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CASE REPORT

An Unusually Large Destructive Nasopalatine Duct Cyst: A Case Report

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Abstract A case of an unusually large expansile nasopalatine duct cyst (NPDC) causing extensive destruction of the hard palate with involvement of the nasal cavity, perforation of the alveolar process and mucosa of the maxilla by pressure of the maxillary anterior teeth is being presented.

Keywords Nasopalatine duct cyst · Mucosal perforation · Alveolar destruction · Non odontogenic cyst · Developmental cyst

Introduction

The nasopalatine duct cyst (NPDC) was first described by Meyer in 1914 [1, 2]. It was believed to arise from remnants of the nasopalatine duct, an embryologic structure connecting the oral and nasal cavities in the area of incisive canal [3, 4]. It is one of the most common non-odontogenic cyst, comprising 10% of jaw cysts and occurring in 1 of every 100 persons with a slight male predilection, the mean age being 42.5 years [5, 6]. These cysts are usually asymptomatic, unless they are secondarily infected. The most commonly reported clinical symptom is swelling in the anterior part of the palate. The NPDCs are often confused with median palatine cysts and other cysts in the palatal region [7, 8].

These cysts are situated in, or near the incisive canals, just behind the maxillary central incisors, and can be seen on radiographs in close relationship to the apices of the upper teeth, which may cause confusion with apical lesions [9]. These entities are usually treated with surgical enucleation [6]. The epithelial remnants located in the midsutural area have a potential to differentiate into stratified squamous and or pseudostratified ciliated columnar epithelium. As the cells proliferate they form a solid mass. The most peripheral cells being adjacent to the connective tissue, assume the position and function of the basal layer. As the basal cells divide, the cyst enlarges. The central cells exfoliate into the center of the cyst where they may rupture and release their contents [10].

The causes are not entirely understood and may include the following: Trauma, infections or spontaneous proliferation. Mucous producing glands found in the epithelium could contribute to secondary cyst formation due to secretory function within [11].

Case Presentation

A 26-year-old female presented to our Maxillofacial and Oral Surgery Department complaining of an anterior facial swelling. History taking revealed that the swelling was first noticed about a year ago with a gradual increase in size. She gave no complaints of pain or paresthesia but stated, however, that there was a sudden increase in size of the swelling over the past few days. Extra-oral examination showed expansion and protrusion of the anterior maxilla (Fig. 1a). There was no palpable or visible lymphadenopathy. The swelling was firm, non-tender, non-fluctuant and without pulsations. No bruit was auscultated.

On intraoral examination, maxillary alveolar expansion and obliteration of the labial sulcus from canine to canine with obvious indentations of the maxillary alveolar mucosa

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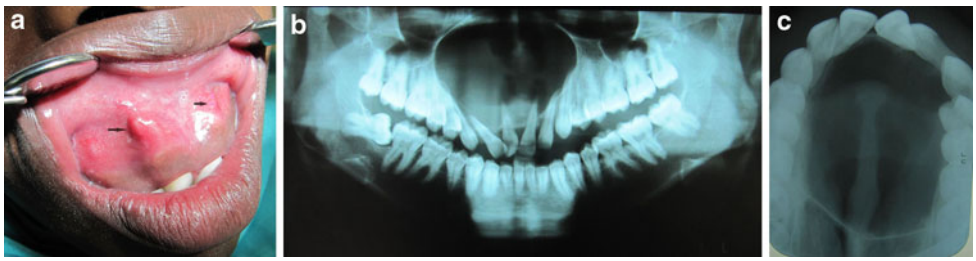

Fig. 1 **a** Intraoral view at initial presentation showing an expansile maxillary alveolus (*arrow*). **b**, **c** Panoral and occlusal radiographs showing the anterior-posterior and lateral bony extent of the cystic lesion

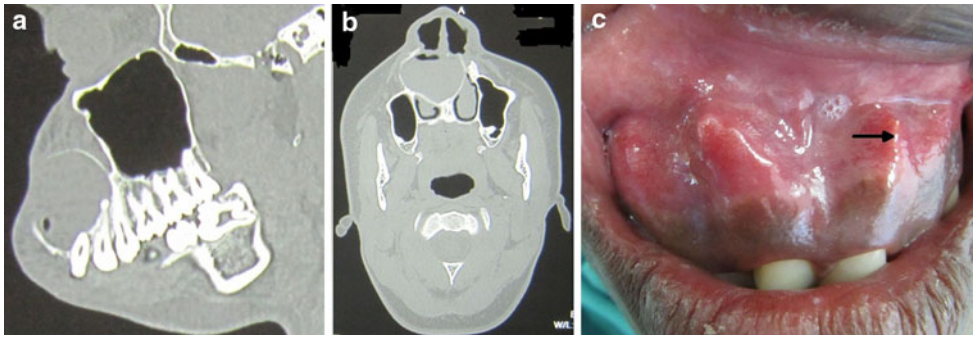

Fig. 2 **a** Saggital and **b** axial CT images. **c** Apical transmucosal protrusion of root canal filling material (*arrow*)

