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Bilateral Postoperative Maxillary Cyst: A Case Report

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Abstract

A postoperative maxillary cyst (POMC) is a benign lesion arising from trauma or surgery involving the midface, specifically the maxillary antrum, as a late complication. It is usually locally aggressive, and accounts for approximately 20% of the lesions seen in Japanese patients undergoing extensive maxillary sinus surgery. According to the available literature, the insertion of mucosal cells between the bony edges of a fracture or osteotomy may result in the cystic degeneration that precedes this type of lesion. The clinical and histological characteristics of a POMC are often mixed, with fibrous connective and myofibroblastic tissue in the surrounding anatomy, which could make the diagnosis difficult or misleading.

Keywords: Postoperative maxillary cyst; Bilateral; Lesion

Introduction

A postoperative maxillary cyst (POMC) is a benign lesion arising from trauma or surgery involving the midface, specifically the maxillary antrum, as a late complication. It is usually locally aggressive, and accounts for approximately 20% of the lesions seen in Japanese patients undergoing extensive maxillary sinus surgery. According to the available literature, the insertion of mucosal cells between the bony edges of a fracture or osteotomy may result in the cystic degeneration that precedes this type of lesion [1]. The clinical and histological characteristics of a POMC are often mixed, with fibrous connective and myofibroblastic tissue in the surrounding anatomy, which could make the diagnosis difficult or misleading. Here we present a clinical case of bilateral POMC with 21 years of evolution and extensive fibrous reparative tissue, which represents an uncommon finding among the American population when compared to the Asian population.

A POMC is also called a surgical ciliated cyst, postoperative paranasal cyst, or respiratory implantation cyst [2,3]. It is a wellknown pathological entity, commonly associated with corrective, reconstructive, or trauma surgery. A POMC usually develops unilaterally as a solitary lesion, with a bilateral appearance being very rare [3,4]. The current American literature shows only a few reports of cases involving unilateral POMCs, but no bilateral cases. The largest report of POMCs known to date consists of 71 patients [5], while the most relevant literature regarding this entity has been published by different Japanese and Korean groups [6]. This lesion appears less frequently in the Western literature likely due to misdiagnoses [6] and a lack of publication; therefore, we encourage the reporting of such a pathology. Moreover, a retrospective study is necessary to determine the incidence of this lesion in the American population.

Some reports have suggested two possible etiologies for POMCs: the closure of the natural ostium and intranasal opening, along with the entrapment of the sinus mucosa, and the retention of tissue fluid/ blood after a surgical procedure [2,7]. These cysts are usually lined by pseudostratified columnar ciliated epithelium of the respiratory type, which may be focally replaced by squamous, cuboidal, or columnar epithelium [8], with surrounding fibrous connective tissue [9]. Radiographically, it appears as a well-defined unilocular radiolucency in the maxillary sinus, capable of eroding the surrounding bony structures if not treated. Its appearance can vary from between 3 and 60 years after the traumatic event occurs [10], with a good long term prognosis once the lesion is removed and normal sinus draining is achieved [3,10,11].

The diagnosis of a POMC can be completed using a biopsy alone, but we decided to perform an immunohistochemical analysis of the extracted tissue to determine the most common markers (Table 1). Actin, desmin, and S-100 are among the most common markers in either normal or pathological fibrous connective tissue, and as expected, POMCs possess similar markers due to their mixed cellular components.

Case Presentation

In 2011, a 56-year-old male presented for the evaluation of slowly progressing right cheek swelling, accompanied by diplopia and dizziness upon walking. Upon examination, the lesion was found to be firm and painless upon manipulation, and covered by smooth erythematous skin (Figure 1). There were no motor or sensory disturbances, and the lesion could not be visualized intraorally. This patient had a history of high blood pressure, and was only hospitalized once in the past, due to a motor vehicle accident in 1989 that resulted in multiple facial

Author	Year	Number of patients
Hayhurst DL et al. [1]	1993	
Kubo I [2]	1927	1
Lee JH et al. [3]	2014	1
Chindasombatjaroen J et al. [4]	2009	7
Kaneshiro S et al. [5]	1981	71
Yamamoto H et al. [9]	1986	60
Cano J et al. [10]	2009	1
Basu MK et al. [11]	1988	23
Heo MS et al. [12]	2000	19
Yoshikawa Y et al. [14]	1982	9
Leung YY et al. [15]	2012	3
Amin M et al. [16]	2003	1
Bulut AS et al. [17]	2010	1
Shakib K et al. [18]	2009	1
Thio D et al. [20]	2003	1
Niederquell BM et al. [6]	2016	284

Table 1: Overview of postoperative maxillary cysts from the literature.

