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Managing a symptomatic large nasopalatine duct cyst – a case report

Abstract:

Nasopalatine duct cysts (NPDCs) are the most common non-odontogenic cysts of the maxilla, originating from the remnants of the nasopalatine duct. Typically asymptomatic, these cysts can cause swelling, pain, and discomfort when they enlarge, often affecting the anterior maxillary region. NPDCs are usually detected incidentally on radiographic examinations, but larger cysts may present with symptoms requiring intervention. This case report presents a 38-year-old male with no systemic diseases who visited Kocaeli Health and Technology University, Faculty of Dentistry with severe pain and swelling in the anterior maxillary region. Clinical examination revealed fluctuation in the palatal mucosa, while cone beam computed tomography imaging showed a homogenous hypodense lesion with sclerotic borders, adjacent to the nasal cavity floor with evidence of bone destruction and perforation of the palatal cortex. The patient underwent an excisional biopsy, and histopathological examination confirmed the diagnosis of a NPDC. This case is distinctive due to the unusually large size and symptomatic presentation. Notably, pain is a rare occurrence in NPDCs.

Keywords: Duct cyst; Cone-beam computed tomography; Maxillary; Odontogenic.

INTRODUCTION

The nasopalatine duct cyst (NPDC) represents the most frequently encountered nonodontogenic cyst in the oral cavity¹. This developmental cyst originates from residual epithelial remnants of the nasopalatine duct, which is located within the incisive canal of the maxilla. The nasopalatine duct, an embryological structure, typically regresses during fetal development, but when remnants persist, they may give rise to cystic formations later in life^{1,2}. Though benign in nature, NPDCs can result in significant clinical symptoms, including swelling, pain,

Statement of Clinical Significance

This case highlights the unusual symptomatic presentation of pain in a nasopalatine duct cyst, a typically asymptomatic condition. This case emphasizes the importance of early identification of nasopalatine duct cysts before the onset of pain, which is an uncommon symptom. In large lesions, as presented in this case, CBCT is essential for accurate diagnosis. In this case report, CBCT played a crucial role in revealing significant bone destruction and perforation, directly influencing the treatment plan. The use of CBCT allowed for a more detailed assessment of the lesion's extent, guiding the clinical approach. Additionally, when pain is present, a vitality test is recommended to rule out endodontic involvement. In this case, all vitality tests returned normal results, and therefore, endodontic treatment was not required. This emphasizes the importance of both CBCT imaging and clinical testing in the comprehensive evaluation of nasopalatine duct cysts.

and pressure in the anterior maxillary region, especially when the cyst enlarges. Given its proximity to critical anatomical structures such as the nasal cavity and the palatal cortex, early detection and treatment are crucial to prevent complications like bone destruction and secondary infections.

CASE REPORT

A 38-year-old male, with no known systemic diseases or chronic drug use, was referred to the Outpatient Clinic of the Faculty of Dentistry at Kocaeli Health and Technology University, presenting with spontaneous severe pain for three days. in

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Received on September 28, 2024. Accepted on November 6, 2024.

<https://doi.org/10.5327/2525-5711.266>



the anterior maxillary region. Intraoral examination revealed swelling in the palatal mucosa, characterized by fluctuation and tender on palpation (Figure 1A). An electric pulp test was conducted on the maxillary anterior teeth (from canine to canine) for vitalometric examination.

Radiographic assessment included panoramic radiography and cone-beam computed tomography (CBCT) (Figure 1B, 1C). Axial CBCT sections showed no evidence of root resorption (Figure 1D). The lesion appeared as a homogenous hypodense area with sclerotic peripheral borders. The lesion was adjacent to the floor of the nasal cavity, where bone destruction was noted. Additionally, palatal cortical bone destruction and perforation were observed. The perforated bone measured approximately 18 mm, while the lesion itself measured 18 x 16 x 9 mm in the CBCT sections (Figure 1E, 1F). Based on these radiographic findings, a preliminary diagnosis of a nasopalatine duct cyst was made. The patient was scheduled for an excisional biopsy at a later date.

The excisional biopsy was performed under local anesthesia. A mucoperiosteal flap was raised, and white-yellow cystic fluid drainage was observed (Figure 2A). The cyst cavity was curetted, and the remaining cystic tissue was placed in formalin for histopathological examination (Figure 2B). Postoperatively, the patient was prescribed antibiotics and non-steroidal anti-inflammatory drugs (NSAIDs) for pain management and infection prevention.

