Deviated Nasal Septum as a Rare Symptom of Nasopalatine Duct Cysts: Two Case Reports and Review of the Literature

Nazopalatin Kanal Kistlerinin Nadir Semptomu Olarak Nazal Septum Deviasyonu: İki Olgu Sunumu ve Literatür Derlemesi

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ABSTRACT A nasopalatine duct cyst develops from the proliferation of epithelial remnants of embryonic nasopalatine ducts present within the incisive canal and it is the most common nonodontogenic cyst in the maxillofacial region. It is usually asymptomatic and has a tendency to occur in males more than females. Nasopalatine duct cysts are one of the most wrongly diagnosed entities owing to location of incisive foramen. In this case report, we presented two rare cases of extensive nasopalatine duct cyst with the symptoms of nasolabial protrusion and nasal septum deviation. Cone beam computed tomography images and treatment of the lesions were presented. Literature review of the nasopalatine duct cysts will also be discussed.

Key Words: Cystic duct; nasal septum; therapy; nonodontogenic cysts

ÖZET Nazopalatin kanal kisti, insiziv kanal içinde bulunan embriyonik nazopalatin kanallarının epitel kalıntılarının proliferasyonu sonucu gelişir ve maksillofasiyal bölgedeki en yaygın nonodontojenik kisttir. Genellikle asemptomatik olup, kadınlardan çok erkeklerde oluşma eğilimi vardır. İnsiziv foramenin konumu dolayısıyla, nazopalatin kanal kisti, en çok yanlış teşhis edilen vakalardan biridir. Bu vaka raporunda, nazolabial protrüzyon ve nazal septum deviasyonuyla birlikte görülen iki nadir, büyük nazopalatin kist olgusu konu edilmiştir. Konik ışınlı bilgisayarlı tomografi görüntüleri ve lezyonların tedavileri sunulmuştur. Nazopalatin kistlerin literatür derlemesi de tartışılacaktır.

Anahtar Kelimeler: Kistik kanal; nazal septum; tedavi; odontojenik olmayan kistler

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Nasopalatine duct cysts (NPDC) are classified among the epithelial non-odontogenic cysts according to the 1992 WHO histological typing of odontogenic tumors. [1] There have been lots of controversy about the etiology of NPDCs but it is largely accepted that the NPDC arises from proliferation of epithelial remnants of the embryologic nasopalatine duct.[2,3] Increased representation in men was found in several studies.[2-5] Most cases of NPDCs are diagnosed by chance at the time of examination and as they are belonged to non-odontogenic pathoses, the teeth interpreted as the cause of lesion are generally vital. Symptoms are found in 13% to 50% of the patients and typically present as a palatal and/or buccal swelling or a fistula.[2-4]

Most of these cysts develop in the midline of anterior maxilla near the incisive foramen. This article reports two cases with clinical presentation, radiographic and pathological findings, treatment and recurrence rates. One feature of these cases is to present large NPDCs with the rare symptom of nasal septum deviation. To our knowledge, only 2 case reports of NPDC associated with nasal septum deviation have been reported in the literature.[6,7]

CASE REPORTS

CASE 1

A 17-year-old female patient was referred to our hospital complaining of an uncomfortable feeling in the anterior palatal region. The patient mentioned that breathing through the nose was getting more and more difficult during the last couple of months. The patient had no history of a dentoalveolar trauma.

Upon extraoral inspection, a protrusion of the upper lip and the nasal alar bases, which were soft on palpation, were noticeable. There was a slight asymmetry on the upper lip area (Figure 1). The skin color was normal, and no enlarged submandibular, submental, or cervical lymph node was found. There was no sensory disturbance. Intraorally, prominent buccal and palatal swellings were visible, which were painless and resilient on palpation. The overlying mucosa appeared normal in color (Figure 2).

Panoramic radiograph taken showed the extensive round radiolucency with a sclerotic border on the anterior midline maxillary bone. The anterior maxillary teeth were deviated but no distinctive root resorptions were seen (Figure 3). Teeth in the anterior maxilla were vital, left maxillary incisor had been extracted before and she had a bridge on the anterior maxillary teeth. Reacting positively to the vitality test, this finding led us to turn our attention to the nasopalatine duct cyst.