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Figure 1: Right cheek swelling, with smooth erythematous skin over the area

fractures. After obtaining his medical records, the past diagnosis of a Le Fort II and right mandibular body fracture from the accident was confirmed, along with a description of the treatment performed (open reduction and internal fixation of both fractures using wires, along with intermaxillary fixation) (Figure 2). Three months after the trauma, a dacryocystorhinostomy was performed to restore the flow of tears into the nose from the lacrimal duct. Both procedures were successful, without any complications seen during the 15-month follow-up. After this, the patient discontinued his appointments and no further information was collected in his record.

The radiographic examination of the region with a new maxillofacial CT showed a large round cyst-like radiolucency with sclerotic borders, occupying 80% of the maxillary sinus and displacing the orbital floor, along with its contents (Figure 3). A similarly shaped, but much smaller lesion was observed in the left maxillary sinus (Figure 4). The aspiration cytology of both lesions showed the presence of mucoid material, along with lymphocytes and foamy macrophages. In addition, the histopathological analysis showed columnar and pseudostratified epithelium, along with dense fibrous connective





Figure 3: Coronal view (left) showing displacement of the orbital contents, along with orbital floor resorption. Axial view (right) showing anteroposterior extension of the lesion, displacing part of the lateral wall of the maxillary sinus.

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tissue. Well-differentiated fibroblasts and myofibroblasts were also present (Figure 5). Immunohistochemistry was performed on the specimen in order provide evidence of the common biomarkers for future publications (Table 2). This patient underwent complete surgical enucleation of both lesions, along with a right orbital floor reconstruction, under general anesthesia. No evidence of local recurrence was seen during the 10 months of follow-up. Moreover, the patient's diplopia and dizziness while walking improved, with no pain or pressure being reported by

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Biomarkers	Surrounding connective tissue	Pathological tissue
Ki67	Low	Low
β catenin	Positive, focal	Negative
BCL-12	Negative (internal control positive)	Negative (internal control positive)
CD-34	Negative (internal control positive)	Negative (internal control positive)
S-100	Positive in fragmented nervous	Positive in fragmented nervous
tissue		tissue
Actin	Positive, focal	Negative
Desmin	Negative	Negative

Table 2: Immunohistochemistry for the most common biomarkers found in normaland pathological fibrous connective tissue. Sample is only positive for actin,desmin, and S-100.

this patient.

Discussion

A POMC is a benign pathology that could appear up to 60 years after a maxillary trauma, without evidence in the literature of any malignant transformation [6]. However, it should be treated promptly, in order to prevent complications associated with the surrounding anatomical structures, and to ease the course of treatment. This will result in a less traumatizing experience for the patient.

In order to determine the extent of the lesion, a maxillofacial CT [12] is important to outline the characteristics of a POMC, which will become very important when planning surgery [13,14]. This pathology is usually misdiagnosed due to the common fibrous findings in a traumatized maxillary sinus, along with a poor clinical description and/or lack of corresponding imaging. The histology of a POMC could be potentially confusing because of the extensive fibrous repair of a previous trauma, and could render the diagnosis difficult [11,15] since the presence of respiratory epithelium is a mandatory finding in the maxillary sinus. Other lesions of the maxillary sinus that must be considered in the diagnosis of a POMC are mucous retention cysts and maxillary pseudocysts [16-18]. According to Gardner [19], histologically, these lesions share pseudostratified columnar epithelium [15,20,21]; however, they do not usually follow an aggressive pattern, displacing or reabsorbing the surrounding bony structures, when compared to POMCs [4].

Conclusion

We have reported this case here because this disease requires more attention in order to obtain an adequate rate of incidence in the Western population. Therefore, we encourage other researchers to conduct a retrospective study of the American literature for a further evaluation of this pathology.

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