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# Coincidence of nasopalatine duct cyst and dentigerous cyst: A case report

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**ABSTRACT**

Dentigerous cyst is one of the most common odontogenic cysts of the jaw that slowly grows around an unerupted tooth crown. Nasopalatine duct cyst is a non-odontogenic common jaw cyst. Co-occurrence of nasopalatine duct cyst and dentigerous cyst in the same location of the same jaw is extremely rare. Odontogenic and non-odontogenic cysts occurring simultaneously in non-syndromic patients is rare. The lesions must be fully extracted by surgery. We report the treatment procedure of a patient suffering from this rare phenomenon.

**Keywords:** Odontogenic cyst; Nonodontogenic cyst; Impacted tooth.

## Introduction

A dentigerous cyst is a benign odontogenic cyst that develops around the crown of an unerupted tooth. It is considered the second most common odontogenic cyst according to the literature [1,2,3]. Dentigerous cysts grow slowly and most patients are asymptomatic [8]. Moreover, this cyst can affect individuals across a wide age range, with the highest frequency occurring between the second and fourth decades of life [4,5]. It is typically discovered incidentally during regular radiography [6,7]. Radiographically, it appears as a well-defined unilocular radiolucency surrounding an unerupted tooth [2]. Enucleation or marsupialization is the treatment of choice for dentigerous cysts, and the selection of the treatment plan is based on various factors such as the cyst's location and size [1,2]. A nasopalatine duct cyst (NPDC), also known as an invasive canal cyst, is one of the most common non-odontogenic cysts in the jaws. It originates from remnants of the embryogenic epithelium of the nasopalatine duct. NPDC typically occurs near the incisive foramen in the anterior midline of the maxilla [9]. Patients with NPDC often experience symptoms such as pain or swelling, although some patients may be asymptomatic, and the cyst may be incidentally discovered during routine radiography. Radiographically, this cyst exhibits a round, ovoid, or heart-shaped radiolucent appearance near the apical part of the maxillary incisors. It is important to note

that NPDC rarely leads to root resorption of the maxillary incisors [10-12]. The simultaneous occurrence of an odontogenic and non-odontogenic cyst in the same site of the same jaw is rare, and to date, there have been no reports of a dentigerous cyst co-occurring with a nasopalatine duct cyst in the PubMed database. This article presents a rare case of these two cysts occurring simultaneously in the maxilla of a 31-year-old male, along with the treatment process.

## Case Presentation

A 31-year-old male presented to the Department of Oral and Maxillofacial Surgery at Shahid Beheshti Medical University with a complaint of a lesion in the anterior region of the maxilla. The lesion was asymptomatic and incidentally detected during routine radiographs. The patient had a maximal inter-incisal space of 40 mm and showed no signs of swelling, tenderness, or drainage. Cone-beam computed tomography (CBCT) revealed a well-defined radiolucency in the midline of the maxilla, surrounding the coronal area of impacted canines measuring approximately 0.2 ×2×0.7cm (Figure 1,2). Both maxillary central incisors had undergone root canal treatment (RCT) and were restored with porcelain-fused-to-metal (PFM) crown bridges. The lesion did not affect adjacent teeth and did not cause any root resorption. A clinical diagnosis of dentigerous cyst was made, and surgical excision of the lesion was proposed. The surgical intervention was performed under local anesthesia using infraorbital block injection and infiltration with 2% lidocaine containing 1:100,000 epinephrine. Crevicular and releasing incisions were made, and the palatal flap was elevated. The lesion was completely enucleated, and the impacted tooth on the right side was extracted. Curettage and irrigation were performed, and the resulting bone defect was filled with demineralized freeze-dried bone allograft (particle size: 500 to 1000μm) and a 2cm×1.5 cm collagen membrane (Hamanandsaz Kish, TRCIR Company, Tehran, Iran) to promote bone regeneration. Simple sutures with 4/0 VICRYL (Supamedical, Tehran, Iran) were placed. The excised tissue was sent to the Oral and Maxillofacial Pathology Department for histopathologic evaluation. The patient was prescribed a comprehensive antibiotic and analgesic regimen, including Amoxicillin 500 mg every eight hours, Novafen every eight hours, and Chlorhexidine 0.2% mouthwash every 12 hours. A soft food diet was recommended for two weeks. After two months, another surgery was performed to extract the left unerupted canine. Similar to the previous procedure, the palatal flap was elevated, crevicular and releasing incisions were made, and the tooth was extracted. Simple sutures with 4/0 VICRYL (Supamedical, Tehran, Iran) were placed. Gross examination of the excised specimen revealed three pieces of brown cystic tissue, with a total measurement of 2.2× 2×0.7cm, showing a corrugated surface and attached to the crown of the canine. Histologic evaluation of the lesion demonstrated a cystic structure lined by thin non-keratinized stratified squamous epithelium (Figure 3). Another cystic structure lined by varying thickness of non-keratinized stratified squamous to respiratory epithelium was also observed. In the underlying connective tissue of the cyst wall, scattered chronic inflammatory cell infiltration, sections of neuromuscular bundles, curetted bone, and hemorrhage were present (Figure 4 A, B). Based on the clinical, radiographic, and histopathologic findings, a definitive diagnosis of nasopalatine duct cyst with dentigerous cyst was established. To assess the surgical outcome after three months, a follow-up CBCT radiograph was obtained. The results showed successful bone regeneration, indicating a satisfactory outcome of the surgery (Figure 5).

