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Case Report

Combined vascular malformation and retention cyst in the maxillary sinus

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ABSTRACT

Objectives: Retention cysts are common in the maxillary sinuses. Combined vascular malformations are abnormalities that involve at least two types of vessels and are frequent in different regions of the organism, especially the skin. The association of a combined vascular malformation and retention cyst in the maxillary sinus is rare. We report the case of a 35-year-old asymptomatic male patient in whom a maxillary sinus cyst was detected during orthognathic surgery. The histopathological findings were compatible with a combined capillary and lymphatic malformation associated to a retention cyst.
Methods: Case report of a combined vascular malformation in a maxillary sinus retention cyst.
Results: Histopathological analysis revealed findings compatible with a combined vascular and lymphatic malformation and a retention cyst lined with ciliated pseudostratified respiratory epithelium. Numerous dilated blood and lymphatic vessels, delimited by a single layer of endothelial cells, were observed near the cyst. Immunohistochemistry for D2-40 antigen was positive in the lining cells of lymphatic vessels and CD34 was detected in endothelial cells of capillary blood vessels.
Conclusion: To our knowledge, this is the first case of the association of a combined capillary and lymphatic malformation and retention cyst in the maxillary sinus. Vascular malformations should be included in the diagnostic hypotheses for cystic lesions of the maxillary sinus usually considered retention cysts. It is possible that other cases of combined vascular malformations exist. In this way, it is important to emphasize the need of histopathological examination of all specimen removed during maxillofacial surgeries.

1. Introduction

Retention cysts are common imaging findings and the differential diagnosis includes unilateral diseases of the maxillary sinuses [1,2]. The prevalence of retention cysts ranges from 1.4 to 35.6%, probably because of the different populations samples selected in the studies or of differences in the accuracy of the diagnostic methods used for their detection, such as panoramic radiography, computed tomography and nuclear magnetic resonance [1].

Most vascular malformations are diagnosed at birth or during childhood. They may cause little or no symptoms during life. In cases of small and well-delimited abnormalities, surgical exeresis may be curative, although incomplete surgical removal can predispose to recurrence of the lesion. In the absence of symptoms, the clinical diagnosis is based on routine medical examination or radiographic records. The final diagnosis is made based on histopathological findings [1,2].

We report a rare association of a retention cyst in the maxillary sinus and a combined capillary and lymphatic malformation.

2. Case report

2.1. Clinical history

A 35-year-old healthy male patient was examined for orthognathic surgery for retrognathia correction. Panoramic radiography revealed an image of dome-shaped and slightly radiopaque lesion

☆ AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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with a cloudy aspect of the right maxillary sinus (Fig. 1). He was asymptomatic and the radiographic diagnosis was of retention cyst of maxillary sinus. After preoperative examinations, he was submitted to surgery. During surgical procedure, a spherical yellowish cystic lesion with irregular surface, measuring around 2.0 × 1.0 cm was found in the base of the right maxillary sinus as seen in the preoperative x-ray. The lesion was easily removed by curettage. There was no drainage of the lesion or excessive bleeding during its removal. Specimen was sent for histopathological analysis.

2.2. Histopathological findings

The mucosa of the maxillary sinus was lined with ciliated pseudostratified columnar epithelium. A cystic lesion lined with ciliated pseudostratified respiratory epithelium was observed in the lamina propria. A glandular duct was present in the cystic capsule. Numerous lymphatic and blood vessels, sometimes congested, were found in the lamina propria, as well as in the cystic capsule (Fig. 2). Immunohistochemistry detected D2-40 antigen staining in lymphatic endothelial cells and CD34 staining in capillary endothelial cells (Fig. 3). The final histopathological diagnosis was combined capillary and lymphatic malformation associated with a retention cyst.

3. Discussion

Retention cysts are common in the maxillary sinuses and are caused by blockage and dilatation of seromucous ducts due to the accumulation of mucin produced by the secretory glands of the ciliated pseudostratified respiratory epithelium [2]. The differential diagnosis should include sinusitis, mucocele, polyps, benign and malign neoplasms and odontogenic cysts [3].

They are mostly asymptomatic and can be found in routine panoramic radiographic, usually the primary resource available for the initial evaluation [1,3,4]. Computed tomography is a more accurate diagnostic image examination that could be used to elucidate eventual doubts about the presence and pattern of the lesions, to help on subsequent surgical procedures and for periodic control [3,4].

