

Nasopalatine canal cyst, a diagnostic twist

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Abstract

We present a case of nasopalatine duct cyst in a 35-year-old female. The cyst was diagnosed based on the presence of only one clinical symptom and no obvious clinical signs, which is a relatively rare occurrence. However, the radiographic and histological presentation of this lesion was typical of a nasopalatine duct cyst. Therefore, this case report aims to highlight the variable presentations of the nasopalatine cyst, which is often misdiagnosed and treated as an endodontic infection.

Keywords: : Nasopalatine cyst, Incisive canal cyst, Cone-beam computed tomography, non-odontogenic cyst.

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Introduction

The incisive canal is a bony conduit through the hard palate, which connects the oral and nasal cavities.¹⁻³ Within this structure lies an epithelium-lined tube, known as the nasopalatine duct. Although the nasopalatine duct offers no utility, it provides the basis for the most common non-odontogenic cystic lesion of the jaws: the nasopalatine duct cyst (NPC).¹ With a prevalence of 1-4%, these lesions are reported to contribute to 80% of oral cystic lesions.¹ Although mostly asymptomatic, once infected, they commonly present as a painful swelling in the mid-palatal region, accompanied with salty discharge.¹ The NPC may also mimic endodontic pathosis, leading to misdiagnosis and unnecessary endodontic intervention.⁴ Interestingly, these lesions are often undetected in the initial stage of the disease which is noteworthy since they tend to enlarge rapidly, involving the adjacent cortical plates, which can be prevented by early diagnosis and intervention.⁵

Although two dimensional radiographs have been frequently used to diagnose NPCs, current guidelines recommend the use of cone-beam computed

tomography (CBCT) to diagnose non-odontogenic pathosis.^{4,6,7} Although these guidelines are relevant when the NPC presents as a lesion that mimics endodontic pathosis, they do not account for the varying and subdued presentation of this cyst, particularly in the initial stage, which may allow early identification and improved management.⁷ Considering this, we present a case of an inflamed NPC which was successfully diagnosed and managed despite having no clinically detectable signs and only one symptom, i.e. salty discharge.

Case Report

A 35-year-old lady with unremarkable medical history, presented to the oral and maxillofacial department at Aga Khan University Hospital, Karachi, on November 20, 2021. She presented with the complaint of a faint discomfort and salty discharge associated with the upper anterior teeth since three to four years. Her extra-oral examination was unremarkable. Intra-orally, a slight redness was noted on the palatal aspect, between teeth #11 and #21 (Figure 1). Moreover, the upper anterior teeth (teeth # 13, 12, 11, 21, 22, 23) showed normal response on pulp sensibility testing using cold test (ENDO-FROST, Coltene™) and electric pulp testing (Gentle Pulse, Pulp Vitality Tester Parkel™). Additionally, there was no



Figure-1: Intra Oral photographs showing upper occlusal (A), Lower occlusal (B), Frontal (C), Right lateral (D) and left lateral (E) views. Diastema of 1 mm is evident between the two central incisors, along with a slight redness on the palatal aspect.

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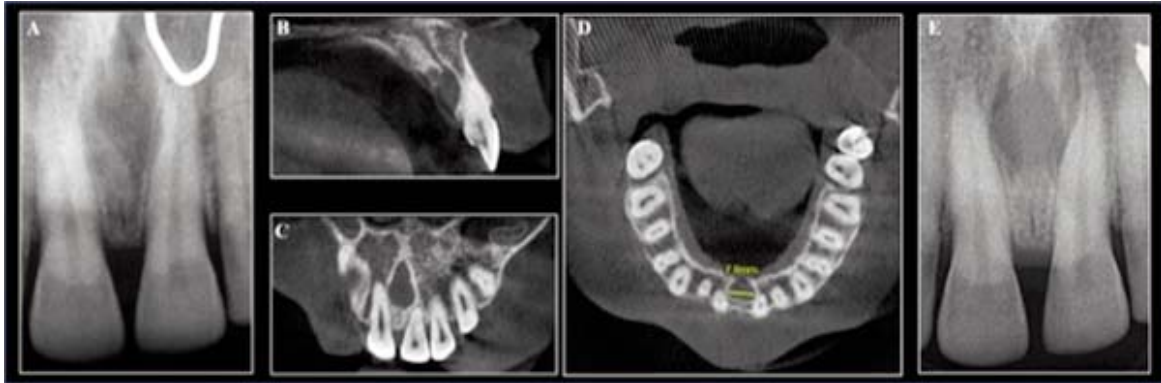


