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Case Report

Dentistry Section

Unusually Large Nasopalatine Duct Cyst: A Case Report

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ABSTRACT

Palatal swellings can at times be a challenging task for clinicians to diagnose, as they may result from various aetiological factors and can derive from the structures within palate or beyond it, either be congenital or acquired in origin. Nasopalatine duct cysts are the most common developmental non-odontogenic cysts account for 10% of maxillary cysts in oral cavity. Though exact aetiology is unknown, these cysts commonly arise from epithelial remnants of nasopalatine duct and usually present as asymptomatic swelling in anterior part of hard palate posterior to the palatal papilla. A unique case report of 28-year-old male patient manifested with a large palatal swelling in the middle of the hard palate with associated symptoms of burning sensation in nose, difficulty while swallowing and change in voice resonance. Radiographic findings revealed well-defined radiolucency in anterior maxilla causing disruption in nasal cavity with histopathological evidence of nasopalatine duct cyst is presented.

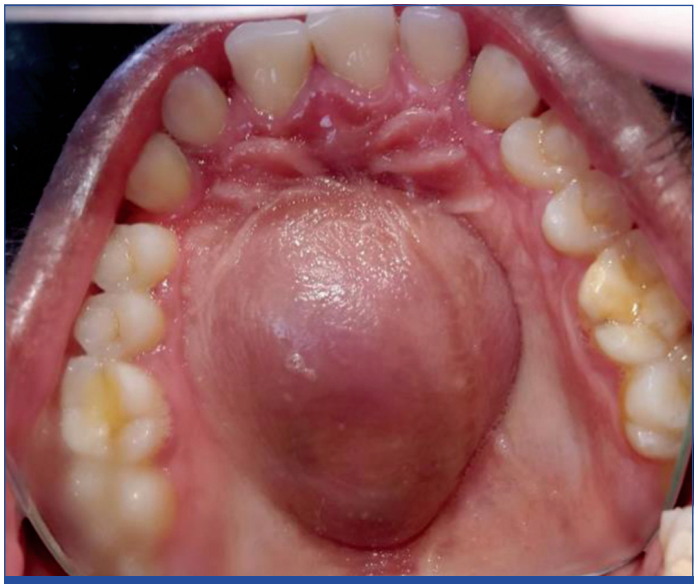
Keywords: Nasal cavity, Non odontogenic cyst, Palatal swelling

CASE REPORT

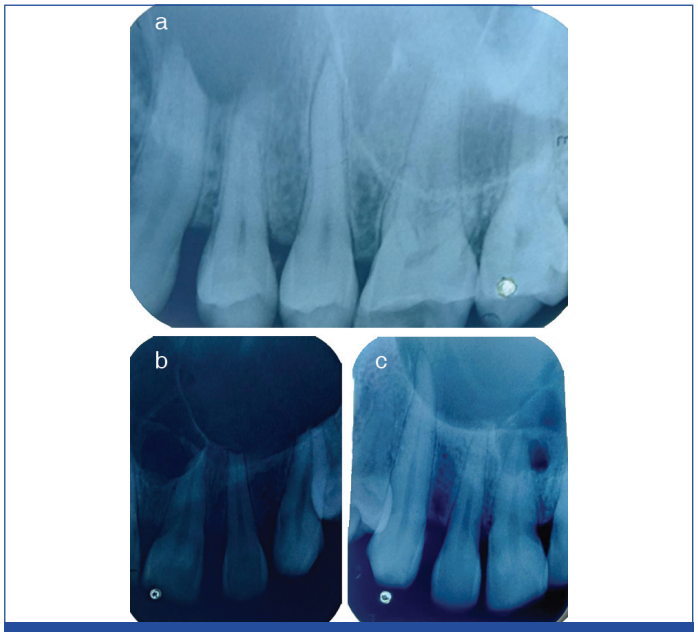
A male patient aged 28-year-old reported to Department of Oral medicine and Radiology with a chief complaint of swelling in his palate which he noticed one month ago. On eliciting the history of presenting illness, patient was apparently normal one month ago. He noticed swelling in his hard palate which appeared large in size. Swelling was insidious in onset and progressed further to attain the present size. Patient revealed that he started feeling discomfort during eating since 5-6 months, also felt burning sensation in nose when he touches the palate since three months. Patient also gave a history of change in voice resonance from past 6-8 months. No h/o pain, pus discharge, paresthesia or trauma was given. Past medical history revealed patient was healthy and had no systemic illness or any deleterious habits.

