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Case Report

A giant nasopalatine duct cyst – A case report

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ABSTRACT

The Nasopalatine duct cysts (NPDCs) are the most common developmental, epithelial and non-odontogenic cysts of the maxilla. They are generally asymptomatic and are diagnosed on routine clinical or radiographic examination. They should however be distinguished from large incisive fossae. On attaining a large size, they may cause symptoms like pain and itching, and may cause root resorption, teeth displacement or bone perforation. We report a case of a giant Nasopalatine duct cyst (NPDC) in a 65 year old female patient with surgical management.

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1. Introduction

Nasopalatine duct cyst (NPDC) arises from the epithelial remnants of the nasopalatine duct, the communication between the nasal cavity and anterior maxilla in the developing foetus. As foetal development continues, this connection gradually narrows as the bones of the anterior palate fuse. This results in the formation of incisive canals that carry the neurovascular bundle, as well as the epithelial rests from the degenerated nasopalatine ducts. These were considered as fissural cysts in the past, now these lesions have been classified by the World Health Organization as developmental, epithelial, non-odontogenic cysts.¹

Clinically, the patients present with an ovoid or round swelling in the anterior region of the midline of the palate.

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In some cases, the swelling is associated with pain due to pressure on the nasopalatine nerves, and other patients may only complain of a discharge giving a salty or foul taste depending on whether the discharge is mucoid or purulent. The cyst may produce bulging of the floor of the nose and displacement of the teeth. In general, symptoms are not severe and patients often disregard them for many years.² They sometimes manifest as an inflammatory process (46%) that rarely produces facial asymmetry, since growth or expansion is intraoral (palatine). The more advanced cases are able to cause pain and itching.³ Radiographically, the lesions are round or ovoid and some may appear heart-shaped, either because they become notched by the nasal septum during their expansion or because the nasal spine is superimposed on the radiolucent area.^{2,4} Histologically, the presence of a cuboidal and respiratory mucosa associated with vessels, nerves and inflammatory cells confirms the

diagnosis of a NPDC.

We present a case of a giant NPDC, that could have been misdiagnosed as periapical lesion with its detailed course and surgical management.

2. Case Report

A 65 year old female patient reported to the department with a complaint of a swelling in the front region of the upper jaw since 15-20 days (Figure 1a,b). The swelling was small in size initially, which increased to the present size. It was associated with dull intermittent pain & unpleasant taste sensation. The patient gave history of removal of the upper front teeth because of pain, one year back, & the extractions were uneventful.

On extra-oral examination, the face appeared asymmetrical due to the presence of a solitary diffuse swelling in the central part of the upper lip, involving the philtrum & more prominent on the left side, obliterating the nasolabial angle. The skin overlying the swelling appeared taut with no sinus or fistula. Intraorally, obliteration of the labial vestibule was seen in the region to 11 to 24, due to the presence of the intraoral swelling. The swelling was roughly trapezoidal in shape on the buccal side with the longer parallel side towards the labial mucosa & the shorter parallel side towards the crest of the anterior edentulous ridge. 11 showed severe abrasion with an open pulp chamber suggesting it to be non-vital. The palatal mucosa over the rugae area also showed a diffuse swelling more prominent on the left side extending upto 25 & medially beyond the midline. The swelling was soft, tender, fluctuant & compressible on palpation. Thus clinically, a differential diagnosis of radicular cyst, residual cyst, nasopalatine duct cyst & central giant cell granuloma was made.

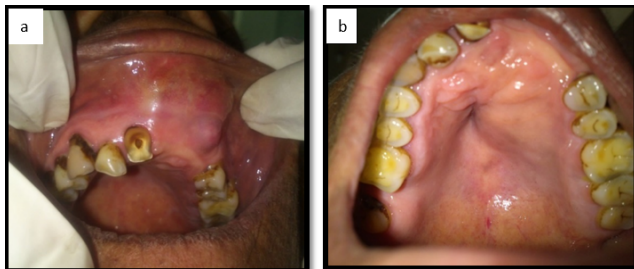


Fig. 1: a: Intra-oral swelling obliterating the labial vestibule; b: Intra-oral swelling on the palate

The patient was advised an intra-oral periapical radiograph of 11, 12 and 13 which showed a well-defined corticated radiolucency at the apical $1/3^{rd}$ of 11, 12, and 13 which was extending further more medially. So an anterior topographic occlusal view of maxilla and a panorex was advised. The anterior topographic occlusal view revealed a solitary, well defined, large, heart shaped radiolucency extending from the anterior region of the hard palate till

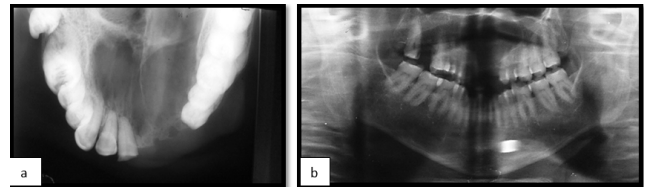


Fig. 2: a: Anterior topographic occlusal radiograph showing heart shaped cystic cavity; b: OPG showing cystic cavity