Figure-2: A: Peri-apical X-ray showing teeth #11, 21, with intact periodontal ligament and peri-radicular area. A faint, tear drop shaped radiolucency is noted between the roots, which appear displaced. B, C, D: Cone Beam Computed Tomography (CBCT) sections showing sagittal (B), frontal (C), and axial (D) sections of the nasopalatine canal, with a 7.8 mm width in axial section (D) E: Peri-apical X-ray taken five months after the surgical intervention, showing teeth #11, 21, with intact periodontal ligament and peri-radicular area. A well-defined lesion is evident between the two central incisors with radio-dense area within the lesion.

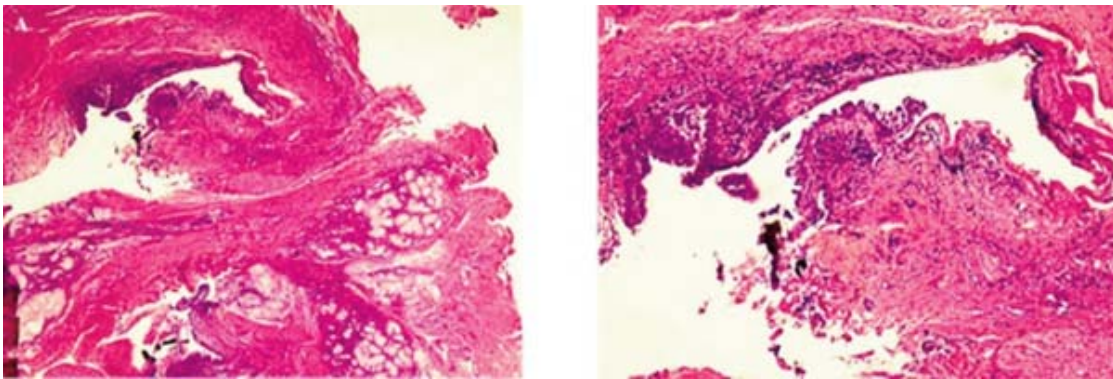


Figure-3: (A): Photomicrograph of Haematoxylin and Eosin stained sections of the cyst lining, showing a well-defined cystic lumen lined with pseudostratified ciliated columnar epithelium (10x10). (B): Inset shows a high powered view of the pseudostratified ciliated columnar epithelium, combined with stratified squamous epithelium and inflammatory infiltrate (40x10)

tenderness on percussion, and the probing depths and mobility were within physiologic limits. A peri-apical X-ray of teeth #11 and #21 revealed a pear shaped radiolucency between teeth #11 and #21 (Figure 2 A). Furthermore, CBCT was also taken, where a nasopalatine canal with a single entry and exit point was noted, classified as type A by Bornstein et al.⁸ (Figure 2 B, C, D). In an axial section taken at the level of the apical third of the roots of the central incisor, the maximum width of the nasopalatine canal was noted, which measured 7.8mm. Sagittal section taken at this point reveals palatal bone erosion, as is noted at the palatal exit point of the canal (incisive foramen). Considering this, a provisional diagnosis of an infected nasopalatine cyst was made and the case was referred to an Oral and Maxillofacial surgeon for enucleation of the cystic lesion under general anaesthesia.

Treatment: Written consent was taken from the patient for the enucleation of the NPC under general anaesthesia.

Moreover, informed consent was taken to publish the case report. Once anaesthetised and under aseptic conditions, a four cornered, full-thickness mucoperiosteal flap was raised from the palatal aspect of the nasopalatine canal. After removing 2mm of the bone, the cyst lining was exposed and the cyst was enucleated in one piece. Gentle irrigation and curettage of the bony cavity was performed, which was then packed with a haemostatic sponge and the flap was re-approximated with interrupted sutures (3-0 vicryl). The removed mass was sent for histopathological analysis, which revealed respiratory epithelium and inflammatory infiltration, confirming the diagnosis of an NPC, as shown in Figure 3 A and B.

