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Case Report
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Fistulized Palatine Duct Cyst: A Case Report

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Abstract

Introduction: Nasopalatine cysts are the most common non-odontogenic cysts of the maxillae, with a prevalence of around 1% in the general population. The age of discovery is between 40 and 60 years. The literature generally reports a male predilection, with a M/F sex ratio of up to 3:1. The aim of our study was to discuss the diagnostic, therapeutic and evolutionary aspects of a cyst of the palatine canal.

Observation: This was a male ENT patient presenting with endonasal swelling for one year, associated with bilateral nasal obstruction, anteroposterior rhinorrhea and headache. On examination, we noted a left endonasal swelling with deviation of the septum to the right, taking up the homolateral hemiface with partial effacement of the nasolabial folds, painless both spontaneously and on palpation. A CT scan of the nasal cavities and facial sinuses revealed an obstructive maxillo-palatal process with bone lysis. Preoperative work up was normal. Surgical excision was performed one month later under general anesthesia. Histological examination of the surgical specimen revealed a remodeled epithelial cyst. Follow-up was straight forward, and the patient regained a symmetrical face 3 months later.

Conclusion: The nasopalatine cyst is of embryonic origin. It must be differentiated from an apicodental cyst. Diagnosis is based on radiology and histology. Treatment is surgical. Exeresis must be complete to avoid recurrence. Recurrence may take more than 5 years. Long clinical follow-up is essential.

Keywords: Palatal cyst; Endonasal mass; Epithelial cyst

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Introduction

First described in 1914 by Meyer, the nasopalatine duct cyst may occur in the nasopalatine duct or in the soft tissues of the palate at the opening of the duct, where it is referred to as the "palatine papilla cyst". The nasopalatine canal, strictly speaking a passageway in the hard palate, usually contains the nasopalatine nerve and vessels, as well as embryological remnants of the nasopalatine duct, which obliterates at birth" [1]. Although the pathogenesis of nasopalatine cyst is still uncertain, it has been suggested that its development is due to probable spontaneous cystic degeneration of the epithelial remnants present in the nasopalatine duct, or to stimulation of these remnants by physical or biological factors, leading to proliferation and degeneration [2-4]. The cells may be activated spontaneously during life, or possibly stimulated by the irritating action of various agents, including infection. The nasopalatine canal cyst is the most common non-odontogenic cyst of the maxilla, with a prevalence of around 1% in the general population [5]. In a study carried out in Brazil on 2,114 pediatric patients presenting with oral and maxillofacial lesions, only 0.8% had presented with a non-odontogenic cyst, 0.2% of which were epidermoid. These lesions are generally slow-growing and expansive, and in some cases are associated with marked bone destruction and recurrence [6,7]. The age of discovery is between 40 and 60. The literature generally reports a male predilection, with an M/F sex ratio of up to 3:1 [2,5].

Thus, appropriate clinical and radiographic examination and careful histopathological analysis are essential to ensure correct diagnosis and establish a conservative surgical approach [3].

The aim of this study was to discuss the clinical and therapeutic aspect of a palatal canal cyst.

Observation

This was an 18-year-old male ENT patient presenting with endonasal swelling that had been present for one year, associated with bilateral nasal obstruction, anteroposterior rhinorrhea and

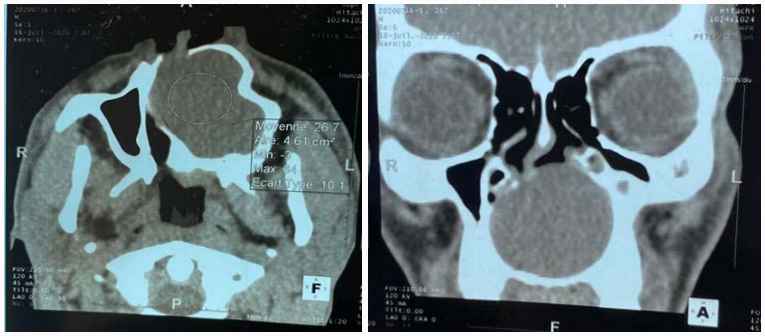

Figure 1: Evidence of bone spur and obstruction of the nasal cavity.

frontal headache. On examination, we noted a left endonasal swelling obstructing the nasal fossa with deviation of the septum to the right, swelling also taking in the homo lateral infra orbital and buccal region, extending from the infra orbital border to the line passing through the left labial commissure with partial effacement of the nasolabial folds, painless spontaneously and on palpation. An otherwise healthy hard palate was also noted.

