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A Nasopalatine Duct Cyst: A Case Report

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Abstract

This article presents a case of a nasopalatine duct cyst in a 26-year-old Thai male. The patient was referred for proper management of a large radiolucent lesion at the mid-line of the anterior maxilla. The lesion was totally enucleated under general anesthesia. A nasopalatine duct cyst was confirmed after histologic analysis.

Keywords: anterior maxilla, nasopalatine duct cyst, non-odontogenic cyst, radicular cyst

Introduction

Nasopalatine duct cysts are the most common non-odontogenic cyst in the oral cavity, found in 1% of the population. The lesion is developed from remnants of the nasopalatine duct in the incisive canal. It occurs more in male patients in the fourth to sixth decades of life.⁽¹⁾ Clinically, a nasopalatine duct cyst may present with swelling at the mid-line of the anterior part of the palate or a labial part of the alveolar bone. The lesion can be a very large bulging floor of the nose. Pain is often associated with swelling, caused by the pressure on the nasopalatine nerve, or by secondary infection.⁽²⁾ The radiography reveals a well-defined unilocular radiolucent lesion at the incisive region with intact lamina dura of the involving teeth.⁽³⁾ In larger pathology, a heart-shaped radiolucency can be seen due to the superimposition of the nasal spine and the cyst. Tooth displacement may occur in a large lesion. The histology is not characteristics, presenting by the variation of the cystic lining from stratified squamous to pseudostratified ciliated columnar epithelium⁽⁴⁾ and by the presence of medium-sized nerves, arteries, and veins.⁽¹⁾ Epithelial lining diversity is caused by the proximity of the lesion to the nasal cavity and the pluripotential of the embryonic epithelial remnants.^(4,5) The treatment of a nasopalatine duct cyst is total enucleation.⁽⁶⁾ The recurrence is rare.^(1,6) Malignant transformation has been reported, but the incidence is extremely rare.⁽¹⁾

Case report

A Thai male patient, 26 years old, was referred from an endodontist for proper treatment of a large radiolucent lesion at the anterior of the maxilla. Ten years ago, the patient had a motorcycle accident but did not confirm head and neck trauma, then last year the patient had swollen palate and gum at the anterior maxilla. He went to a local clinic, had an emergency endodontic treatment at tooth 21, then was referred to the Endodontic Division, Department of Restorative Dentistry and Periodontology, Chiang Mai University, for endodontic treatment of teeth 11, 12, 21, 22.

When the patient presented at the Department of Oral and Maxillofacial Surgery, his face appeared symmetrical, with no lymphadenopathy, and patent both nares. An intraoral examination showed normal mucosal coverage. There was no shallow labial vestibule, swollen palate, or

bony expansion at the anterior maxilla (Figure 1). Root canals of teeth 11, 12, 21, 22 were treated with mineral trioxide aggregate (MTA).



Figure 1: Intraoral photographs show normal mucosa coverage with no bony expansion.

The periapical radiographs showed a well-defined unilocular radiolucent lesion at the anterior of the maxillary region extending from the periapical area of tooth 12 to tooth 22. In the vertical dimension, the lesion extended beyond the alveolar part of the upper incisors. Root resorption was detected at tooth 11 (Figure 2A).

Cone-beam computed tomography showed a well-defined unilocular hypodensity lesion extending from the alveolar part of the upper incisors to the nasal floor. Buccal cortex and nasal floor perforation were displayed at nasal floor level (Figure 2B). Palatal bone perforation was presented in coronal images (Figure 2C).

The differential diagnoses were given as seen below.

1. Radicular cyst due to the lesion involving pulp necrosis teeth
2. Nasopalatine duct cyst due to the position of the lesion
3. Odontogenic kercyst due to the radiographic findings

Since the radicular cyst was the first differential diagnosis and the incisional biopsy was not performed, the treatment plan was total enucleation of the lesion with root resection of tooth 11 under general anesthesia. The reason behind root resection was because, in the endodontist's opinion, tooth 11 was suspected to be the source of the lesion from the root resorption sign. However, root resorption alone was not enough evidence. It was also possible that the lesion developed from other teeth that were to be seen in the operating room and only one root to be resected.

In the operation, a trapezoidal flap was performed labially extended from teeth 14-24. The barrier bone was removed (Figure 3A), and the lesion was completely enu-



Figure 2: Radiography findings (A) Periapical radiographs show a well-defined unilocular radiolucent lesion at the midline of the maxillary region. There was root resorption at tooth 11. (B) From axial CBCT images, the arrows show labial and nasal floor perforation. (C) From coronal CBCT images, the arrows show palatal and nasal floor perforation.

