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A swelling in disguise: A rare case of nasopalatine duct cyst

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Abstract
Nasopalatine duct cyst is considered among commonly occurring non-odontogenic cysts. We are reporting here regarding a patient, who is a 26 year old female with nasopalatine duct cyst. She primarily presented with a swelling in the oral cavity for which total surgical enucleation of the cyst was done. Radiographically, a clearly circumscribed radiolucent area with an inverted pear-shape was noted between the maxillary incisor roots. Cyst wall is fibrocartilaginous. Recurrence rate, post-surgery is usually uncommon.

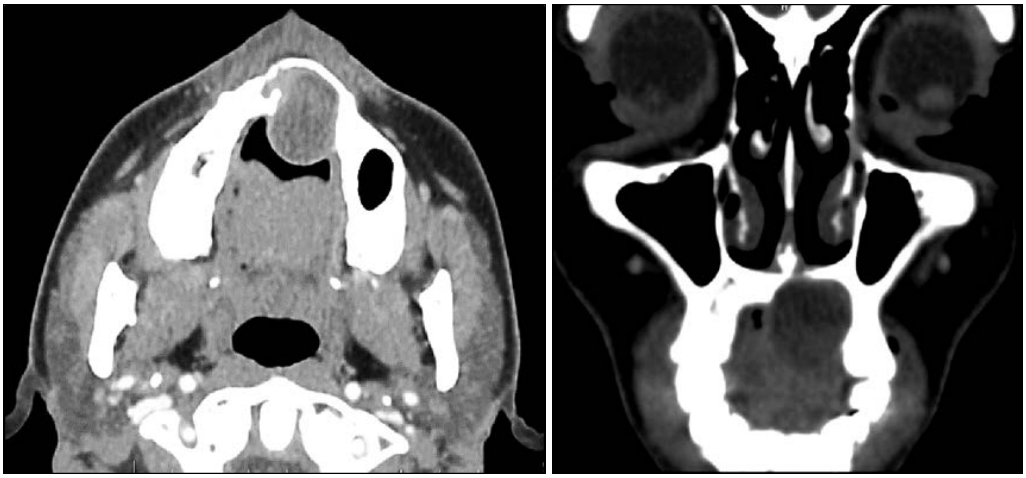
Keywords: Nasopalatine duct cyst (NPDC), maxilla, non-odontogenic, enucleation

Introduction
NPDCs, because of their anatomical location, often known as incisive canal cysts, are developmental abnormalities believed to originate from embryonic nasopalatine duct epithelial remnants, which is a connection between anterior maxilla and nasal cavity in growing foetus within the incisive canal. They are the most commonly occurring nonodontogenic cysts with a higher male predeliction [1]. In 1914, Meyer was first to describe it [2]. According to reports, prevalence of these cysts ranges from 1 - 11.6% of all jaw cysts. NPDCs are found in all age groups but more predominantly seen between 30 and 60 years of age [3].

Case Report
A 26 year old female presented with a swelling in oral cavity for 4 months which gradually increased in size, associated occasionally with mild pain and watery discharge. Intraoral examination revealed a solitary ovoid midline swelling on anterior part of hard palate extending more to left half of palate. The swelling measured about 3x3 cm and was tense cystic in consistency and was nontender on palpation (Figure 1). The mucosa over the swelling was normal. None of the teeth were mobile nor displaced.

Fig 1: Pre-operative image of the nasopalatine duct cyst over hard palate

Contrast enhanced computed tomography of paranasal sinuses was done which showed evidence of well-defined minimal peripherally enhancing hypodense cystic lesion approximately measuring 2.2(AP) x 1.6(ML) x 1.8(CC) cm involving the maxilla in the midline region extending along the left side of the hard palate with overlying bone erosion of the maxilla. (Fig. 2.A and B)