On extraoral examination, no gross facial asymmetry and no regional lymphadenopathy. Intraoral examination of lesion proper revealed a solitary swelling oval in shape present on the hard palate measuring about 2.5 X 2.5 cm extending anteroposteriorly from posterior aspect of incisive papilla to junction of soft and hard palates, mediolaterally from mid palatine raphe at the center to junction between alveolar ridge and palate on either side. Surface over swelling appears smooth with well-defined margins, no changes in overlying mucosa and surrounding areas were noticed. No pus discharge/sinus tract was seen [Table/Fig-1]. On palpation, all inspection findings were confirmed and the swelling was soft to firm in consistency, and non tender. Other findings include anterior traumatic bite, dental caries irt 46, 47, 37, supra gingival calculus and gingival recession irt 31,41. Electric pulp test were performed which revealed a normal response irt 11,12,13,14,15,16,21,22,23,24,25,26. Based on the history and clinical findings a clinical diagnosis of median palatine cyst and differential diagnosis of Nasopalatine duct cyst and pleomorphic adenoma of minor salivary glands were considered.

The patient was subjected to radiological investigation like Intraoral Periapical Radiograph (IOPAR) irt (23,24,25), (11,12,13) and (21,22,23) revealed well-defined unilocular radiolucency with ill-defined margins. Internal structure was radiolucent, periodontal ligament space appears normal and break in continuity of lamina dura seen in periapical region of 22 23 [Table/Fig-2a-c]. As the lesion was large and to identify its extensions, a maxillary occlusal radiograph was performed which revealed a well-defined ovoid radiolucency extending anteriorly between the apical 1/3rd of central incisors to posterior aspect of 16,26 with

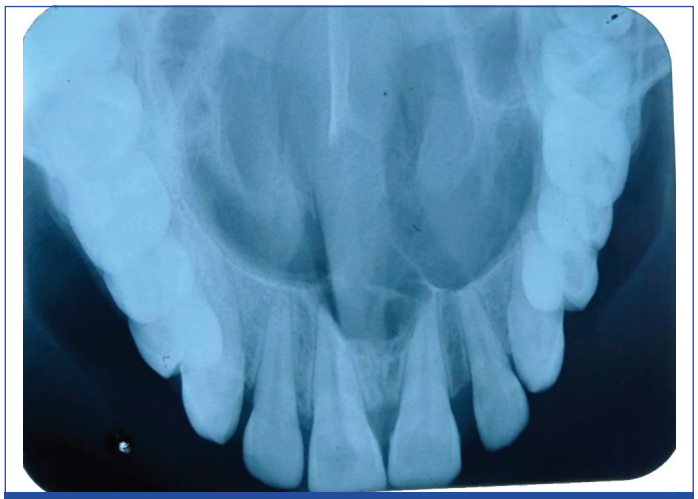


[Table/Fig-1]: Solitary oval shaped swelling in the hard palate.

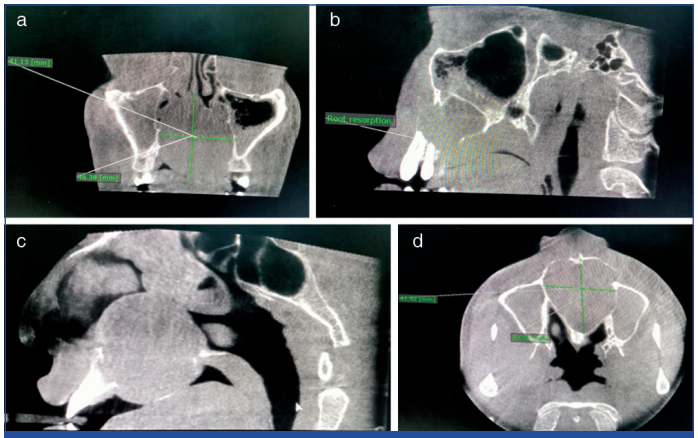


[Table/Fig-2]: a) IOPAR irt 23 24 25; b) IOPAR irt 21 22 23; c) IOPAR irt 11 12 13.