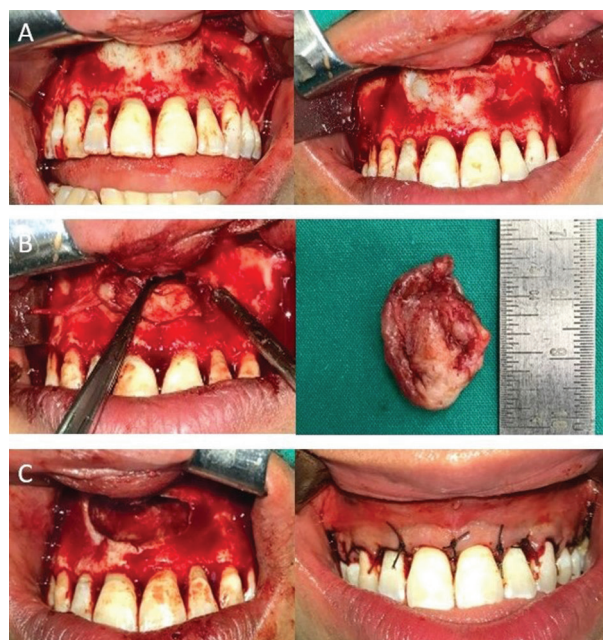


Figure 3: Intraoperative photographs.

cleated (Figure 3B). There was a change of plan. Root resection was not performed, because the lesion did not attach to any tooth. A nasopalatine duct cyst became the first differential diagnosis, but again there was no confirmation that the lesion would be a nasopalatine duct cyst. Gel foams were used to manage death space, and primary closure was done (Figure 3C). The specimen was sent to the Department of Oral Pathology.

Histological microphotographs showed a cystic wall lined by pseudostratified ciliated columnar epithelium with some goblet cells (Figure 4A) or nonkeratinized stratified squamous epithelium. The fibrous connective tissue wall demonstrated a moderate chronic inflammatory cell infiltrate consisting primarily of lymphocytes, plasma cells, and several eosinophils. Nerve bundles (Figure 4C) and variable-sized blood vessels were included within the connective tissue wall (Figure 4B). Despite histology, there was a possibility that the lesion could be a radicular cyst that had an epithelial transition due to its position near the nasal floor. In radiology experts' opinion, it was likely to be a nasopalatine duct cyst given the position of the lesion.

Two months after surgery, the wound had completely healed with no pain. No positive finding was found in the periapical films when compared with the original ones (Figure 5)

Discussion

The differential diagnoses of a nasopalatine duct cyst could be a radicular cyst, a large incisive foramen, and an odontogenic keratocyst (OKC). A radicular cyst can be distinguished from a nasopalatine duct cyst by the response of the pulp sensibility test. The endodontic origin lesion is associated with a non-vital tooth and may present with loss of continuity of lamina dura.⁽³⁾ On the other hand, the nasopalatine duct cyst is usually associated with vital tooth and continuity of lamina dura.⁽³⁾ The challenge would increase if the involving tooth does not respond to the pulp test or has already been endodontically treated,⁽²⁾ which was likely to happen in this case. Other than the response of the pulp sensibility test, altering the horizontal angulation of the periapical radiograph would change the position of the nasopalatine image while the radicular cyst remained the same in its position at the apex of the tooth.^(7,8) The distinction of a nasopalatine duct cyst from a large incisive fossa could be made by size; a normal incisive

