ORIGINAL ARTICLE

Clinical Analysis and Surgical Results of 58 Paranasal Sinus Mucoceles

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Received 19 April 2014; accepted 4 June 2014

KEYWORDS
Mucoceles; Paranasal sinuses; Surgical management; Results

Abstract

Introduction: Mucoceles are slow-growing, benign lesions found in the paranasal sinuses that are locally destructive, causing bony resorption and displacement of adjacent structures. We present our experience in the surgical treatment of these lesions.

Methods: This was a retrospective review of 58 paranasal sinus mucoceles in 54 patients between 1989 and 2012. We describe patient age and sex, mucocele location, clinical features, surgical approaches employed, recurrence and complications.

Results: The mean age of patients in this series was 59 years; there were 31 males (57%) and 23 females (43%). Thirty-two cases (55%) were located in the frontal or ethmoid–frontal system, 8 (14%) in the ethmoid sinus, 14 (24%) in the maxillary sinus and 4 (7%) in sphenoid sinus. Predisposing factors were present in 55% of the patients and 45% cases were primary. Endoscopic treatment was given to 71% of mucocele patients, while 29% were treated with external or combined approaches. Recurrence appeared in 4 patients (7%), 2 in the endoscopic surgery group and 2 in the external surgery group.

Conclusions: The procedure of choice for management of paranasal sinus mucoceles is endoscopic drainage. It is a safe approach that gives good results.

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Palabras clave
Mucoceles; Senos paranasales; Manejo quirúrgico; Resultados

Análisis clínico y resultados quirúrgicos en 58 mucoceles nasosinusales

Resumen
Introducción: Los mucoceles son lesiones benignas que afectan a los senos paranasales, de crecimiento lento, con capacidad de reabsorción ósea. Exponemos nuestra experiencia en el tratamiento quirúrgico de estas lesiones.

Métodos: Se realizó un estudio retrospectivo de 58 mucoceles nasosinusales diagnosticados en 54 pacientes entre los años 1989-2012. Se analizaron las diferentes localizaciones, distribución por edad y sexo, características clínicas, tipo de abordaje quirúrgico, recidivas y complicaciones.

Resultados: La edad media de los pacientes en el momento del diagnóstico fue de 59 años. El 57% de los pacientes (31/54) fueron varones y el 43% (23/54) mujeres. En el 55% de los casos (32/58) el mucocele estaba localizado en el seno frontal o en la región frontoetmoidal, un 14% (8/58) en el seno etmoidal, un 24% (14/58) en el seno maxilar y un 7% (4/58) en seno esfenoidal. En el 55% de los casos se identificó algún factor predisponente, siendo el 45% mucoceles primarios. El 71% de los pacientes fueron tratados con un abordaje endonasal endoscópico y en el 29% se realizó un abordaje abierto o combinado. Observamos un total de 4 recidivas (7%), 2 en el grupo abordaje endonasal endoscópico y 2 en el grupo tratado con cirugía abierta.

Conclusiones: El tratamiento de elección de los mucoceles nasosinusales es la marsupialización por vía endonasal endoscópica, siendo una técnica segura y con buenos resultados.

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Introduction

Mucoceles are pseudocystic, expansive, slow-growing formations. They are benign processes which affect the paranasal sinuses. The external wall comprises the healthy mucosa of the sinus cavity itself and the content is an aseptic fluid which when superinfected is termed mucopyoce. To expand they complete a dynamic process of bone erosion–resorption.