FIGURE 1: Protrusion of the upper lip.

FIGURE 2: Swelling at the anterior palate.

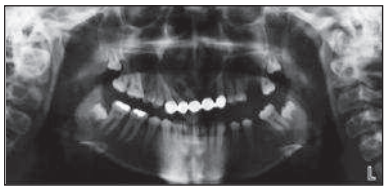

FIGURE 3: Round shaped cyst and displaced roots of the anterior teeth are seen in panoramic radiograph.

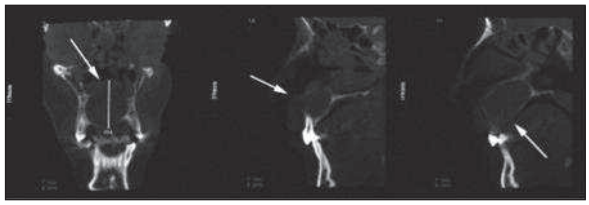

FIGURE 4: Elevated nasal floor on (A), buccal (B) and palatinal bone (C) resorptions on are revealed by CBCT scan images.

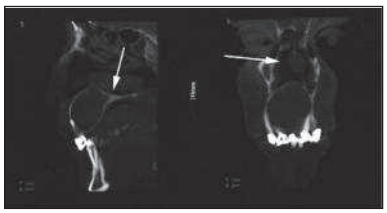

FIGURE 5: Note that the cystic enlargement through the incisive canal (A), and the nasal septum deviation to the left side (B) (Case 1).

A cone beam computed tomography (CBCT) scan revealed the large defect which was 33,6 x 31,6 x 28,8 mm in size. The buccal and palatal cortical bone plates both exhibited resorption, and the lesion extended into the nasal cavity (Figure 4). The floor of the nasal fossa was elevated and the nasal septum was deviated (Figure 5).

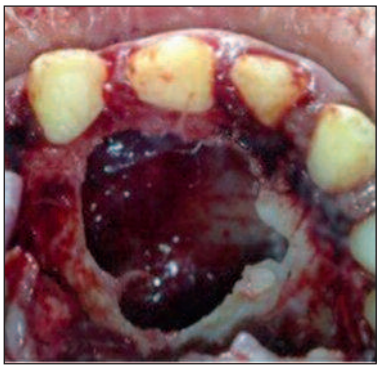

FIGURE 6: Extensive NPDC was enucleated (Case 1).

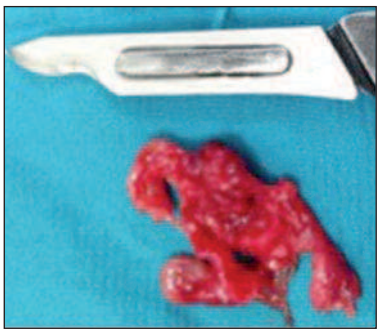

FIGURE 7: The NPDC specimen (Case 1).

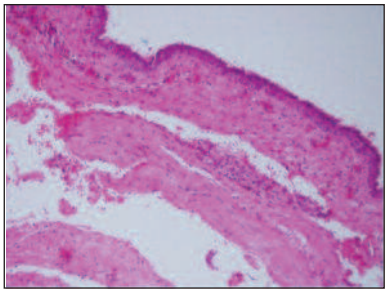

FIGURE 8: Microscopically, cyst lined by ciliated pseudostratified columnar epithelium. Mild chronic inflammation was seen on the cyst wall (Case 1).

Cystectomy was performed after confirming by CBCT. Surgically enucleated lesion adhered to the palatal flap and nasal mucosa (Figures 6, 7). Histologic examination confirmed the diagnosis of NPDC (Figure 8).

CASE 2

In the other case (Case 2), A 55 year-old female patient was referred to our hospital complaining of painless swelling involving anterior palate. The patient gave history that she had not had any systemic diseases or a story of dentoalveolar trauma.