Histopathological analysis revealed that the cyst wall consisted of connective tissue rich in collagen fibers, containing vascular and neural structures. The inner lining of the cyst was composed of cylindrical epithelium of respiratory epithelium in some areas and thin, multilayered non-keratinizing squamous epithelium in others (Figure 3). The definitive diagnosis of the lesion was confirmed as a nasopalatine duct cyst. During the follow-up appointment the next day, the patient reported no pain. At the one-week follow-up, sutures were removed, and the patient was found to have no postoperative complaints (Figure 4).

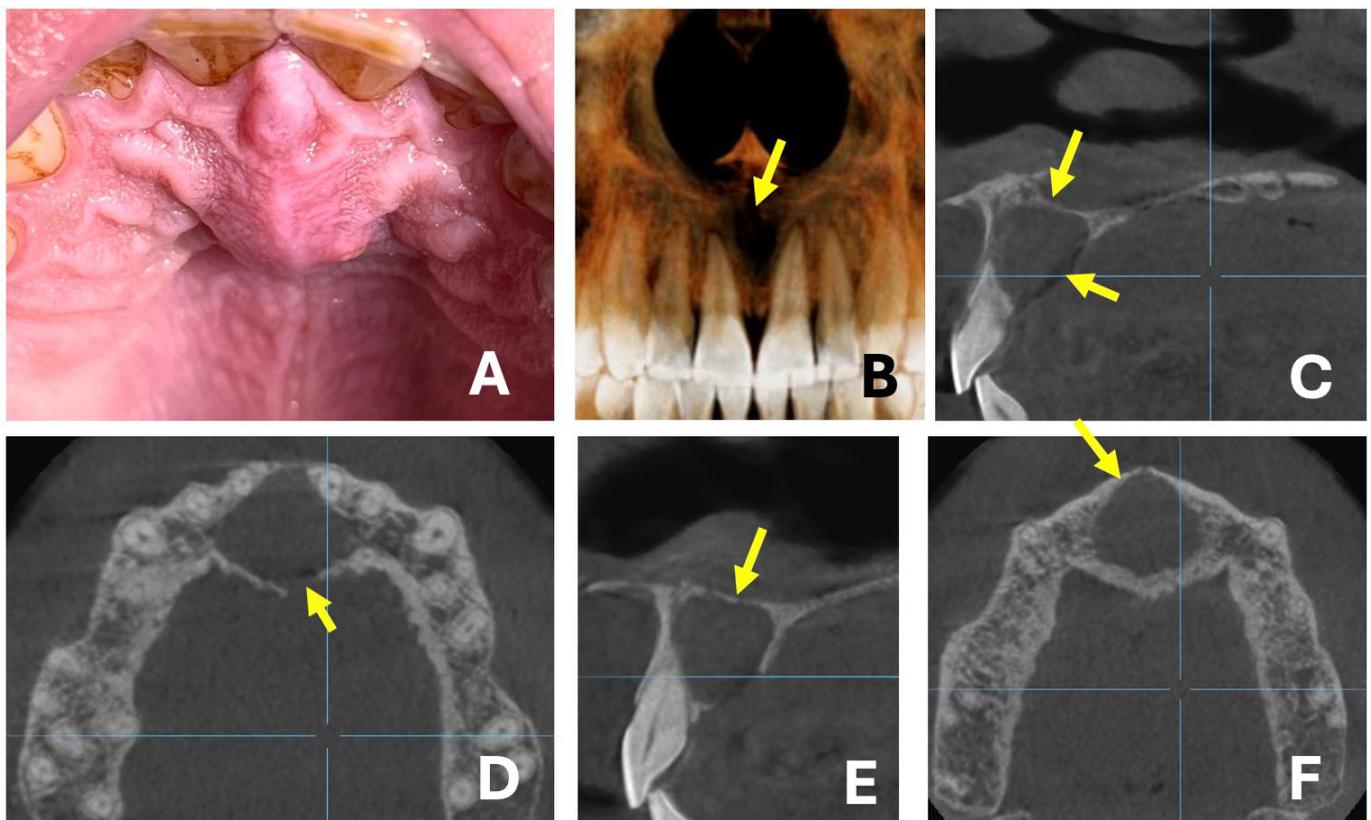


Figure 1. (A) Intraoral view demonstrating the swelling in the palatal region. (B) 3D reconstruction of the maxilla illustrating the intrabony lesion in the anterior maxillary region. (C) Arrows indicate the areas of bone destruction observed in the floor of the nasal cavity and the palatal cortex in sagittal CBCT section. (D) The perforation of the palatal cortex is clearly visible in axial CBCT section. (E) The lesion is located adjacent to the nasal cavity as shown in sagittal CBCT section (F) The lesion extends and breaches the labial cortex of the maxilla as shown in axial CBCT section.