poorly defined sclerotic margins [Table/Fig-3]. To ascertain the findings and visualise the lesion effect on surrounding structures, patient was subjected to Cone Beam Computed Tomography (CBCT) examination of nasomaxillary complex. Coronal section revealed a well-defined radiolucent lesion present in the midline of maxilla between roots of 11 21, which has extended till first molar region on either side with breach in floor of nasal cavity. Entire right maxillary sinus shows opacification [Table/Fig-4a]. Sagittal sections showed the expansion of labial cortical plate and break in palatal cortical plate. Root resorption irt 22 23 24 seen [Table/Fig-4b,c]. Axial section showed- thinning of medial wall of maxillary sinus with complete opacification on right side [Table/Fig-4d]. From CBCT findings nasopalatine duct cyst was considered as radiographic diagnosis.



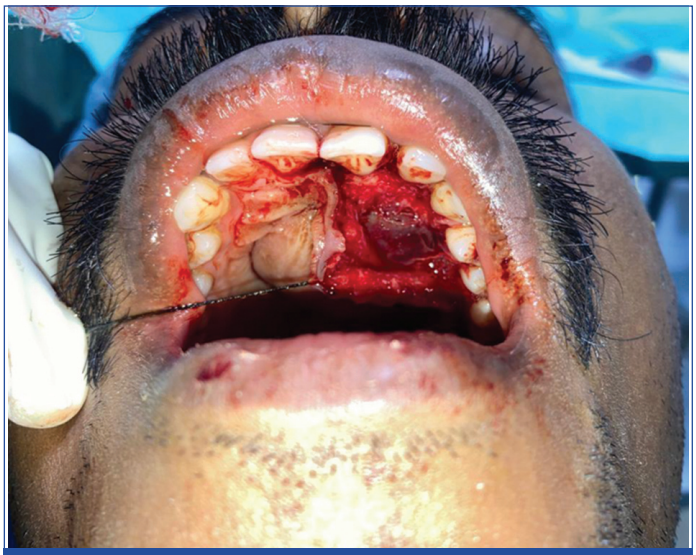
[Table/Fig-3]: Maxillary occlusal radiograph.



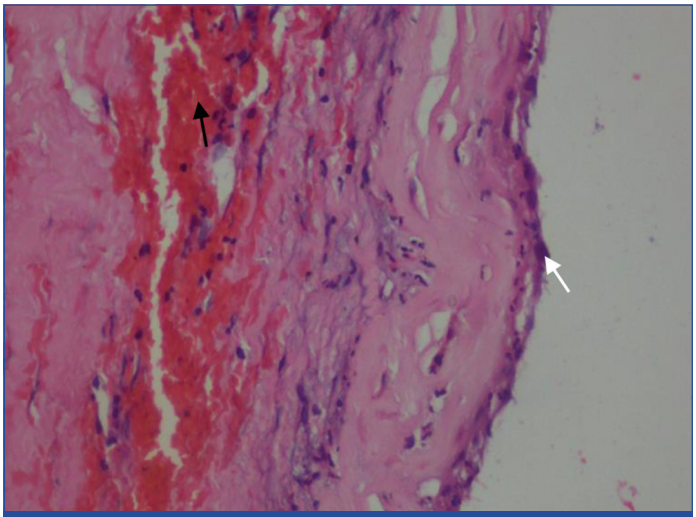
[Table/Fig-4]: a-d) CBCT (Coronal, Sagittal and Axial sections) revealing well defined radiolucent lesion with expansion of labial cortical plate and break in palatal cortical plate.