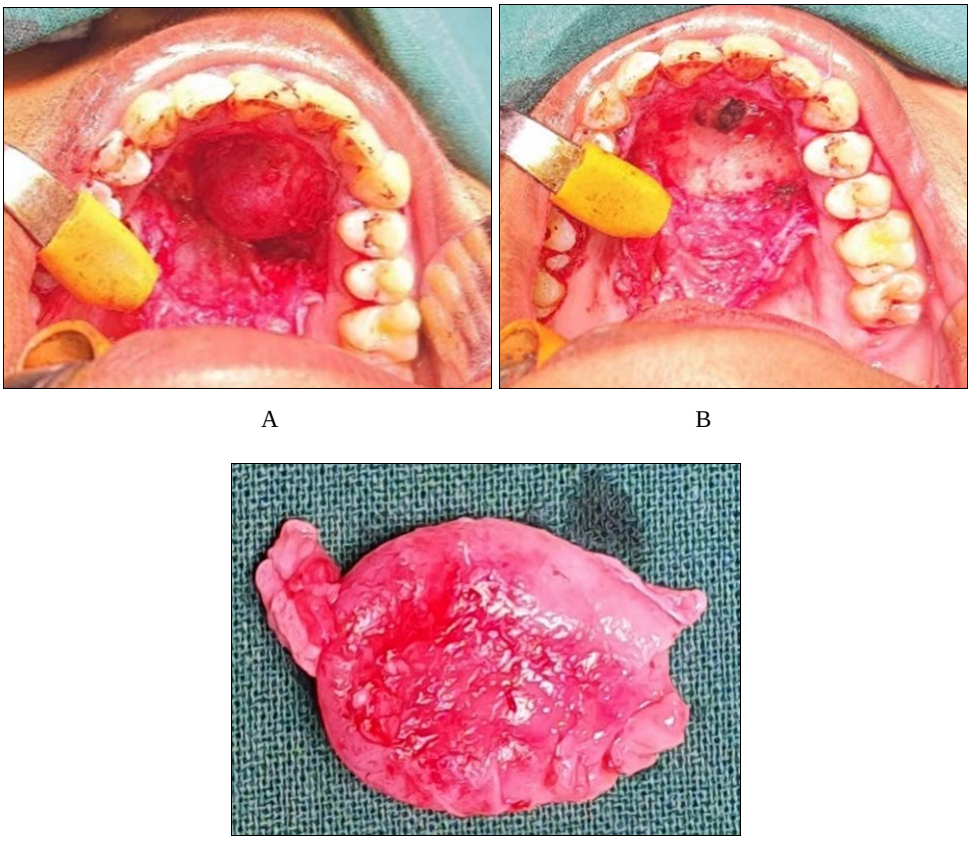


A B

Fig 2: Contrast enhanced computed tomography of paranasal sinuses showing well defined non-enhancing cystic hypodense lesion involving the maxilla in the midline region extending along the left side of hard palate with overlying bone erosion of the maxilla. (A) Axial view (B) Coronal view.

Clinical examination, computed tomography, and fine needle aspiration cytology were primarily utilized to establish the identification of a nasopalatine duct cyst.

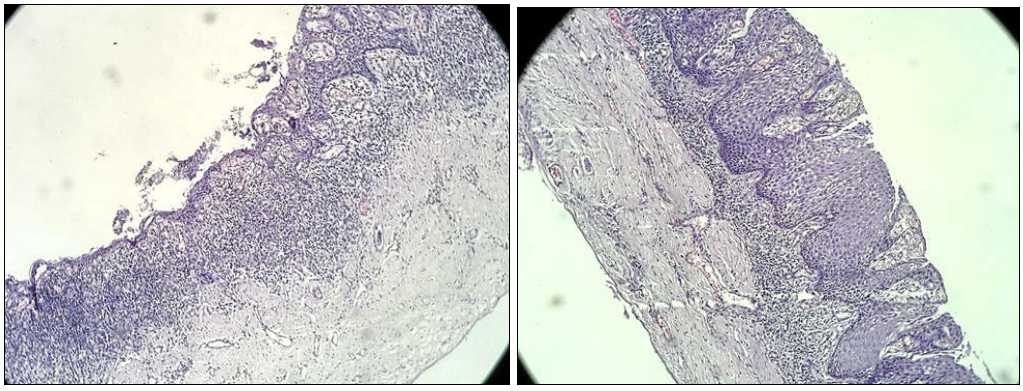
Utilizing transpalatal and intraoral method, complete enucleation of the cyst measuring 2.5 cm in greatest dimension was done (Figure 3. A, B, C). Histopathological examination under 100x magnification by H & E staining of multiple sections taken from the excised specimen showed cyst lined by stratified squamous epithelium and granulation tissue as shown in Figure 4 (A, B). Confirmation of the nasopalatine duct cyst as diagnosis was made. Post-operative period was uneventful.



A B

C

Fig 3: (A) Intra-operative image showing the nasopalatine duct cyst after dissection from surrounding structures. (B) Image showing post enucleation of the cyst with a small area of erosion in the hard palate. (C) Specimen of nasopalatine duct cyst removed in toto.



A B

Fig 4: (A&B) Photomicrograph of H & E stained sections from different areas of the same cyst showing squamous epithelium with epithelial capillary proliferation and dense lymphocytic response in 100x magnification.