They were first described as hydatides by Langenbeck in 1818 and Rollet suggested the term mucocele for this pathology in 1896. Onodi made the first histological description in 1901.1–3

Their etiopathogenesis is unknown, although it seems likely that ostium drainage obstruction and sinus inflammation need to coincide for a mucocele to form. There are two theories to explain their formation: the necrosing pressure theory, where the pressure exerted by mucus in an obstructed sinus causes necrosis of the underlying bone,4 and the inflammation theory, where activation of leukocytes and lymphocytes is observed producing inflammatory mediators (IL-1, IL6),5,6 which in turn cause the activation of interleukins (PGE2, collagenase),7 all of which promotes bone resorption and bone remodelling for the mucocele to expand. Mucoceles appear in patients between the 4th and 7th decade of life. There are no differences in terms of gender. They are generally unilateral and principally affect the ethmoid–frontal complex (60%–89%).8 The clinical symptoms depend on the location and the size of the mucocele. Ocular symptomatology predominates in anterior ethmoid–frontal locations whereas jugal and vestibular swelling is the most common presentation sign in maxillary mucoceles. Neurological signs can be seen in very developed mucoceles and are found relating to cerebral compression and/or compression of the optic or cranial nerve.9 These lesions are treated surgically in order to marsupialise the mucocele to the nostril. The objective of our study is to describe the epidemiological and clinical characteristics of these lesions and to analyze the different types of surgical approaches used, and the results in terms of recurrences and possible complications.

Methods

A retrospective study was undertaken on 58 mucoceles diagnosed in 54 patients between January 1989 and January 2012. The different locations, distribution by age and gender, predisposing factors, clinical characteristics, localization, type of surgical approach, recurrences and postoperative complications were analyzed. Minimum follow-up was 12 months. Diagnosis was based on the patient’s clinical history, endoscopic exploration and on imaging tests. All the patients underwent computerized tomography prior to surgery, to assess the location and extension of the mucocele; this was complemented with magnetic resonance imaging in cases where there was extension to the orbit or the base of the skull.

Results

The mean age of the patients at the time of diagnosis was 59 with a range between 12 and 93 years of age. 57% of the patients (31/54) were males and 43% (23/54) were females. Four patients presented 2 independent mucoceles in different locations; therefore the total number of diagnosed
mucoceles was 58. The anatomopathological result of 11 of them was mucopyoce.

In 55% of the cases (32/58) the mucocele was located in the frontal sinus or in the ethmoid–frontal region, in 14% (8/58) in the ethmoidal sinus, in 24% (14/58) in the maxillary sinus and in 7% (4/58) in the sphenoid sinus. Fig. 1 shows the principal locations.

45% (26/58) were primary mucoceles, whereas some risk factor was identified in 55% (32/58) of the cases. The 32 secondary mucoceles were: 20 patients with a surgical history, 6 patients diagnosed with concomitant paranasal sinus polyposis which had not been operated, 3 patients with a history of previous facial trauma with fracture, 2 patients had a mucocele secondary to an osteoma and one patient had a mucocele secondary to an inverted papilloma.

Table 1 shows the relationship between surgical history and location.

The mean latency time between the history of previous surgery and diagnosis of the mucocele was 16 years (range between 5 and 32 years).

Table 2 shows the most common clinical presentation according to location.

All the mucoceles were treated surgically, 41 using an endoscopic endonasal approach, 7 using an external approach, and 10 mucoceles using a combined approach. Table 3 shows the type of approach according to location.

No major postoperative complications arose. Two patients experienced epistaxis in the period immediately after surgery that resolved with nasal packing. Collapse of the anterior diploë was seen in 2 patients with frontal mucoceles.

Recurrence was diagnosed in 4 of the cases (7%), all located in the frontal sinus: after endoscopic endonasal route in 2 of the patients and after an external approach in the other 2.

The latency period between surgery and recurrence varied between 6 and 17 years.

Discussion

Mucoceles are benign lesions which most frequently affect the ethmoid–frontal complex, followed by the ethmoidal sinus and less frequently the maxillary sinus. Our case studies coincide with other literature in that more than half of the cases are located in the ethmoid–frontal region. However, the percentage of maxillary mucoceles was higher than that published in most series (24% vs 10%).