Hematological investigations were within the normal limits. Fine Needle Aspiration Cytology (FNAC) was performed which showed a dark brown color fluid [Table/Fig-5]. An incisional biopsy of the lesion under local anaesthesia was performed [Table/Fig-6], subjected to histopathological investigation which revealed 1-2 cell thick cuboidal epithelium and connective tissue showed bundles of collagen with chronic inflammatory cells and area of haemorrhage- giving a histopathological diagnosis of nasopalatine duct cyst [Table/Fig-7]. From the above clinical, radiographic and histopathological findings a final diagnosis of Nasopalatine duct cyst was considered. Since the lesion appeared large and extending to the nasal floor, decompression of cystic lesion was performed [Table/Fig-8]. Within six months of follow-up no recurrence of lesion was noted and the drainage tube is changed in every one month follow-up appointment [Table/Fig-9].

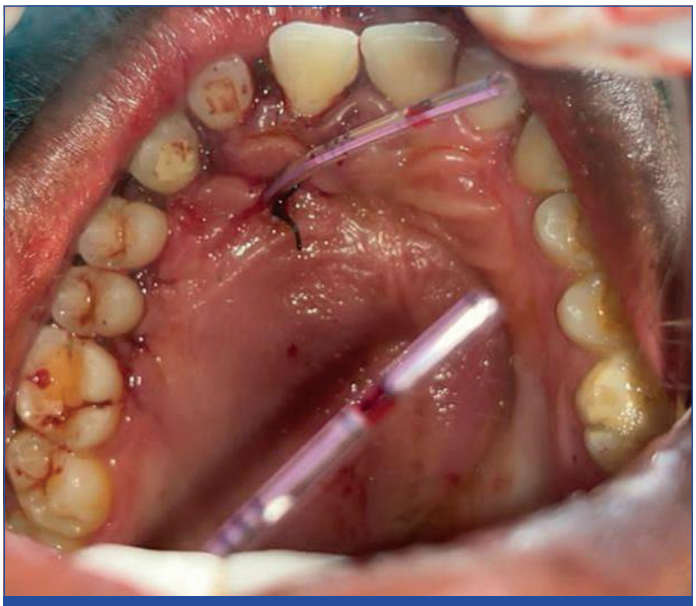
[Table/Fig-5]: FNAC showing dark brown coloured fluid.



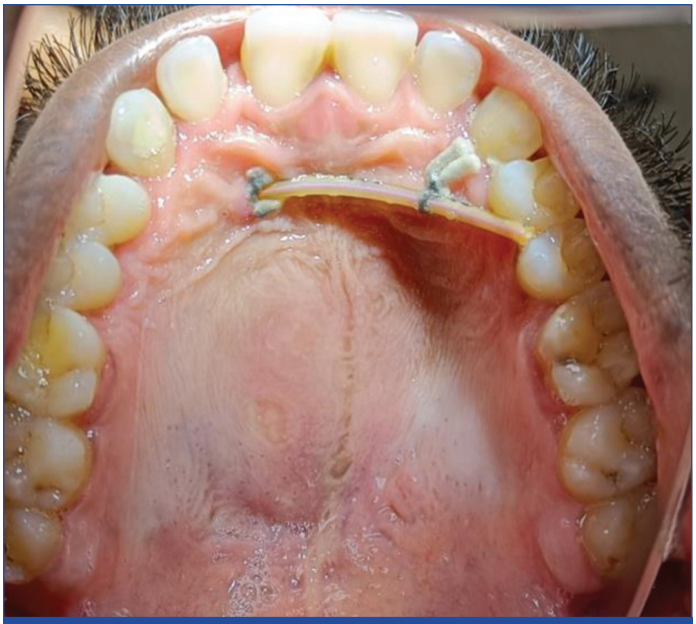
[Table/Fig-6]: Incisional biopsy of the lesion under local anesthesia.



[Table/Fig-7]: High power (40x) photomicrograph showing extravasated RBC (black arrow) and cuboidal epithelium cells (white arrow).