Endoscopic or surgical treatment of these cysts is indicated in the presence of clinical symptoms such as headache, nasal obstruction, facial pain, post-nasal drip and rhinorrhea, although most patients exhibit spontaneous remission or asymptomatic lesions as observed in the present case [1,4].

Vascular anomalies represent a group of diverse lesions related to vascular development whose manifestation ranges from a “spot” at birth to life-threatening conditions. The correct diagnosis is fundamental for adequate treatment, which is often multidisciplinary [5–7]. According to the recent classification of the International Society for the Study of Vascular Anomalies (ISSVA) [8], vascular anomalies can be divided into two groups according to histopathological findings: lesions with a pathological proliferative component (vascular tumors) and relatively static lesions (vascular malformations) [5].

Vascular malformations are the result of errors that occur during embryogenesis [5]. They are classified into four large groups according to the predominant type of vessel involved or to their flow characteristics (slow flow: capillary, venous, lymphatic; high flow: arteriovenous): simple, combined, large vessels, or associated with other anomalies [5,7,8]. The presence of two or more types of malformations in a single lesion is called combined vascular malformation [8].

Hemangioma is a generalized term frequently used to designate arterial, venous, lymphatic or combined malformations. The term lymphangioma is used for lymphatic malformations. These malformations differ in terms of evolution and treatment [5].

Lymphatic malformations are benign lesions that arise from the endothelial cells of lymphatic vessels. The majority of these

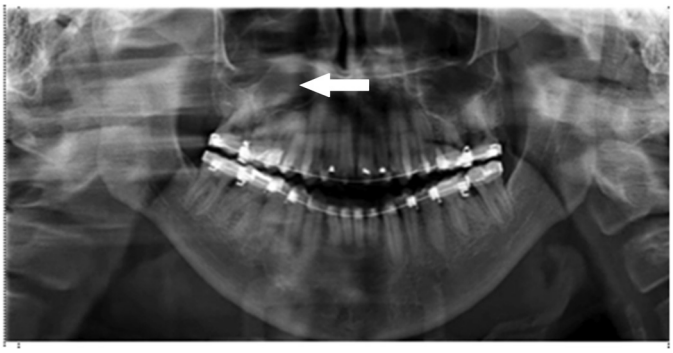

Fig. 1. Panoramic radiograph showing an image of cup-shaped morphology that caused partial opacification of the right maxillary sinus (arrow).

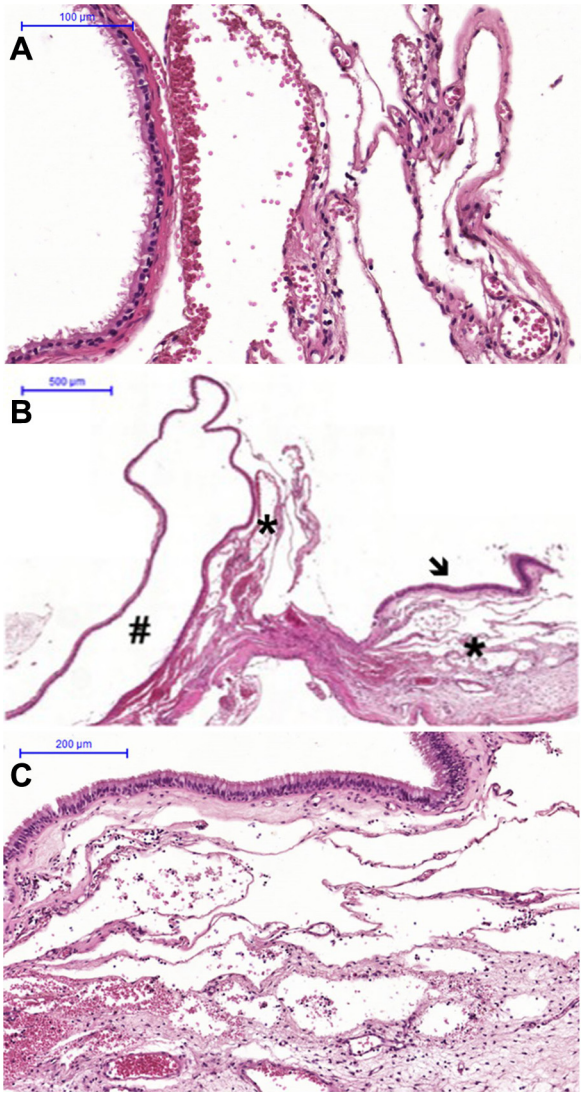

Fig. 2. (B) H&E-stained section showing the maxillary sinus mucosa on the right (↘) and retention cyst on the left (#), with lymphatic and capillary malformation in the lamina propria and cystic capsule (*). (A) Detail of the wall of the maxillary retention cyst, with capillary and lymphatic malformation in the connective tissue capsule. (C) Detail of the maxillary sinus mucosa lined with respiratory epithelium and capillary and lymphatic malformation in the capsule.