Follow-up: A follow-up examination after five months revealed complete resolution of the previously reported symptoms and radiographic evidence of healing, visible as radio dense areas within the well circumscribed radiolucent lesion (Figure 2 E). Additionally, pulp

sensibility testing of the upper anterior teeth (teeth # 13, 12, 11, 21, 22, 23) was re-assessed at the follow-up visits and normal response was noted. There was no tenderness on percussion, and the probing depths and mobility were within physiologic limits.

Discussion

There have been numerous reports of lesions that mimic endodontic pathosis, which have led to unnecessary interventions as a result of misdiagnosis.^{5,9} NPCs most commonly present with patients complaining of a swelling in their palate or of salty tasting discharge that takes away the flavour of food.¹⁰ In severe cases, NPCs cause expansion of the bony cortical plate, causing a swelling in the mid-palatal area between the two central incisors. These cysts may also extend labially and present as a distortion of the labial frenum, or even distort the nasal septum by expanding into the nasal cavity.

In this case, we encountered an NPC which presented with no clinically detectable signs but only one long standing symptom of salty discharge on the palatal surface. It is noteworthy that there are several reports of NPCs that have been misdiagnosed as lesions of endodontic origin, leading to unnecessary root canal treatments. However, a meticulous diagnostic process and CBCT analysis allowed the timely identification and management of the cyst. Interestingly, although the symptoms were subdued, the lesion had already led to thinning of the palatal cortical plate, which further increases the importance of early diagnosis.

Several reports have highlighted the importance of early diagnosis of NPCs using CBCT evaluation. This includes a report by Hilfer et al where the misdiagnoses of a NPC eventually led to the extraction of the upper central incisors.¹¹ Although the current guidelines recommend the use of CBCT for the evaluation of non-odontogenic pathoses, it is critical to note that while these lesions are often misdiagnosed as endodontic pathoses, this is not always the case.⁶ To date, there is a gap in the literature regarding the specific signs that may warrant CBCT assessment, leaving the clinician in a conundrum considering the rule of ALARA (as low as reasonably achievable).⁴ Additionally, although the rate of recurrence has been reported to be low, there is little information on the follow-up protocol for such pathologies.^{1,6}

The present case had an atypical clinical presentation, although the radiographic and histopathological features are commonly encountered. Radiographically, the displaced roots of the central incisors suggested the presence of a pathology, along with a poorly defined

radiolucency present between the roots of the maxillary incisors. Since the upper anterior teeth had a positive response on pulp sensibility testing, the diagnosis of a periapical pathosis was ruled out. However, several differential diagnoses were considered, which included a wide nasopalatine canal, Odontogenic Keratocyst (OKC) and ameloblastoma. In this case, the width of the NPC was measure to be 7.8mm. Considering the normal reported range of 2.62 (\pm 0.91) mm for the Pakistani population, an NPC was suspected.^{3,12} Moreover, the upper central incisor roots appeared to be flared, which is another classic sign of a NPC.⁹ The OKC is a benign odontogenic cyst that commonly presents in the maxilla; anteriorly between the canine and lateral incisor; or posteriorly, around the third molar teeth.¹³ An OKC may be located surrounding the crown of an unerupted tooth, surrounding the periapical region of a root or be present lateral to the root, resembling an NPC.¹³ Moreover, a unicystic ameloblastoma may also present as a well-defined radiolucency in the anterior maxilla with flaring of the adjacent teeth.¹⁴ However, in this case, the biopsy revealed inflammatory infiltration and a cyst lined by respiratory and stratified squamous epithelium, which is diagnostic for a nasopalatine duct cyst.¹²

One limitation of this case is that a radiograph could not be obtained at the two-year follow-up, since the patient had relocated. However, the six-month and one-year follow-up revealed satisfactory healing clinically and radiographically (Figure 2 E). The importance of this case lies in its subdued presentation, which urges clinicians to develop an improved understanding of this pathology. Thorough diagnosis and early intervention may avoid complications of the NPC, such as bone loss and root resorption.

Conclusion

This case report adds to the body of evidence surrounding the variable presentation of the nasopalatine duct cyst. The report highlights the specific signs and symptoms of these lesions to allow early detection and possibly avoid unnecessary endodontic treatment.

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Conflict of Interest: None to declare.

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Authors' Contributions

MK: Conception, design, acquisition, analysis, interpretation of data, drafting, revising it critically.

RG: Final approval and agreement to be accountable for all aspect of the work.

HK: Drafting, revising it critically.