This finding is probably due to the greater frequency of Caldwell-Luc procedures performed in the 1980s, primarily in the treatment of maxillary rhinosinusitis. Seven percent of our cases were diagnosed in the sphenoid sinus. Har-El et al. presented similar data finding 11% sphenoid mucoceles and Rombaux et al., in a series of 178 mucoceles, found 8% sphenoid mucoceles. Exceptionally, cases have been described in the concha bullosa, in

Table 1 Relationship Between Surgical History and Location of the Mucocele.

<table>
<thead>
<tr>
<th>Location of Mucocele</th>
<th>ENS</th>
<th>External</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Frontal/ethmoid-frontal</td>
<td>4</td>
<td>2(^{a})</td>
<td>6</td>
</tr>
<tr>
<td>Ethmoidal</td>
<td>2</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>Maxillary</td>
<td>1</td>
<td>8(^{b})</td>
<td>9</td>
</tr>
<tr>
<td>Sphenoid</td>
<td>3</td>
<td>0</td>
<td>3</td>
</tr>
<tr>
<td>Total</td>
<td>10</td>
<td>10</td>
<td>20</td>
</tr>
</tbody>
</table>

\(^{a}\) Subciiliary approach.

\(^{b}\) Caldwell-Luc approach.
the lateral sphenoid recess and the orbital floor (Haller cell).\textsuperscript{14}

Several predisposing factors have been identified such as a history of facial trauma, the presence of a paranasal sinus polyposis or paranasal sinus tumours and previous paranasal sinus surgery, the latter being the most common factor. A predisposing factor was identified in more than half of the cases in our series, 45\% being primary mucoceles. Bokmühl et al.\textsuperscript{15} presented 66\% secondary mucoceles in 185 operated patients. Serrano\textsuperscript{16} and Kyung\textsuperscript{17} published similar figures.

Findings of inflammation are common in all chronic rhinosinusitis, including paranasal sinus polyposis, and only a small proportion of them develop a mucocele. The risk of developing a mucocele associated with nasal polyposis is estimated at less than 1\%.\textsuperscript{17} In our case studies, a bilateral concomitant paranasal sinus polyposis was diagnosed in only 6 unoperated patients. Therefore, inflammation like the obstruction of the ostium, which is associated with these polyposes, is not a sufficient condition for these lesions to develop.

Headache and orbital tumour were the most common clinical manifestations in the ethmoid–frontal location at the time of diagnosis, followed by diplopia caused by displacement of the eyeball. The presentation sign in the maxillary sinus, in half the cases, was a jugal tumour. Ocular symptoms predominated in the sphenoid sinus, due to the proximity of the optic nerve and the oculomotor cranial nerves. Two patients presented diplopia due to involvement of the third nerve and one patient presented a loss of visual acuity caused by compression of the optic nerve. Loss of visual acuity is a situation where emergency surgery is indicated. The literature reflects a high visual recovery rate after surgical drainage.\textsuperscript{18–20} In 14\% of the mucoceles the patients did not present any symptoms, these data are similar to other series which put the percentage of asymptomatic patients between 8\% and 11\%.\textsuperscript{19,20}

Treatment of these lesions is surgical. The objective of surgery is to marsupialise the cavity of the mucocele towards the nostril, attempting to ensure good ventilation and drainage of the affected sinus preserving the external wall of the mucocele, since this mucus is healthy, and to resume its normal appearance when it is well ventilated. In our series, the patients treated in the 1980s were operated using the external route, and the endoscopic endonasal approach was introduced in our centre from the 1990s. This is currently the approach of choice, although a number of situations are accepted as relative contraindications for it. The frontal sinus is the location which presents more surgical limitations.\textsuperscript{21} We indicate an external approach in our centre for frontal mucoceles which only affect the most external and posterosuperior region of the sinus, when the appearance of the mucocele is the consequence of an endonasal process which separtes the sinus, such as an osteoma or major sclerosis in the region of the frontal recess, or when the mucocele is the consequence of orbital herniation. These situations are shown in Fig. 2.

