Case Report

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Paucisymptomatic Post-Surgical Megamucocele of the Sphenoid Sinus

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Abstract

Paranasal sinus mucocele is a benign pseudocystic slowly growing and locally invasive mass characterized by accumulation and retention of mucoid secretion. It is lined by a respiratory epithelium. It usually occurs in adults. The frontal sinus is the most frequently affected site followed by the anterior ethmoid sinus. Sphenoidal mucocele is rare, accounting for 1 to 2 % of all paranasal sinus mucoceles. We report the story of a 75-year-old lady who developed a paucisymptomatic megamucocele of the sphenoid sinus 20 years after resections of an ossifying fibroma by a neurosurgeon via a fronto- orbital approach. She underwent a wide endonasal endoscopic marsupialization of the lesion. One year after the ENT surgery the surgical opening has remained open. We report the case, the imaging and review the pertinent literature.

Keywords: *Mucocele of Sphenoid Sinus; Pseudocyst; Magnetic Resonance Imaging; Frontal Sinus; Fronto-orbital Approach; Marsupialization; Primary; Secondary; Ethmoid sinus; Neuro-surgical approach; paranasal sinus; expansile; recurrence.*

Introduction

Paranasal sinus mucocele is a benign pseudocystic slowly growing and locally invasive mass, which is characterized by the retention of mucoid secretion. This accumulation leads to thinning, distension and erosion of one or more of its bony walls.

Mucocele of the frontal sinus is the most commonly affected sinus, followed by the anterior ethmoidal sinus. Sphenoid sinus mucocele is rare, accounting for 1% to 2% of all the paranasal sinus mucoceles [1].

We report the case history of a lady who developed a megamucocele of the sphenoid sinus 20 years after she had undergone resection by a neurosurgeon of an ossifying fibroma.

Case Report

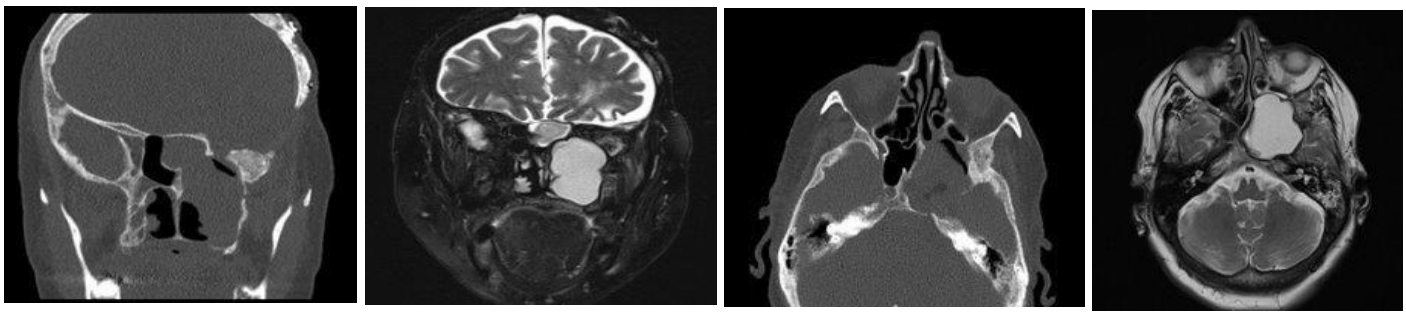
This is the story of a 75-year-old lady with a past medical history of an ossifying fibroma resection which was performed by a neurosurgeon via a neurosurgical and frontal approach. The resection required 3 surgical operations and the patient received postoperative radiotherapy. As a sequela of the management of this fibroma, she is blind on the left side.

More recently, she was seeking the advice of an ENT doctor due to some nonspecific nasal complaints such as pruritus, nasal discomfort, but no significant headache or diplopia.

A sinus CT-scan was performed and which revealed the presence of a huge expansible process within her left sphenoid sinus, extending anteriorly into her posterior ethmoid and inferiorly behind the posterior wall of her maxillary sinus. The size of the lesion was about 6 cm in the craniocaudal axis, 4 cm in the right and left axis and 4 cm in the antero-posterior axis. Posteriorly there was a dehiscence of the bony wall of the carotid canal. The CT scan features of the lesion confirmed the position of the lesion that would necessitate the adoption of a neurosurgical frontal approach for her operation to excise the lesion.

Due to the size of the lesion, the patient was referred to our department for management.