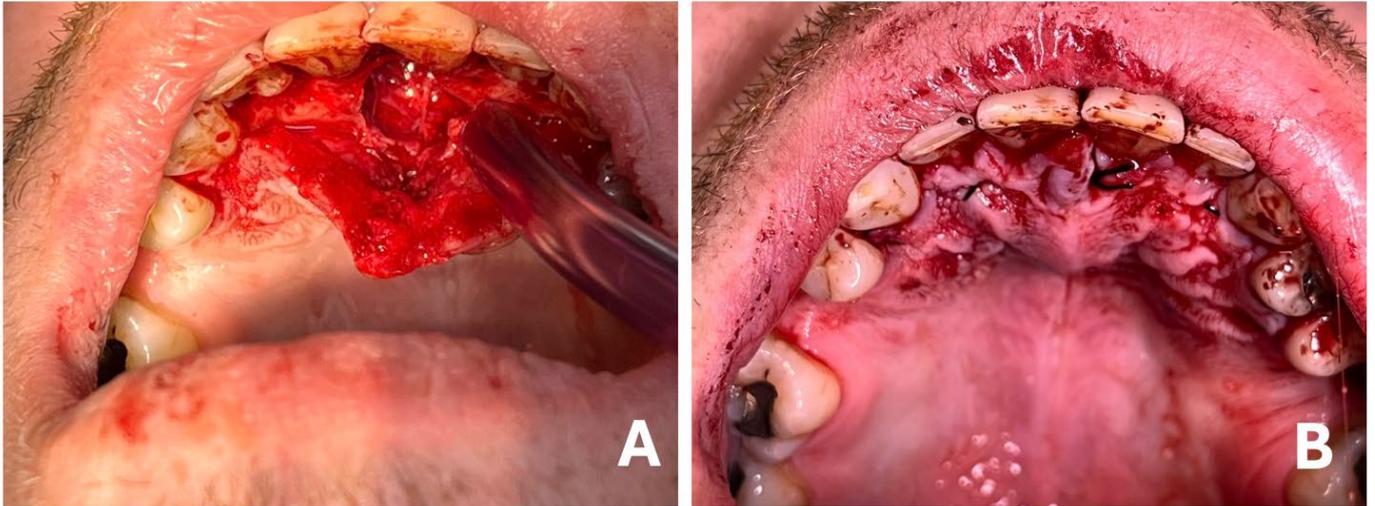


Figure 2. (A) The bony cavity of the lesion following excision. (B) Postoperative appearance of the sutured site. (C) One week after the surgery.

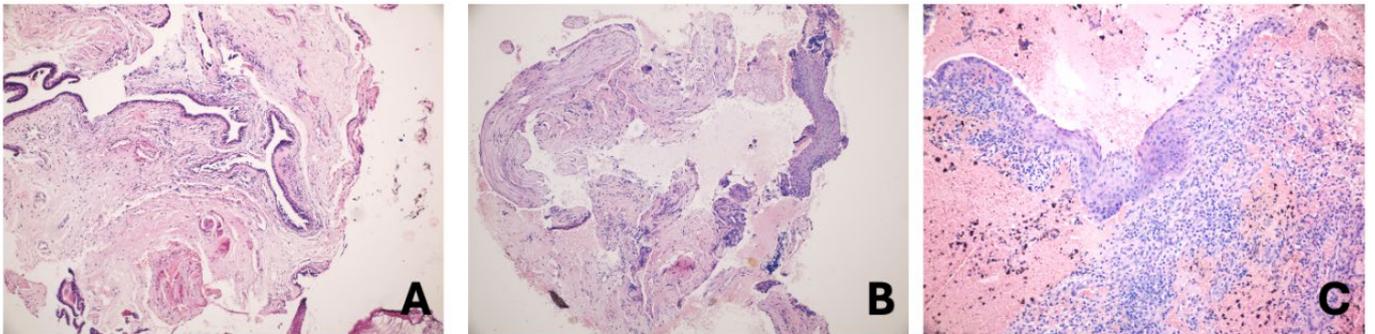


Figure 3. (A) Vascular-rich connective tissue containing nerve sections is lined with cylindrical respiratory epithelium (H&E X100). (B) The inner surface of vascular-rich connective tissue containing multiple nerve sections is lined with stratified squamous epithelium (H&E X100). (C) Between hemorrhagic areas, connective tissue with lymphoplasmacytic cell infiltration lined by stratified non-keratinizing squamous epithelium (H&E X200).