The percentage of recurrence is less than 10\% according to most contemporary series. In our case studies 4 recurrences were observed, all localized in the frontal sinus: two patients after an endonasal approach and in two patients after an external approach. This greater percentage of recurrence in the externally operated group could be due to the fact that the mucoceles were larger and more complex surgically.

Recently, Courson et al.\textsuperscript{21} published a meta-analysis in which they compared the results of 957 ethmoid–frontal mucoceles operated using the endoscopic endonasal approach to those of patients on whom external approach was used. The results in both groups were similar.\textsuperscript{21}

<table>
<thead>
<tr>
<th>Location</th>
<th>Type of surgery</th>
<th>ENS</th>
<th>External approach</th>
<th>Combined</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Ethmoid–frontal</td>
<td></td>
<td>15</td>
<td>7</td>
<td>10</td>
<td>32 (55%)</td>
</tr>
<tr>
<td>Ethmoidal</td>
<td></td>
<td>8</td>
<td>0</td>
<td>0</td>
<td>8 (14%)</td>
</tr>
<tr>
<td>Maxillary</td>
<td></td>
<td>14</td>
<td>0</td>
<td>0</td>
<td>14 (24%)</td>
</tr>
<tr>
<td>Sphenoid</td>
<td></td>
<td>4</td>
<td>0</td>
<td>0</td>
<td>4 (7%)</td>
</tr>
<tr>
<td>Total</td>
<td></td>
<td>41</td>
<td>7 (12%)</td>
<td>10 (17%)</td>
<td>58 (100%)</td>
</tr>
</tbody>
</table>
Figure 2  Indications of an external or combined approach. (A) Right frontal mucocele septated by an osteoma. (B) Right frontal mucocele secondary to orbital herniation. (C) Mucocele in the external left frontal region associated with paranasal sinus polyposis. (D) Mucocele external left frontal region.

Table 4  Results of Main Contemporary Published Series.

<table>
<thead>
<tr>
<th>Authors (year)</th>
<th>No. of mucoceles</th>
<th>Location</th>
<th>Approach</th>
<th>Recurrence (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Rombaux (2000)</td>
<td>178</td>
<td>All</td>
<td>ENS + combined</td>
<td>2</td>
</tr>
<tr>
<td>Har-El (2001)</td>
<td>108</td>
<td>All</td>
<td>ENS</td>
<td>0.1</td>
</tr>
<tr>
<td>Khong (2004)</td>
<td>41</td>
<td>All</td>
<td>ENS</td>
<td>0</td>
</tr>
<tr>
<td>Bockmühl (2006)</td>
<td>185</td>
<td>All</td>
<td>ENS + combined</td>
<td>2.2</td>
</tr>
<tr>
<td>Serrano (2006)</td>
<td>60</td>
<td>All</td>
<td>ENS</td>
<td>0</td>
</tr>
<tr>
<td>Sautter (2008)</td>
<td>57</td>
<td>All</td>
<td>ENS</td>
<td>0</td>
</tr>
<tr>
<td>Obeso (2009)</td>
<td>81</td>
<td>All</td>
<td>ENS</td>
<td>10</td>
</tr>
<tr>
<td>Dhepnorrarat (2012)</td>
<td>40</td>
<td>Frontal-ethmoidal</td>
<td>ENS+ combined</td>
<td>2.5</td>
</tr>
<tr>
<td>Courson (2014)</td>
<td>121</td>
<td>Frontal-ethmoidal</td>
<td>ENS+ combined</td>
<td>10.7</td>
</tr>
<tr>
<td>Our series</td>
<td>58</td>
<td>All</td>
<td>ENS+ combined</td>
<td>7</td>
</tr>
</tbody>
</table>

The results of the main series published in recent years are shown in Table 4.

Conclusions

Paranasal sinus mucoceles are lesions which primarily affect the ethmoid–frontal complex. The endoscopic endonasal approach is the treatment of choice, as it is a safe technique with good results. The frontal sinus involves more surgical limitations and recurrences are more common in this location.

Conflict of Interests

The authors have no conflicts of interest to declare.

References

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