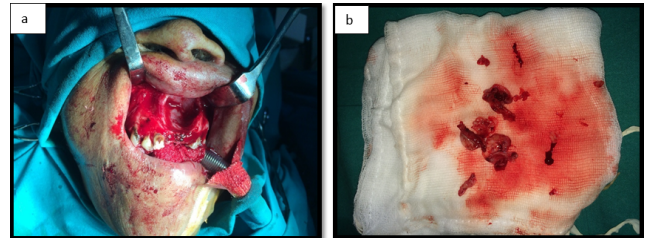


Fig. 3: a: Surgical enucleation of the cyst; b: Cyst lining in pieces

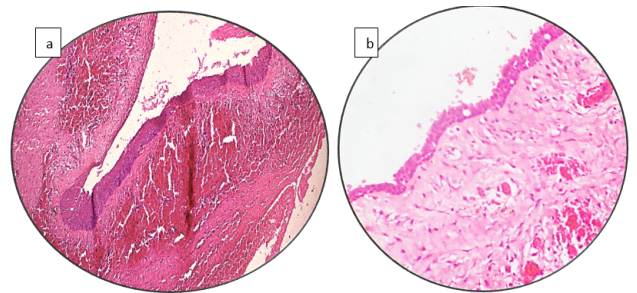


Fig. 4: a: Cyst lining and connective tissue wall showing nerve bundles and inflammatory infiltrate; b: Pseudo-stratified ciliated epithelial lining of the cyst

the level of 16 and 26. It was approximately 5 X 4cm in size. The posterior border of the radiolucency was well defined & corticated, while the anterior border appeared to merge with the edentulous alveolar ridge. (Figure 2a) In the panorex view, a pear-shaped radiolucency was seen in the midline, extending medio-laterally from the apices of 13 to 26 & superio-inferiorly from the floor of the nasal cavity till the apical $1/3^{rd}$ of 11, 12, 13 & edentulous space of 21, 22, & 23. (Figure 2b). Here too the radiolucency appeared to be corticated all around except in the inferior region near the anterior edentulous space. On the basis of radiological findings, a differential diagnosis of nasopalatine duct cyst or residual cyst was made. A fine needle aspiration cytology was performed which showed inflammatory cells.

Since the size of the cyst was large & appeared to be extending into the nasal cavity clinically, the cyst enucleation was planned under general anaesthesia. After a thorough preanaesthetic work up, the patient was taken up for enucleation. (Figure 3a,b) This was followed by histopathological evaluation.

Microscopic examination revealed a cystic cavity lined by pseudostratified epithelium with a fibrous connective tissue wall. The fibrous wall consisted of neurovascular bundles along with dense chronic inflammatory infiltrates at places mainly comprising of lymphocytes and plasma cells. (Figure 2a,b). The clinico-radio-histopathological correlation was suggestive of infected nasopalatine duct cyst.

3. Discussion

The nasopalatine duct cyst (NPDC) was first described in 1914 by Meyer⁵ It is also known by other names such as anterior middle cyst, maxillary midline cyst, anterior middle palatine cyst and incisor duct cyst.¹

NPDC is a common non-odontogenic cyst, occurring in approximately 1% of the general population.⁶ These cysts, formally classified as fissural cysts were believed to originate in entrapped epithelium in embryonic fissures during the development of the orofacial region.² The pathogenesis is now believed to arise from remnants of the embryonic nasopalatine duct.⁷

According to Hedge et al, although, the nasopalatine duct cysts may develop at any age, they occur frequently in the fourth to sixth decade of life with a slight male (M:F = 3:1) and Caucasian predominance. Vasconcelos et al too found a higher incidence of NPDC among males than females. (1) In the present case however it was seen in a female patient in her sixth decade.

The etiology of these lesions is not clear; in addition to the hypothesis of spontaneous proliferation from embryonic tissue remains, other possible etiologies have been proposed – including prior trauma, poorly fitting dentures, the existence of local infection, or the influence of genetic and racial factors.⁸ In this patient, no history of trauma or dentures was available, only history of uneventful extractions in that region was obtained. The cyst seemed to be fairly asymptomatic causing a fluctuant swelling on the palate, and the patient only complained of an unpleasant taste and itchy sensation in the nasolabial area, on appearance of a swelling in the same area.

The NPDC is usually detected on routine clinical and radiographic investigations as it is asymptomatic, but in this case the cyst had grown to a massive size so as to perforate the labial cortical plate and obliterate the nasolabial angle. The cyst also seemed to encroach into the nasal cavity.

In establishing a diagnosis of NPDC, it is important to attempt to exclude the possibility of a periapical lesion by testing the pulp vitality of the incisor teeth. In the case presented here, clinically a differential diagnosis of either a radicular or a residual cyst was made. Radicular cyst could be a possibility, as one of the anterior tooth showed severe abrasion and was non-vital. Due to history of extraction of the other anterior teeth, it could be a residual cyst. Radiographically, the typical heart shaped appearance on

the cyst, was suggestive of a NPDC. Diameters ranging from 10 to 40 mm have been reported.⁹ Tariq Salammet al reported an extensive NPDC in a 55 year old female patient with a labial swelling measuring 2 X 2 cm. Tozoglu et al also reported a huge NPDC with a maximum diameter of 30 mm in male patient. In the case reported here, the cyst was of a giant size measuring approximately 4 X 5 cm.

MRI is highly reliable in diagnosing NPDCs, discarding root cysts or any other cysts of odontogenic origin.¹⁰ However, due to economical constraints, this was not advised. The treatment of choice is surgical removal of the cyst, although some authors propose marsupialization of large NPDCs. In this case, surgical enucleation was done and the postoperative recovery was uneventful. The patient is on a routine follow-up for more than one year with no signs of recurrence.

4. Conclusion

NPDCs are of unknown etiology, but should be treated as soon as they are diagnosed to avoid complications of teeth displacement, bone perforation and malignant transformation. The treatment may vary from marsupialization to enucleation to en-bloc resection, depending on the size of the cyst.

5. Source of Funding

None.

6. Conflicts of Interest

There are no conflicts of interest.

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