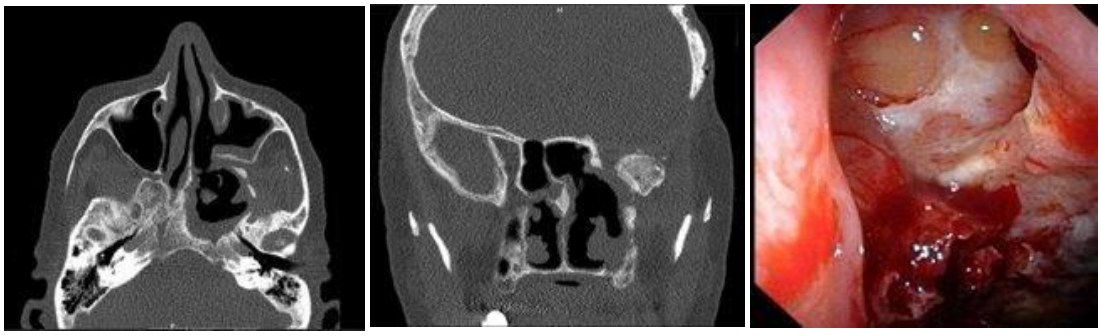
She had a Magnetic Resonance Imaging (MRI) scan which confirmed the diagnosis of a megamucocele that had extended into her posterior ethmoid and behind the posterior wall of her maxillary sinus. This mass was filled with mucoid secretion with a heterogeneous signal (the lesion was hypo signal on T1 weighted sequences with some areas in hypersignal on T1 and T2). After injection of gadolinium there was no peripheral enhancement. (*Figure 1*).



A. Coronal view of the CT-scan B. Axial view of the CT-scan C. Coronal view of the MRI D. Axial View of the MRI

Figure 1: Preoperative views

The patient underwent a large marsupialization of the mucocele via an endonasal endoscopic approach associated to an opening of the posterior ethmoid and the opening of the pterygopalatine fossa. The postoperative period was uneventful. One year after the surgery we did not notice any recurrence of the mucocele. (*Figure 2 shows the result*).



A. Axial view of the CT scan B. Coronal view of the CT scan C. Endoscopic view

Figure 2: Postoperative views.

Discussion

Paranasal sinus mucocele is a benign, slowly growing, expansible lesion originating from a paranasal sinus cavity. It most commonly occurs in adults in the frontal sinus, followed by the anterior ethmoid sinus. Sphenoidal localization is very rare [1].

Typically, a combination of chronic inflammation and obstruction of the main sinus ostium plays a significant role in the pathogenesis [2]. Mucoceles are classified into 2 categories: idiopathic or secondary mucoceles [3].

Secondary mucoceles can be observed as a late complication after previous sinus surgery whatever the route used (external or endonasal one). The time between the onset of the disease and diagnosis after a sinus surgery can vary considerably, ranging from 3 days to 38 years, with an average delay of 4 years [4]. In the present case the delay between the first surgery and the diagnosis of the sphenoid mucocele was 20 years.

The initial clinical presentation is variable and can be non-specific. In the present case the patient had no headache, only pruritus, sneezing and nasal discomfort.

The diagnosis can be an incidental finding on a sinus CT scan which has been undertaken as part of the diagnostic workup of non- specific sinonasal complaints or when the patient has had in the past a sinunasal surgery.

However, the first common classical symptom associated to a sphenoid mucocele is headache [5,6].

Typically, it is reported as persistent deep-seated headache. It can irradiate to the occiput, vertex or at any point of the skull. The headache is related to the expansion of the lesion whether it is associated or not to an acute infection. When there is thinning and erosion of the planum sphenoidale or the clivus, then the headache may be explained by stretching of the dura mater.

The differential diagnosis of such headache must include migraine or cephalalgia.

Visual disturbances (diplopia or even blindness) are the second most common clinical presentation [7-11]. This is explained by the fact that on the lateral wall of the sphenoid sinus lays the cavernous sinus with inside the oculomotor cranial nerves (III, IV, VI).

Compression of the VI nerve causes a diplopia very quickly because the VI nerve is the longest nerve inside the cavernous sinus and is very sensitive to any compression.

The diplopia can result to a compression of the IIIrd cranial nerve as well. Ophthalmoplegia is another presentation which typically is irreversible[10]. Blindness is also a possible symptom due to a compression of the optic nerve. The bony canal surrounding the optic nerve can undergo dehiscence due to its erosion by the mucocele. In the present case the patient has been blind since the first operation performed 20 years earlier.

The diagnosis of a sphenoid mucocele requires a nasal endoscopy and the undertaking of a radiology imaging [1-4].

In patients who underwent in the past of a sinus surgery we can visualize a yellow bulging of the mucosa in the sphéno-ethmoidal recess but usually the nasal endoscopy is not contributive and that's why an imaging classically reveals the lesion.

On a sinus CT scan without injection of any contrast medium we can see a complete opacity of the affected sphenoid cavity with either sclerosis or thinning, distension and erosion of the bony walls. In some cases, the clivus and or the planum sphenoidale can be very thin as well [12,13].