Discussion

Nasopalatine duct cyst" was recognized formally in 1971 as a WHO classification, recognizing its connection to nasopalatine duct remnants, vestigial embryonic structure that lies between oral and nasal canals and is connected via incisive canal [5].

Several theories have been suggested regarding the development of nasopalatine duct cysts (NPDCs). NPDCs may be caused by entrapment of epithelial remains upon embryonic fusion of the nasal cavity and anterior maxilla, according to one such idea, known as embryonic epithelial entrapment theory. Growth of nasopalatine duct remnants becomes subject of another theory, which suggests that these cysts develop as result of duct remnants' natural development inside incisive canal.

Additionally, the Wnt signaling pathway (Wingless-type MMTV integration site family) plays a crucial role in craniofacial formation, encompassing nasopalatine duct's development and regression. Cystic degeneration susceptibility can be increased by any disruption of significant pathways, particularly Wnt signaling, Notch signaling, bone morphogenetic proteins (BMPs), fibroblast growth factors (FGFs), and E-cadherin [4].

Most nasopalatine duct cysts (NPDCs) are non-symptomatic and are typically found during routine clinical examinations or radiological imaging. Swelling in center of anterior palate or pain from improperly fitting denture are most common symptoms [5].

In case of an infected cyst, pain and drainage could develop [2]. Pain may also arise due to nerve compression. It can also present with purulent discharge and in cases where cyst is large and found in the top portion of the duct; nasal obstruction can also happen. Trauma, infection, and spontaneous occurrence have been identified as potential triggers for the development of a NPDC. If the size of the cyst is large in the superior part of the duct extending into the nasal cavity, they can also present with nasal obstruction.

A typical radiographic result is heart-shaped radiolucency caused by overlap of nasal septum or anterior nasal spine, or well-defined radiolucent area in center of anterior maxilla close to roots of central incisors [5].

Histologically, NPDCs have been lined with stratified squamous epithelium, either by itself or in conjunction with other forms of epithelium, like simple cuboidal epithelium, simple columnar epithelium, or pseudostratified columnar epithelium (with or without cilia). Epithelial composition varies depending on its proximity to the nasal cavity. Superiorly, the predominant lining is respiratory epithelium, which gradually transitions to cuboidal epithelium and eventually to squamous epithelium as it extends inferiorly toward the oral cavity [4].

Thin, nonkeratinized stratified squamous epithelium often seen in cystic linings is by ciliated pseudostratified columnar epithelium containing goblet cells. In contrast, columnar or cuboidal epithelium lesser frequent and typically consists of 2 to 5 cell layers. Additionally, since nerves and blood vessels frequently pass via the incisive canal, they may be present in some specimens. In certain cases, hyaline cartilage or lobules of minor salivary glands may also be present within the fibrous cystic wall of NPDC.

Malignant cyst transformation has been documented as a potential consequence, however it is uncommon.

Immunohistochemical staining helps to determine the origin of tumor cells but it is not necessary to establish the source of the lining epithelial cells in a nasopalatine duct cyst (NPDC) [6].

Complete surgical excision, usually by a transoral palatal or vestibular strategy, is standard treatment for a nasopalatine duct cyst. In their study, Pei-Wen Wu *et al.* employed "transnasal endoscopic marsupialization" to treat large nasopalatine cysts with nasal involvement, and they discovered that procedure had a very low failure rate. However, for large NPDCs, total excision becomes more challenging and increases the chances of complications in the post-operative period. These may include wound dehiscence, anterior palate paraesthesia, infection, injury to nasopalatine neurovascular bundles, damage to tooth roots, facial swelling, submucosal hematoma, and formation of oronasal fistula. [5] In case of failure to excise an NPDC, it can precipitate infection followed by perforation of the bone and subsequent fistula formation, as stated by Schott *et al.* they also suggested that in cases where denture fixation is required, cyst has to be removed prior to the said procedure, contrary to which trauma can occur which leads to further infection [7]. The recurrence rates of the cysts are usually found to be lower than 2%.

The NPDCs' differential diagnosis comprises of “radicular cysts, lateral periodontal cysts, central haemangiomas and periapical granulomas, median palatal cyst, primordial cysts associated with mesiodens, and rarely, chondrosarcomas which develops from cartilaginous rests native to the incisive papilla or nasopalatine duct region” [4].

Conclusion

A prompt diagnosis along with Histopathological analysis serves as gold standard combined with appropriate care like surgical enucleation, crucial to getting a favourable result for this rare condition. The preferred treatment is enucleation, ensuring complete removal to prevent recurrence. Total excision is necessary, followed by long-term postoperative follow up. In cases of large cysts lacking bony architecture, marsupialization might be considered to reduce risk of a persistent postoperative fistula [2].

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