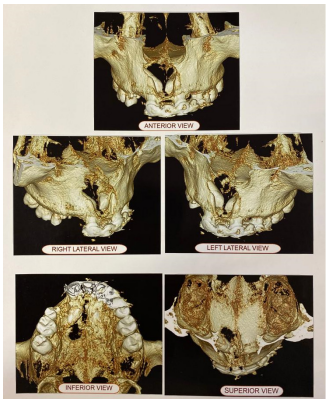


*Figure 1.* The 3D CBCT view shows a well-defined radiolucency in the midline of maxilla, around coronal area of impacted canines measuring about 2×2×0.7 cm.

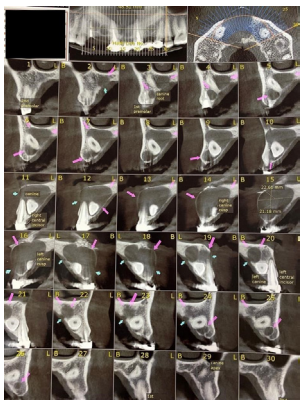

*Figure 2.* CBCT sagittal sections show the precise extent of the lesion and well defined margins.

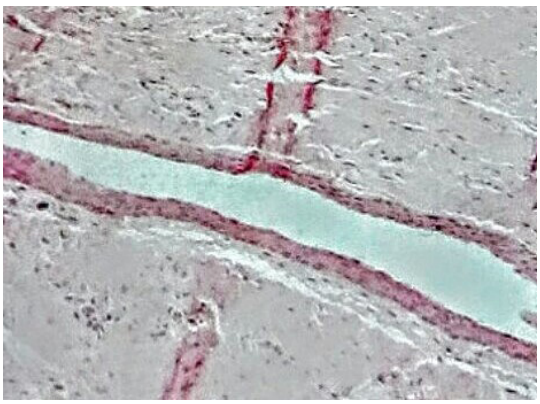

*Figure 3.* Histopathological picture of dentigerous cyst showing the lining epithelium which is thin and non-keratinized, covering the cyst wall (H&E, original magnification×200).

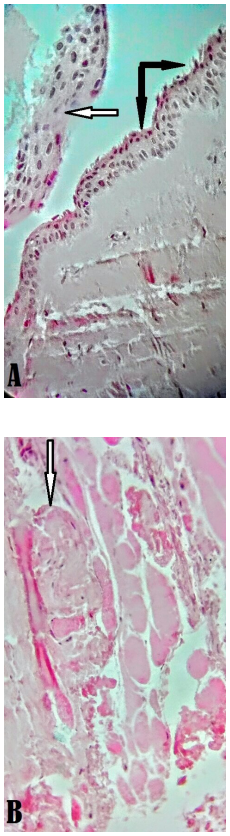


*Figure 4.* Histopathologic sections show (A) Varying thickness of non-keratinized stratified squamous epithelium (white arrow) and respiratory epithelium (black epithelium) (H&E, original magnification ×400). (B) Section of neuromuscular bundle (arrow) and hemorrhage (H&E, original magnification ×400).