[Table/Fig-8]: Post operative photograph.



[Table/Fig-9]: Follow-up photograph.

DISCUSSION

Meyer in the year 1914 was the first to describe the nasopalatine duct cyst [1]. It is a developmental cyst, epithelial and non-neoplastic in nature [2]. Its location is peculiar, specific and it affects the midline anterior maxilla [3]. It may originate within the nasopalatine canal, at the opening of the canal or in the soft tissues of the palate where it is called the 'cyst of the palatine papilla'. The term 'nasopalatine duct cyst' is preferred to the synonymous 'incisive canal cyst. The aetiology of the nasopalatine duct cysts is unknown, but few of the possible causes could be infection and trauma [4,5]. No history of trauma was recorded as such in our case. In recent years, uncertainty about its origin has been advocated as to whether the 'median palatine cyst' is an entity or the cyst in this region is posterior extensions of nasopalatine duct cysts [3]. These cysts are considered as a fissural cyst which is arising from the epithelium trapped during the embryonic facial processes fusion (Wiesenfeld, 1989) [4]. The side-to-side fusion concept has been eliminated. It is now considered that the Nasopalatine Duct Cyst (NPDC) develops from the embryonic epithelial remnants of the nasopalatine duct and is most common of all the non-odontogenic cyst in maxillofacial region, representing 1.7% and 11.9% of all cysts of the jaw reported by Allard et al., 1981; Berlove, 1956 [6]. NPDC commonly occur in 4th to 6th decades of life with 3:1, male to female ratio and shows predilection for Caucasian individuals [7-10]; whereas in our case, it was noticed in a younger male.

Clinically, it is located specifically in the midline of the anterior maxilla and appears as a swelling measuring few mm to cm in size [11] associated with discharge and salty taste in the mouth. Cysts range in size, with an average diameter of approximately 1.5 cm [12]; wherein case swelling was noted in the mid of palate measuring about 2.5 cm in diameter. These cysts are usually asymptomatic, can undergo secondary infection [13], which was found similar in our case. It may perforate the palatal bony plates and cause tooth displacement. It is often associated with burning sensation and numbness as the nasopalatine nerve is affected [8]. Comparing our case we noticed burning sensation in nose when pressure is applied over the swelling. Pain is an uncommon symptom in cases of NPDC and Vasconcelos RF et al., found that none of the patients reported pain at presentation which was similar in our case [4].

Radiographically, NPDC appears as a radiolucency which is well circumscribed in or close to the midline situated between the roots of the maxillary central incisors and lamina dura continuity is maintained [14]. In our case, it showed a typical unilocular radiolucent lesion in midline at the roots of maxillary central incisors. Occasionally, radiographic differentiation of NPDC with normal anatomical incisive foramen is difficult [7]. The most characteristic radiographic sign of the incisive canal cyst is a partial lack of a cortical margin at the most inferior and superior aspects of some lesions. The distinct appearance is heart shaped due to superimposition of anterior nasal spine over the superior portion of the lesion. The central incisors may be displaced [15], whereas in our case we noticed the perforation in hard palate, breach in the nasal floor and root resorption but displacement of adjacent teeth as such was not noticed.

Histopathologically, it is characterised by stratified squamous epithelium lining alone or in combination with pseudostratified squamous epithelium, simple columnar epithelium and simple cuboidal epithelium. The present case revealed cuboidal epithelial lining in 1-2 cell thickness. The fibrous wall shown blood vessels and nerves with small islands of cartilage and minor salivary gland tissues. If cyst found to be infected chronic inflammatory cells will be seen throughout the specimen [16,17]. The size and the positioning of the cyst determine the ideal surgical approach. However, marsupialisation has been proposed as an alternative treatment for large cysts [9]. In our case, cystic decompression was performed. Long-term follow-up of these cysts are warranted despite being low recurrence rate ranging 0% to 11% [18,19].