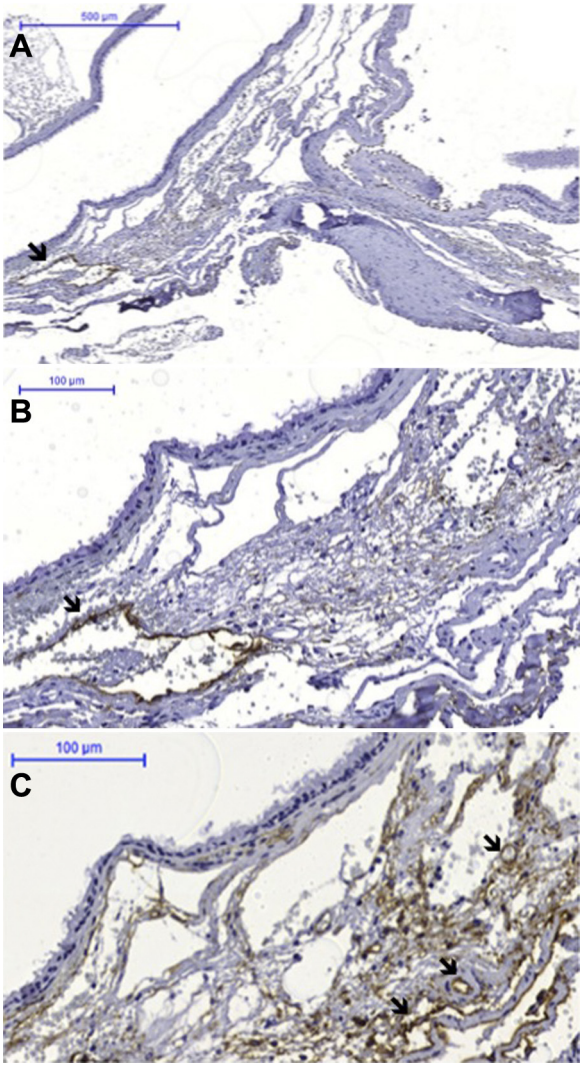

Fig. 3. (A) Immunohistochemistry for D2-40 antigen showing positive staining in the lymphatic vessel lining (➘). (B) Detail of positive D2-40 staining in the lymphatic endothelium (➘). (C) Immunohistochemistry for CD34 showing positive staining in endothelial cells of capillaries and arterioles (➘).

malformations is congenital or emerges within the first year of life. Lymphatic malformations can be found throughout the body, with a preference for the axilla, head, neck and inguinal region, and are often associated with genetic syndromes. They are classified as microcystic, macrocystic or combined according to the size of the vascular spaces found [9].

Mattila et al. [6] described 435 cases of vascular anomalies diagnosed in children, in which one-third of the cases were vascular tumors and 55.2% were vascular malformations. However, only 89.2% of the tumors and 38% of the malformations were correctly diagnosed by multidisciplinary teams.

The exclusive finding of vascular malformations in the maxillary sinus is rare. Hudson and Meszaros [10] described a case of lymphangioma in the maxillary sinus of a 39-year-old man with a history of chronic sinusitis, rhinitis, facial pain and x-ray image of an occlusive lesion in the left maxillary sinus. In the present case, the patient was asymptomatic.

To our knowledge, there are no reports on combined vascular malformations in the maxillary sinus. In the present patient X-ray evaluation revealed dome-shaped lesion compatible to maxillary retention cyst. During orthognathic surgery for retrognathia correction the lesion was removed. The histopathological diagnosis was of combined vascular malformation associated with a retention cyst in the maxillary sinus.

Retention cysts are mostly asymptomatic and do not need medical or surgical treatment [1,3,4]. It is possible that other cases of combined vascular malformations exist. Thus, vascular malformations should be included in the diagnostic hypotheses for the maxillary sinus lesions usually considered retention cysts. In this way, it is important to emphasize the need of histopathological examination of all specimen removed during maxillofacial surgeries.

Conflicts of interest

There is no conflicts of interest involving the authors of this paper.

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