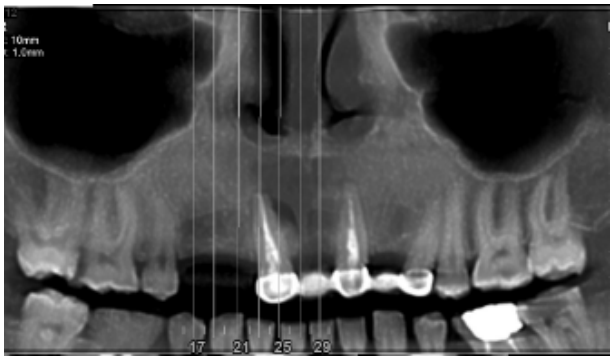

*Figure 5.* CBCT radiograph demonstrates the success of bone regeneration.

## Discussion

In the present research case study, a patient was diagnosed with both a dentigerous cyst (DC) and a nasopalatine duct cyst (NPDC). A dentigerous cyst, also known as a follicular cyst, is characterized by the growth of a cyst within the dental follicle that surrounds the crown of an unerupted tooth. Smaller dentigerous cysts are often asymptomatic and are commonly detected incidentally during radiographic examinations. However, larger cysts, exceeding 2cm in size, may manifest with symptoms such as swelling, tooth displacement, mobility, and sensitivity. Radiographically, dentigerous cysts appear as well-defined radiolucent lesions surrounding the crown of an impacted tooth, exhibiting a clearly demarcated sclerotic border [1,2,3].

On the other hand, the nasopalatine duct cyst originates from the remnants of the nasopalatine duct and typically develops in the anterior maxilla, close to the incisive foramen [11]. Although nasopalatine duct cysts may remain asymptomatic, they can still cause swelling, discomfort, and drainage from the anterior hard palate. Radiographically, nasopalatine duct cysts present as distinct radiolucent lesions along the anterior midline of the maxilla [10–12].

In the radiographic assessments of the aforementioned case, the observed lesion was described as a single radiolucency with a radiopaque rim, leading to the conclusion of it being a cystic lesion. Considering the prevalence of dentigerous cysts, the location and size of the lesion, and the presence of an impacted tooth, a diagnosis of dentigerous cyst was made. Other potential differential diagnoses included glandular odontogenic cyst, which is a relatively uncommon cystic lesion of the jaws with a frequency ranging from 0.012% to 1.3% among all jaw cysts, and unicystic ameloblastoma, which shares similar features but predominantly

occurs in the mandible [20,21]. Although the simultaneous occurrence of multiple odontogenic tumors or central lesions in the jaws has been reported in some cases, it is uncommon in non-syndromic patients. For instance, Kumar et al. (2019) presented a case involving a calcifying ghost cell odontogenic tumor (CGCOT) in a 12-year-old child, with a compound composite odontoma on the right side and an odontoma on the left side of the jaw. The authors proposed that incomplete differentiation of the epithelium and CGCOT on the right side supported the notion that an odontoma can arise as a secondary phenomenon to the development of CGCOT. The treatment and prognosis of CGCOT are influenced by its association with other odontogenic tumors such as adenomatoid odontogenic tumor, ameloblastic fibro-odontoma, ameloblastic fibroma, and ameloblastoma [13]. Additionally, Murgod et al. (2017) reported a case of central odontogenic fibroma associated with a dentigerous cyst, where the formation of the dentigerous cyst was attributed to the displacement of tooth #23 by the central odontogenic fibroma [14]. Bakhtiari et al. (2016) described a case of concurrent compound odontoma with cemento-ossifying fibroma, although further studies are required to establish a definitive connection between these two lesions [15]. Nogueira et al. (2012) reported the simultaneous occurrence of a dentigerous cyst and a persistent cyst in the maxilla, recommending a surgical approach due to the potential for bone resorption, tooth damage, and anatomical tissue impairment, particularly in large cystic lesions [16]. Furthermore, Pushapanshu et al. (2013) documented a case of non-syndromic co-occurrence of a calcifying odontogenic fibroma (WHO type) with traumatic bone cyst (TBC). Calcifying odontogenic fibromas are rare neoplasms that can arise from the dental papilla, periodontal ligament, or dental follicle, depending on the presence of odontogenic epithelium. TBC, on the other hand, has been defined differently by various authors, with research suggesting that it arises due to venous congestion and interstitial fluid drainage obstruction in a region of rapidly resorbing and remodeling cancellous bone [17-19].

## Conclusion

DC and NPDC are both independently occurring lesions that happened in the same area as the incisive canal and impacted canine were located close together. According to Pubmed database, there have never been cases of non-odontogenic and odontogenic cysts coexisting. The two cysts do not associate, making this rare coincidence purely coincidental.

## Conflict of Interest

There is no conflict of interest to declare.

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