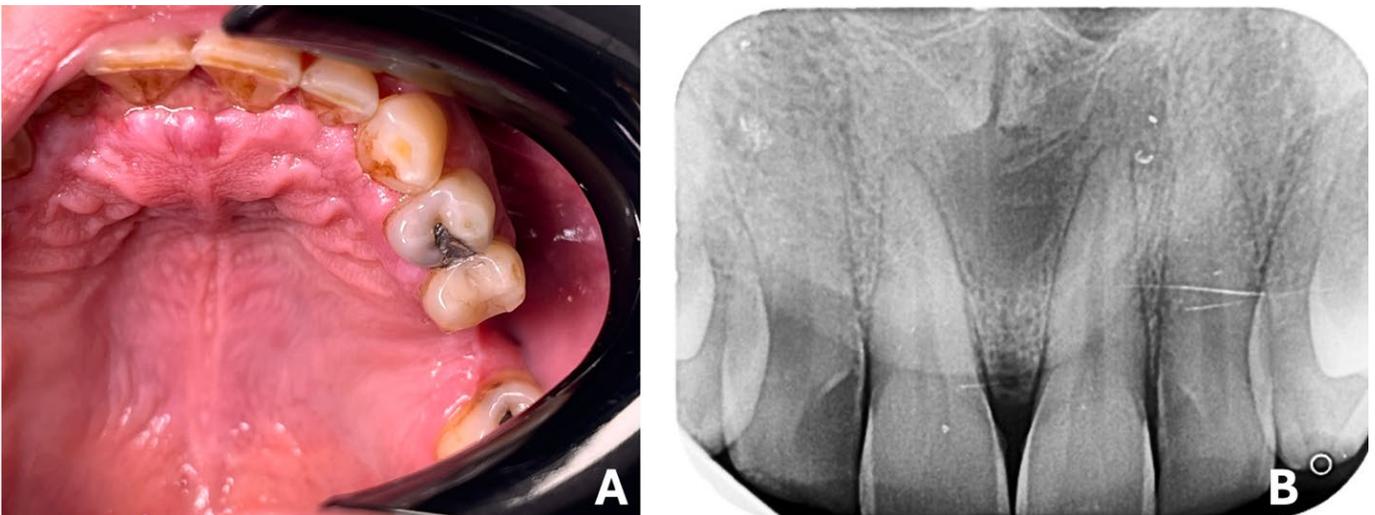


Figure 4. One week following surgery, (A) the oral mucosa demonstrates satisfactory healing, and (B) the periapical radiograph reveals a radiolucent area corresponding to the empty cystic cavity in the anterior region.

DISCUSSION

The nasopalatine duct, also referred to as the incisive canal, is an embryological structure that serves as a connection between the oral and nasal cavities. It is located in the midline of the maxilla on the palatal aspect. The nasopalatine neurovascular bundle traverses this duct and exits through the nasopalatine foramen³. While the duct typically closes after birth, remnants may persist in some individuals, leading to the development of a NPDC. First described by Meyer in 1914⁴, the NPDC is also known by various other names, including anterior midline cyst, maxillary midline cyst, anterior middle palatine cyst, and incisive duct cyst, previously classified as a fissural cyst⁵.

NPDC is a non-odontogenic developmental cyst that typically manifests in the anterior maxilla, specifically in the midline region behind the upper incisors. It is the most common non-odontogenic cyst of the oral cavity, and although it is often asymptomatic, it may occasionally present with symptoms such as swelling, pain, or drainage. While the NPDC is one of several pathologic processes that can affect the jaw bones, it is distinct in that it arises exclusively in the midline of the anterior maxilla. This cyst can occur at any age but is most frequently observed in individuals between the ages of 30 and 60^{3,6}.

The nasopalatine ducts typically degenerate over time; however, the persistence of epithelial remnants can later become a source of epithelium that leads to the formation of a NPDC. This may occur either through spontaneous proliferation⁷ or in response to external factors such as trauma (e.g., removable dentures), bacterial infection, or mucous retention³. Additionally, genetic factors have been proposed as a potential cause^{3,6}. The presence of mucous glands within the proliferating epithelium may further contribute to cyst formation by secreting mucin into the enclosed structure⁸. Nevertheless, the exact etiology remains uncertain.