Computed tomography of the nasal cavities and facial sinuses revealed a maxilla palatine process blowing the cortices with lysis in places measuring 55 mm × 38 mm, bulging into the left maxillary sinus with significant reduction of the sinus lumen and pushing back the nasal septum on the right with evidence of bone spur and obstruction of the nasal cavity (Figure 1). Surgical removal was performed one month later under general anesthesia. Histological study of the operative specimen revealed a cystic wall lined by a cylindrical coating, the chorion infiltrated with lymphocytes, epithelioid cells and giant cells, with cholesterol crystals associated with fibrosis and hemorrhage, concluding in a remodeled epithelial cyst. Follow-up was straight forward, and the patient regained a symmetrical face 3 months later. Two years later, follow-up was marked by the absence of recurrence.

Discussion

Nasopalatine canal cysts, also known as incisive canal cysts, are the most common non-odontogenic developmental cysts in the jaws [8]. It is also known by other names such as anterior midline cyst, maxillary midline cyst and anterior middle palatine cyst. It was considered a Fissural cyst in the past, but today, according to the World Health Organization (WHO) classification, they are considered developmental, epithelial and non-odontogenic cysts of the maxilla along with nasolabial cysts [9]. Most of these cysts develop on the midline of the anterior maxilla, close to the incisive foramen [5]. Nasopalatine duct cysts affect a wide age range; however, most occur between the fourth and sixth decades of life, and most studies show a significantly higher frequency in men than in women, with a ratio of 2.5:1 [5,10]. This could be due to the fact that women generally seek dental help earlier than men. Due to a lack of representative studies, it is not entirely clear whether nasopalatine duct cysts are more common in Caucasians, blacks or Asians. Nasopalatine duct cysts are asymptomatic in 30% to 50% of cases, and are discovered incidentally on dental panoramic radiography, rhino-sinus tomography or Dentascan. It has been reported that the time to consultation or discovery can range from 12 to 48 months [11,12]. In our case, it was 12 months. The clinical presentation of the cyst is often varied and presents a diagnostic difficulty and frequently misdiagnosed as a developmental or inflammatory odontogenic cystic lesion originating from the incisors due to the proximity of the cyst to the incisors [13]. A typical feature of cysts is their growth potential, which can compress neighboring structures. Occasionally (in 17% of cases), patients report pain due to compression of structures adjacent to the cyst, particularly when the cyst becomes super infected. This extension was also the cause of nasal obstruction and facial deformity (asymmetry) in our patient.

As dental panoramic and occlusal views with or without cyst opacification were no longer available, we opted for a CT scan of the facial mass [11,14-16]. In our case, the scan revealed an obstructive maxilla palatal process with bone lysis, particularly of the medial wall of the left maxillary sinus. The type of epithelium lining the nasopalatine canal is highly variable, depending on the relative proximity of the nasal and buccal cavities. The uppermost part of the canal is characterized by a respiratory type of epithelial lining, and as it descends, the lining transforms into cuboidal epithelium. In the lowermost part, closest to the oral cavity, squamous epithelium is the usual type. The cyst wall may contain a chronic inflammatory reaction of lymphocytes and plasma cells. The presence of neurovascular bundles, mucous glands and adipose tissue is also helpful in diagnosing nasopalatine duct cysts [9].

In our case, in addition to the cylindrical coating, we noted the presence of lymphocytes and fibrosis, epithelioid cells and giant cells with cholesterol crystals, leading to the conclusion of a reworked epithelial cyst.

The reference treatment in the literature is surgical, notably enucleation under general or local anesthesia *via* the vestibular or palatal approach for cysts larger than 6 mm, and surveillance for asymptomatic cysts smaller than 6 mm [11,14-16]. Given the extent of the lesion, we opted for vestibular surgery under general anesthetic.

Two years after surgery, we noted no recurrence, which is rare in the literature [1,2,4,11,17].

Conclusion

Palatal canal cyst is an entity associated with bone lysis that remains relatively rare in the literature. In this case, it is often symptomatic and the limits of extension are well determined by CT scan of the facial mass. Surgery is an excellent alternative treatment, and the recurrence rate remains low.

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