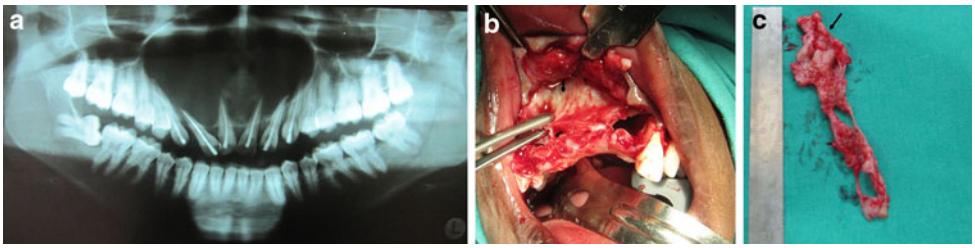

Fig. 3 **a** Post RCT panoral. **b** Intraoperative view of cystic lining (*arrow*). **c** Cyst lining

was noticeable. These indentations appeared to correspond to the outline of the roots of the maxillary incisors (Fig. 1a). Some of the maxillary incisor crowns were mobile with some degree of rotation and malalignment with respect to the maxillary arch and their adjacent teeth. The palatal mucosa appeared normal in colour with a soft consistency posterior to the incisive papilla. The maxillary anterior teeth were vital.

The patient had no pre-existing underlying medical conditions that was being treated or required treatment. Cigarette smoking, alcohol consumption, tobacco or betel nut chewing, nor the use of lime or snuff formed part of her social habits. Baseline hematological investigations included full blood count, urea and electrolytes, liver function tests and erythrocyte sedimentation rate. These were found to be within normal limits.

Imaging studies included plain radiographs as well as computed tomography (CT). The occlusal and panoral radiographs showed a large oval lesion with well-defined sclerotic borders (Fig. 1b, c). CT showed a 31 mm × 38 mm × 51 mm unilocular, expansile lesion in the anterior aspect of the maxilla. The lesion extended superiorly and inferiorly with cortical breakdown of the hard palate and loss of bony support to the maxillary incisors. The lesion communicated with the nasal cavity (Fig. 2a, b).

On the basis of clinical and radiographic findings, a provisional diagnosis of NPDC was made.

Enucleation biopsy of the lesion was planned under general anaesthesia following root canal treatment (RCT) of her maxillary anterior teeth from canine to canine. On her return post-RCT 3 weeks later to the maxillofacial department, intraoral examination revealed that the apices of the maxillary incisors had perforated the buccal cortex as well as the buccal alveolar mucosa, thus exposing the root canal filling material to the oral cavity. The patient confirmed that no root apex or RCT filling material perforation of the mucosa was noticed before or after RCT. The lesion appeared to have grown larger in size radiographically (Fig. 3a).

Under general anaesthetic, a friable hemorrhagic cyst like lining was enucleated from the bony walls as well as from the palatal mucosa and the exposed nasal septum (Fig. 3c). Due to the increased mobility and poor prognosis of the anterior teeth, extractions were done from canine to

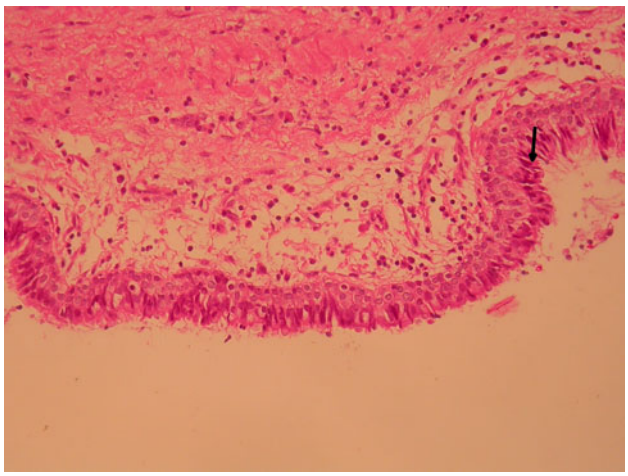

Fig. 4 Micrograph showing cystic cavity lined by non-keratinizing squamous epithelium with ciliated pseudo-stratified columnar epithelium

canine so as to facilitate an easier mucoperiosteal flap raise of the already thinned and perforated mucosa (Fig. 3b). The cavity left behind by the cyst was approximately 4 cm × 5 cm with an exposed nasal septum. The mucoperiosteal flap was sutured back in position. The patient was reviewed and discharged the following day on antibiotic and analgesic cover. Weekly reviews were planned to monitor the progress of the patient. However, the patient was lost to follow up after the third weekly review. No complications were noted at the last review. The orthopantomograph taken after 3 weeks showed a large radiolucent cavity left behind by the cyst.

Micrograph showed a cystic cavity lined by non-keratinizing squamous epithelium with ciliated pseudo-stratified columnar epithelium in areas (Fig. 4) the macroscopic; radiographic and histological findings were in keeping with that of a nasopalatine duct cyst.