MRI scan is therefore a complementary tool to analyze the content of the mass [14,15]. The intensity of the signal depends on the age of the mucocele, the stage of development, the protein content and the viscosity of the liquid.

The intensity of the signal can be moderate or low on T1 weighted images and high on T2 weighted images.4 In some cases there is a peripheral enhancement after administration of contrast material. In our case the signal was high on T2 weighted sequences. When the liquid is very thick or dry the signal is very low or even absent in T2.

Differential Diagnosis [1-4,12-16]

Mucocele of the sphenoid sinus is rare and underdiagnosed particularly when the clinical presentation is subtle. In the present case the patient had non-specific nasal complaint but had reported a story of a past sinus surgery. In the case, when the nasal endoscopy is non-contributive an imaging must be requested.

The imaging scan can find an isolated sphenoid opacity. This can be compatible with a bacterial sinusitis, fungal ball or a granulomatous reaction but we must also rule out arachnoid cyst or meningocele from the Steinberg canal.

The MRI scan is a necessary complementary examination to make the right definitive diagnosis. The examination confirms the presence of liquid within the mass but can also rule out other cystic lesions such as an arachnoid cyst, meningocele, Rathke cleft cyst, pituitary adenoma, or craniopharyngioma.

In case of headache the differential diagnosis also includes ophthalmology causes (like acute angle closure glaucoma) or neurology causes (like causes of increased intracranial pressure). Expanding sphenoid sinus mucocele may compress the optic nerve or the III, IV and VI cranial nerves. So ophthalmologic examination and imaging of the skull base and cavernous sinus is mandatory.

In case of painful ophthalmoplegia we must rule out lesions such as pituitary apoplexy, ruptured intracranial berry aneurysm involving posterior communicating artery or the cavernous segment of the internal carotid artery, carotico-cavernous fistula.

Treatment [17-19]

Treatment of sphenoid mucocele is surgical. The purpose of the surgery consists with a large drainage of the lesion into the nasal cavity. Before the development of the endoscopic endonasal surgery a trans-facial or transcranial approach was recommended with complete removal of the fibromucosa of the mucocele. Since the development of the endoscopic techniques the marsupialization of the lesion is the gold standard in most of the cases with preservation of the lining of the lesion. Commonly, the approach is preformed trans-nasally via the sphéno-ethmoidal recess.

In the present case, the opening has been enlarged inferiorly and laterally, behind the posterior wall of the maxillary sinus due to the extension of the mass. Postoperative closure of the surgical opening can occur particularly when the mucocele is the late sequela of a surgery with subsequent huge fibrosis. The use of a rescue flap is then recommended [18,19].

Conclusion

Sphenoid sinus mucocele is a rare pathology that is sometimes difficult to diagnose due to their non- specific clinical presentations. Therefore, a comprehensive clinical and radiographic examination is essential even if the patient has few symptoms particularly when the patient has undergone a sinus surgery in the past or radiotherapy. Imaging is essential to make the diagnosis and demonstrates its expansion. Marsupialization of the lesion via an endoscopic approach is nowadays the best option. A follow up is mandatory to rule out any recurrence.

Take Home Message

We emphasize the need of an imaging of the paranasal sinuses in every patient operated upon for a sinus problem many years ago when the nasal endoscopy cannot check all the paranasal cavities. The patient's complaints were not correlated to the size and expansion of the mucocele.

Consent Declaration

We declare that the following case report titled "Paucisymptomatic Post-Surgical Megamucocele of the Sphenoid Sinus" has been prepared with the respect for patient privacy and confidentiality. The information presented in this case report is based on the clinical details of a specific patient who was diagnosed with this medical condition. The purpose of sharing this case report is to contribute to medical knowledge and education within the healthcare community.

The patient involved in this case report has provided informed consent for the utilization of their clinical information for educational and research purposes. All personal identifiers have been removed to ensure the patient's anonymity and privacy.

Conflict-of-Interest Statement for Dr. Sapa Vadjeru

Dr. Sapa Vadjeru declares that there are no conflicts of interest associated with the case presented in this work. The author has received no financial support or any form of compensation from any organization or entity that could potentially influence the content of this work.

Conflict-of-Interest Statement for Prof. Eloy

Professor Eloy affirms that there are no conflicts of interest concerning the content discussed in this publication. The author has not received any personal benefits from organizations or individuals that might affect the objectivity of this work.

Conflict-of-Interest Statement for Dr. De Coen:

Dr. De Coen confirms the absence of conflicts of interest relevant to the subject matter of this manuscript. The author has not been

involved in any financial arrangements or partnerships that could potentially impact the integrity or impartiality described in this work.

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