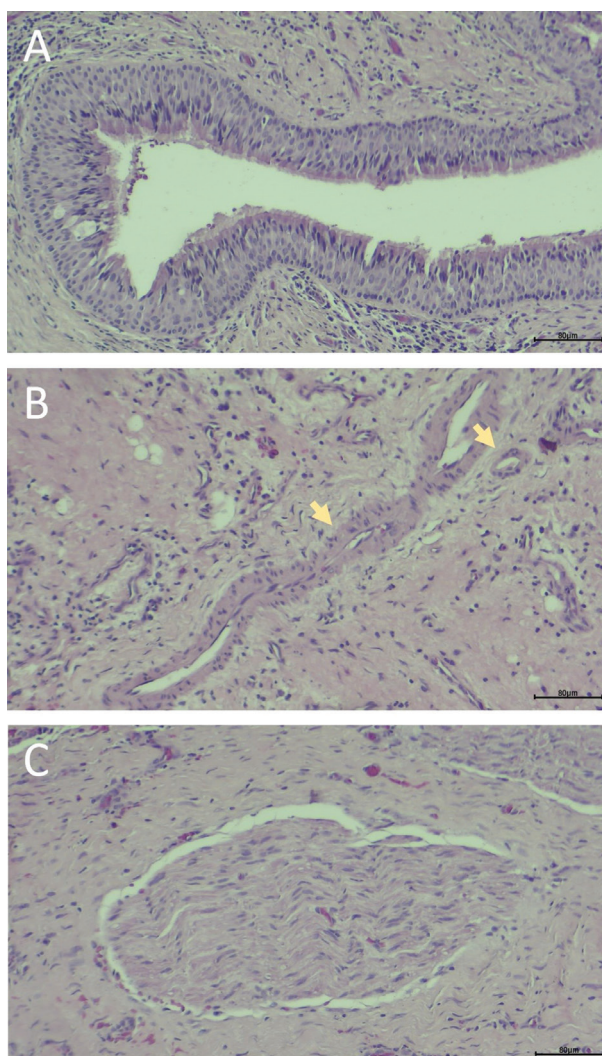


Figure 4: Histological microphotographs (A) Pseudostratified ciliated columnar epithelium (H&E, x100) (B) Variable-sized blood vessels within the connective tissue wall as shown by the arrows (H&E, x100) (C) Nerve bundles (H&E, x100)

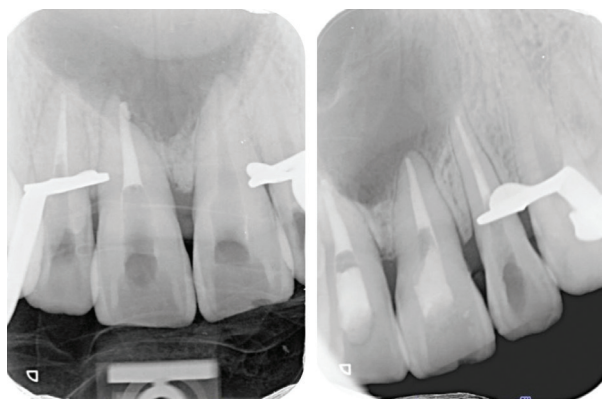


Figure 5: Periapical radiographs, a 2-month follow-up after total enucleation.

foramen can be up to 6 mm,⁽¹⁾ and the lesion exceeding 10 mm in diameter should be regarded as a nasopalatine duct cyst.⁽²⁾ Meanwhile, a radiolucency of 6-8 mm with an ill-defined border is regarded as a large incisive fossa.⁽⁴⁾ Cysts usually present with a well-defined margin and tend to have a superior margin at a higher level than an incisive fossa.⁽⁵⁾ In addition, Aspiration could help distinguish the two lesions.⁽⁴⁾ Lastly, although the midline maxillary region is uncommon for an Odontogenic keratocyst, due to the radiographic data, OKC should be considered as one of the differential diagnoses.⁽⁹⁾

The definitive diagnosis of the nasopalatine duct cyst is based on clinical, radiographic, and mainly histopathologic findings. It was reported that 0.65% of clinical impressions as periapical pathology were of non-endodontic origin from microscopic evaluation.⁽⁶⁾ This emphasizes the importance of microscopic evaluation of all excised tissues, especially in cases where clinical and radiographic findings are inconclusive.

Conclusions

This case report reports the management of a nasopalatine duct cyst and also contains a review of nasopalatine duct cysts, including demography, clinical, radiographic, histopathology, differential diagnoses, and treatment.

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Conflicts of interest

The authors declare no conflicts of interest.

References

1. Lang MJ, Lee YP, Hwang MJ, Chiang CP. Nasopalatine duct cyst - case report. *J Dent Sci.* 2021;16(3):1047-9.
2. Ricucci D, Amantea M, Girone C, Feldman C, Rocas IN, Siqueira JF, Jr. An unusual case of a large periapical cyst mimicking a nasopalatine duct cyst. *J Endod.* 2020;46(8):1155-62.
3. Gnanasekhar JD, Walvekar SV, Al-Kandari AM, Al-Duwairi Y. Misdiagnosis and mismanagement of a nasopalatine duct cyst and its corrective therapy. a case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 1995;80(4):465-70.
4. Allard R, Van Der Kwast W, Van Der Waal I. Nasopalatine duct cyst: review of the literature and report of 22 cases. *Int J Oral Surg.* 1981;10(6):447-61.
5. Abrams AM, Howell FV, Bullock WK. Nasopalatine cysts. *Oral Surg Oral Med Oral Pathol.* 1963;16(3):306-32.
6. Elliott KA, Franzese CB, Pitman KT. Diagnosis and surgical management of nasopalatine duct cysts. *Laryngoscope.* 2004;114(8):1336-40.
7. Vosough Hoseini S, Moradzadeh Khiavi M, Aghbali A, Lotfi M, Sarvani S, Kadeh H. Unilateral nasopalatine duct cyst, differential diagnosis and treatment plan: a case report. *J Dent.* 2011;12(1):95-9.
8. Srivastava S, Misra N, Agarwal R, Pandey P. Nasopalatine canal cyst: often missed. case reports. *BMJ Case Rep.* 2013;2013:bcr2012007548.
9. Neville BW, Damm DD, Allen CM, Chi AC. *Oral and maxillofacial pathology*: Elsevier Health Sciences; 2015.