CONCLUSION(S)

A case of large nasopalatine cyst in a young individual with asymptomatic swelling on hard palate is presented. With its clinical presentation and considering the extension to maxillofacial complex, it also highlights the role of radiographic investigations like CBCT, which also aids the maxillofacial surgeons to assess the extensions in treatment planning and follow-up.

REFERENCES

[1] Dedhia P, Dedhia S, Dhokar A, Desai A. Nasopalatine duct cyst. Case Rep Dent. 2013;2013:869516.
[2] Shrivastava A, Sur J, Khan F, Prasad S, Roy S. Cone beam computer tomography a crossway in diagnosis of nasopalatine cyst- A case report. Int J Maxillofac Imaging. 2019;5(1):15-18.
[3] Rajendran R, Sivapathasundharam B. Shafer's Textbook of Oral Pathology. 7th edn. Elsevier; 2012.
[4] Tozoglu U, Sumbullu MA, Bilge M. CBCT imaging of extensive nasopalatine duct cyst: A case report. Cumhuriyet Dent J. 2013;16(4):327-30.
[5] Panjwani S, Arora S, Rai S, Malik R. Role of cone beam computed tomography in the prompt diagnosis of a nasopalatine duct cyst. J Indian Acad Oral Med Radiol. 2014;26(3):338-41.
[6] Vasconcelos RF, De Aguiar MCF, Castro WH, de Araújo VC, Mesquita RA. Retrospective analysis of 31 cases of nasopalatine duct cyst. Oral Diseases. 1999;5(4):325-28.
[7] Chatterjee S. Swellings of the palate: A review. Indian J For Med Toxicity. 2020;14(4):9040-45.
[8] Cavalcante IL, Barros CC, Cunha JL, Cruz VM, Pedrosa GA, Santos Ad, et al. Clinicopathologic features of nasopalatine duct cysts: A retrospective study in two Brazilian oral and maxillofacial pathology referral centers. Med Oral Patol Oral Cir Bucal. 2021;26(5):676-83.
[9] Patil S, Khandelwal S, Doni B, Rahman F. A review of 9 palatal swellings. J Pak Dent Assoc. 2013;22(2):133-39.
[10] Angel TP, Mariela PM, Eduardo S, Jose HD, Consolaro A, Rubira-Bullen IRF, et al. Nasopalatine cyst: A silent growing lesion. Biomed J Sci &Tech Res. 2017;1(3):881-85.
[11] Tanaka S, Lida S, Murakami S, Kishino M, Yamada C, Okura M. Extensive nasopalatine duct cyst causing nasolabial protrusion. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2008;106(4):46-50.
[12] Meyer AW. A unique supernumerary paranasal sinus directly above the superior incisors. J Anat. 1914;48:118-29.
[13] Perumal CJ. An unusually large destructive nasopalatine duct cyst: A case report. J Maxillofac Oral Surg. 2013;12(1):100-04.
[14] Aparna M, Chakravarthy A, Acharya SR, Radhakrishnan R. A clinical report demonstrating the significance of distinguishing a nasopalatine duct cyst from a radicular cyst. BMJ Case Rep. 2014;2014:bcr2013200329.
[15] Langlais RP, Langland OP, Nortje CJ. Diagnostic Imaging of the jaws. USA: Williams and Wilkins; 1995.
[16] Nelson BL, Linfesty RL. Nasopalatine duct cyst. Head Neck Pathol. 2010;(4):121-22.
[17] Neville BW, Damm DD, Allen CM, Bouquot JF. Developmental defects of the oral and maxillofacial region. In: Oral and maxillofacial pathology. WB Saunders, Philadelphia 2005.

[18] Oliveira LJ, Leao PLR, Alvarenga RR, Horta MCR, Souza PEA. Nasopalatine duct cyst – diagnosis, treatment and postoperative complications: Report of two cases. J Bras Patol Med Lab. 2017;53(6):407-13.

[19] Lang MJ, Lee YP, Hwang MJ, Chiang CP. Nasopalatine duct cyst - case report. J Dent Sci. 2021;16(3):1047-49.

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