In view of the fact that post-operative mucocoele has a high incidence in patients showing a strong tendency to fibrotic cicatrization or adhesion, we speculate that there may be an unexpectedly low incidence of patients showing severe pathological changes. If adhesion of the middle meatus occurs due to lateral deviation of the middle turbinate, the anterior and posterior ethmoid sinuses become isolated, and the basis for development of an anterior/posterior mucocoele is formed.

symptom mucocoele	exophthalmus	swelling	double vision	eye pain	headache	visual disturbance
AE	○ ○	○ ○ ○ ○ ○	○	○ ○	○	
AE+PE	○ ○ ○ ○ ○ ○ ○ ○ ○ ○ ○ ○ ○ ○ ○	○ ○	○ ○ ○	○ ○ ○		○
PE	○ ○	○		○	○ ○	○ ○ ○ ○
PE+S	○		○		○ ○	○ ○ ○
S				○ ○ ○	○	○ ○ ○ ○ ○ ○

AE : Anterior ethmoid sinus
PE : Posterior ethmoid sinus
S : Sphenoid sinus

FIG. 5

Clinical symptoms of a mucocoele.

The changes in the surgical wound after an operation for sinusitis differ with the severity of the disease and surgical method employed. If the ethmoid sinus is insufficiently treated, that is if inflammatory cells or the ground lamella are left unremoved, local infection may occur again even if temporary healing is achieved. As a result, due to subsequent disturbance of the ciliary movement and/or endonasal airflow, an obstructive lesion may develop due to granular proliferation, fibrosis. Moreover, even when a healed surgical cavity in the ethmoids is achieved, there is a possibility of formation of a closed cavity due to long-term post-operative changes in the region, such as adhesion, cicatrization.

9. Treatment

It is not necessary to completely marsupialize the ethmoidal and sphenoidal mucocoele, but it is absolutely necessary to open the mucocoele in order to establish routes for drainage of the contents. That is, permanent healing can be achieved by sufficiently opening the anterior and posterior walls of the mucocoele by an endonasal approach. Not all mucocoeles are amenable to an endonasal approach, although ethmoidal and sphenoidal mucocoeles are the most accessible. The operation can be performed even more safely if objects are accurately observed using an endoscope (Kennedy *et al.*, 1989). The mucocoele will not recur if sufficient of the wall is removed. In fact, during follow-up for one to 10-year periods, recurrence has not been seen in any of the patients included in the present study.

Some investigators (Canalis *et al.*, 1978; Close and Oconner, 1983) report the occurrence of post-operative complications, such as transient leakage of CSF or orbital cellulitis after adequate excision of the mucosa of the mucocoele wall by an extranasal or transantral approach. However, there have been no such complications reported after endonasal opening of the mucocoele. Thus, this method can be recommended. Long-term post-operative follow-up is necessary for cleaning of the area during healing to maintain patency.

A CT scan is necessary for adequate visualization of the anatomy distorted by previous surgery and pathology. A mucocoele is not always unilocular; multilocular mucocoeles containing septa can occur. It is therefore important to confirm the positional relationship



FIG. 6

Axial CT scan showing extensive ethmoidal mucocoele.

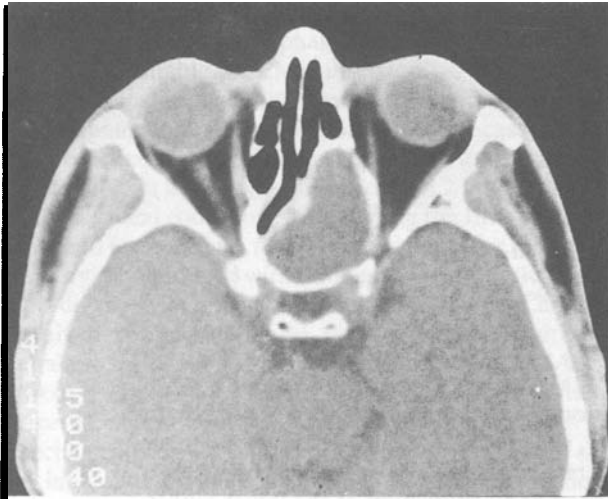


FIG. 7

Axial CT scan of patient with sphenoidal mucocoele accompanied by visual disturbance.

between the mucocoele and the surrounding structures by pre-operative CT imaging. In addition, the anterior wall of the mucocoele is not always as thin as a membrane. Moreover, even in the case of a mucocoele whose anterior wall is hard and thick due to bony hyperplasia, the floor of the anterior cranial fossa or the bony wall of the optic canal is absent in some patients.

Conclusion

On the basis of the results of a clinical study on 47 patients (49 lesions) with a mucocoele in the anterior ethmoid, posterior ethmoid or sphenoid sinuses who were treated during the 10-year period from 1980 to 1989, the authors emphasize the following points:

1. In about half of mucocoele cases, the middle meatus is normal or shows only slight changes, such as mild oedema. Although the incidence of adhesion of the middle meatus is high, the incidence of polyposis is low.

Key word: Mucocoele

2. Many mucocoele cases are post-operative, *i.e.*, they had previously undergone anasal for sinus surgery. In most such cases, the mucocoele develops 15–24 years later.
3. Many mucocoele patients first visit a department of ophthalmology. Thus, ophthalmologists should be thoroughly conversant with this disease.
4. Some mucocoeles are multilocular (2–3 compartments), and thus it is important to check the anatomy by pre-operative CT scanning.
5. An endonasal approach with sufficient opening of the mucocoele wall is recommended for surgical treatment of ethmoidal and sphenoidal mucocoeles.
6. In order to prevent development of a post-operative mucocoele, it is important to sufficiently resect the ground lamella and lamellae in the ethmoid sinus at the time of the primary paranasal sinus surgery.

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