Discussion

Nasopalatine duct cysts are the most common developmental, epithelial and non-odontogenic cysts of the maxillae [6, 9, 12–14]. They are cysts derived from proliferation of embryonic epithelial remnants of the nasopalatine duct. It may occur at any age but most commonly seen in the fourth to sixth decades of life [1]. The sensation in the anterior palate is not disturbed following severance of the nasopalatine nerve. The greater palatine nerves innervate most of the palatal mucosa. However, the nasopalatine nerve, which regenerates following its severance, only innervates the incisive papilla and the palatal marginal gingiva of the central incisors [15, 16]. The aggressive nature of these cysts should not be underestimated as they can cause massive destruction and disability. Further, squamous cell carcinoma in the maxilla originating from NPDC was also reported and therefore warrants early removal if diagnosed [17].

The clinical findings, e.g., vital teeth ruled out origin of the lesion from a periapical lesion. Surgical defects associated with vital teeth can heal well without need for RCT [8]. A decision regarding RCT should be based on clinical and radiological findings as well as patient acceptance. In this case it was decided to do RCT of the anterior maxilla from canine to canine. Following root canal therapy, the cystic content was expected to drain spontaneously via the root canals with resultant decompression of the cyst, however, minimal. Whilst there may have been some drainage via the canals, it can be assumed that there was further expansion due to an increase in the internal pressure of the cyst within a short period of time, which was evident in the mucosal perforation of the maxillary alveolar mucosa.

A well-circumscribed, heart-shaped, midline radiolucency that is inter-radicular in location between the roots of vital maxillary incisor teeth is virtually pathognomonic for nasopalatine duct cysts. However, other benign entities that develop within the jawbones with some frequency can mimic nasopalatine duct cysts and should be considered in the clinical differential diagnosis such as:

1. Odontogenic cysts (e.g., lateral radicular cyst, lateral periodontal cyst, odontogenic keratocyst [OKC]).
2. Odontogenic tumors (e.g., ameloblastoma, odontogenic myxoma).
3. Nonodontogenic tumors (e.g., central giant cell tumor, brown tumor of hyperparathyroidism, central hemangioma) [18].

Race findings are controversial, since some studies report the same incidence in both Negroes and whites [2, 4, 8], and any difference between them may be attributable to lesser financial resources among the former [12], which could explain the late presentation of such patients for treatment as in this case.

The radiographic appearance of the lesion (splaying of roots, more than 6 mm, surgeons notation of its location near incisive canal as well as the histopathological report together comprise an almost classic description of a nasopalatine duct cyst. By comparison, other possibilities (e.g., OKC or other odontogenic cysts, median maxillary cyst) become remote.

The decision to use autogenous, allogenic or, osteoinductive materials to fill the defect left behind by the lesion was based on patient acceptance and institutional resources. Osteogenesis of a bony defect in the jaws begins with the formation of a blood clot, which is later replaced by osteogenic granulation tissue. In a small defect, the blood clot is replaced by immature bone within a few weeks. This process is completed by osteoblasts, which differentiate

from the endosteum or marrow spaces, with little or no part being played by the periosteum. Larger bony defects may fill in from the periphery over a period of many months. Incomplete osteogenesis, mainly in the maxilla, may be seen and such defects may fill with scar tissue partially or completely. There is little cancellous bone in the maxilla that can contribute to endosteal osteogenesis and the likelihood of complete bony repair may be reduced in large cavities when both cortical plates are perforated [19].

Included amongst the various options for the treatment of cystic defects believed to stimulate new bone fill may include:

1. A combination of Bioplast® fibrin powder, thrombin, patients blood and antibiotics [20].
2. Platelet rich fibrin [21].
3. Xenograft bone with aspirate bone marrow [22].
4. Recombinant bone morphogenic protein—2 (rh BMP-2) in conjunction with rib graft [23, 24].

It is worth noting that other substances capable of delivering BMPs to tissues for longer periods, such as fibrin and collagen sponges, hydroxyapatite, calcium sulphates and synthetic materials like copolymers have been used as carriers in most studies. They are believed to increase the osteoinductive capacity of BMPs [25].

Financial constraints did not allow treatment by any such methods and hence the cavity was closed. Patients Megakaryocyte-derived BMPs, it is hoped may promote bone growth and repair together with patient replaced osteogenic granulation tissue [19, 26].

Marsupialization of the lesion was not considered due to the size of the lesion, tooth mobility and the destruction caused.

Conclusion

Whilst the destructive nature of the nasopalatine cyst is well known, this case demonstrates the destructive potential of an untreated nasopalatine cyst, which caused destruction of bone, mucosa, and movement of teeth and disability. To the best of the author's knowledge, no such phenomenon has been reported in which a single nasopalatine duct cyst had caused tooth displacement, destruction of the palate and labial bone, perforation of the labial mucosa with exposure of root canal filling materials that manifested itself transmucosally in the oral cavity. Early diagnosis and treatment is needed.

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