On radiographic images, a nasopalatine duct cyst typically appears as a round or ovoid radiolucency situated between the roots of the central incisors. In some cases, superimposition of the nasal spine may give the lesion a heart-shaped appearance. Most of these cysts exhibit a well-defined sclerotic border^{3,6,8}. Additionally, a prominent incisive canal may sometimes present as a radiolucent area, mimicking an NPDC. It is generally accepted that a radiolucency in the incisive canal larger than 6 mm should be considered potentially

pathological and warrant further investigation^{7,8}. Magnetic resonance imaging (MRI) of NPDCs typically reveals a well-defined cystic lesion in the midline of the maxilla, often between the roots of the central maxillary incisors. MRI offers the advantage of detailed soft tissue characterization without radiation, making it useful for identifying cyst contents and the relationship to adjacent structures⁹. Cone-beam computed tomography (CBCT), on the other hand, provides high-resolution images that clearly demonstrate the bony margins and the extent of the cyst, typically presenting as a round, ovoid, or heart-shaped radiolucency^{10,11}. The size of the cyst is particularly significant, as larger NPDCs often require more extensive surgical intervention, increasing the invasiveness of the procedure. This can lead to a greater risk of complications, a longer operative time, and a more challenging postoperative course for the patient¹¹. In contrast to smaller NPDCs, which are often managed with less invasive techniques, large cysts may necessitate wider exposure and a more complex closure to ensure complete removal and minimize recurrence^{6,7}. Additionally, patients with larger NPDCs may experience more pronounced postoperative discomfort, swelling, and longer recovery times, emphasizing the importance of careful planning and patient counseling prior to surgery¹¹. This case report presents a large and symptomatic NPDC. Despite its large size, no non-vital teeth were observed in this case, and no complications occurred during the surgical procedure or in the postoperative period. This aligns with similar cases reported in the literature^{6,7,11,12}, where large NPDCs have been managed without significant complications. However, given the potential for recurrence commonly noted in larger cysts, routine radiological follow-up will be conducted for this patient to monitor for any signs of recurrence.

Distinguishing a nasopalatine duct cyst (NPDC) from other lesions, such as a radicular cyst, can be challenging, especially in the early stages or when the lesion is near the apices of the central incisors¹¹. Radicular cysts are odontogenic in origin, typically arising from inflammation or infection associated with a non-vital tooth. Radiographically, both NPDC and radicular cysts may appear as well-defined radiolucencies, and their proximity to the root apices can complicate the diagnosis. However, unlike NPDC, radicular cysts are usually linked to a history of dental trauma or decay¹³. The differential diagnosis for NPDC should also encompass other odontogenic and non-odontogenic conditions. Among odontogenic cysts, lateral radicular cysts, lateral

periodontal cysts, and odontogenic keratocysts should be considered. Additionally, odontogenic tumors such as ameloblastoma and odontogenic myxoma, as well as non-odontogenic tumors like central giant cell tumors, brown tumors related to hyperparathyroidism, and central hemangiomas, should also be included in the diagnostic process. The final diagnosis of a NPDC is confirmed through histopathological examination, which typically reveals a cystic lesion lined by stratified squamous or pseudostratified columnar epithelium, often with mucous glands, nerve bundles, and blood vessels in the cyst wall^{3,14,15}.

Performing a vitality test is crucial to avoid unnecessary endodontic interventions^{12,13}. In this case, the patient reported severe pain in the maxillary anterior teeth; however, since the vitality test yielded a positive response, endodontic treatment was deemed unnecessary. Radiological evaluation is key in diagnosing NPDCs, and while panoramic radiography is useful, additional imaging techniques such as periapical and occlusal radiographs, along with computed tomography (CT), are recommended for a more comprehensive assessment. Two-dimensional radiographs often fail to provide sufficient detail. In this particular case, CBCT revealed no evidence of external root resorption. Although significant expansion and perforation of the palatal cortical plate was observed. Perforation of the palatal cortex may have been missed on traditional two-dimensional imaging modalities. This highlights the value of advanced imaging techniques in the accurate diagnosis and management of NPDC.

CONCLUSION

NPDCs, although commonly asymptomatic, can present with significant clinical symptoms when they enlarge, particularly affecting the anterior maxillary region. This case demonstrates the importance of comprehensive diagnostic approaches, including advanced imaging techniques such as CBCT. In this case, the positive vitality test results ruled out the need for endodontic therapy although the patient had severe pain. Additionally, the histopathological confirmation of NPDC underscores the necessity of biopsy in cases where imaging alone may not provide definitive diagnosis.

AUTHORS' CONTRIBUTIONS

MO: Conceptualization, Data curation, Investigation, Writing –original draft, Writing –review & editing.

ANA: Investigation, Methodology, Writing –original draft, Writing –review & editing. VO: Formal analysis, Project administration, Resources, Supervision, Validation, Visualization, Writing –review & editing.

CONFLICT OF INTEREST STATEMENT

Funding: The authors declare that no funds, grants, or other support were received during the preparation of this manuscript.

Competing interests: The authors have no relevant financial or non-financial interests to disclose.

Ethics approval: The patient approved that the data, images and medical history can be used for scientific purposes.

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