Extraoral examination revealed no obvious facial asymmetry and intraorally the patient was examined and it was found out that the patient had a painless swelling and expansion involving anterior palate (Figure 9). Panoramic radiography revealed the round shaped radiolucent lesion just above the apexes of maxillary incisor teeth (Figure 10). The pulp vitality test applied from right canine to left first premolar and teeth were found out as vital. This finding was suggestive of a NPDC.

Cone beam computed tomography images revealed that well defined hypodense lesion, and palatal and nasal floor resorptions (Figure 11, 12). The size of the lesion was 26.0 x 46.8 x 25.8 millimeters. Nasal septum was also deviated but not as much as the case 1. Under local anaestethics, full thickness mucoperiosteal flap was raised palataly

FIGURE 9: Preoperative intraoral image of the Case 2.

FIGURE 10: NPDC is seen just above the roots of the anterior maxillary incisors on the panoramic radiography (Case 2).

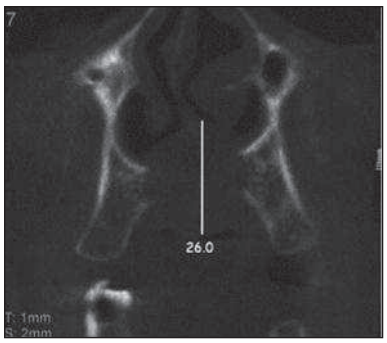

FIGURE 11: Coronal section of CBCT scan shows that radiolucent expansion at the anterior palate and nasal septum deviation to the right side (Case 2).

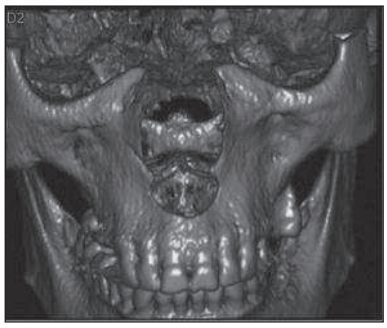

FIGURE 12: 3D reconstructed CBCT image shows the bone resorption (Case 2).

FIGURE 13: Enucleated cyst region (Case 2).

FIGURE 14: Postoperative intraoral image of the Case 2.

and the cyst was enucleated from the bony wall as well as from the palatal mucosa and exposed nasal floor (Figure 13). Postoperatively there was no pain and disturbances (Figure 14).

DISCUSSION

The nasopalatine duct cyst was first ever described by Meyer in 1914.[8] It develops from the proliferation of epithelial remnants of embryonic nasopalatine ducts present within the incisive canal and it is the most common nonodontogenic cyst in the maxillofacial region with a prevalence of 2.2%.[5,9-11] It is seen as rounded, hearth-shaped radiolucency at the midline of the anterior maxilla with a sclerotic border. The hearth-shaped radiolucency is due to the anterior nasal spine which is centrally superimposed on the bone defect. In some situations, however, from time to time it can present as unilateral radiolucency and rarely causes root resorption.[5,9,12] NPDCs can cause divergence of the roots and in our case 1, divergent roots of the maxillary incisors can be seen. Recurrence of NPDC after enucleation has been reported but it is between 0% to %11.[5,13] About 20% to 30% of cases enucleated have been reported to heal by scar tissue, and have not shown bone regeneration.[3,14] In our presentation, both cases were enucleated, and no graft or regenerative materials were used.

The diameter of NPDCs varies from 6 mm to mean 17mm, and is likely to be less than 20 mm. A size smaller than 6 millimeters is considered to be a normal anatomic shape of incisive foramen if there are no clinical signs and symptoms.[3,4,13,15,16] Both of our cases exceeded the sizes of normal anatomic variations.

Most of the NPDC cases are asymptomatic, and they are usually encountered on radiographs of routine dental examinations by chance.[3,4,9,13] In our cases, both of the patients presented the pathology as palatal and labial swellings. According to the previous case reports, swelling of a palate and/or vestibule is regarded as the most common symptom of the NPDCs.[3,4] There were no complaints of pain and this finding is compatible with NPDCs general features. If pain is involved, infection may be the cause.[2,4,5,13] Cortical bone and root resorptions were also reported. In our case 1 and 2, both palatal and buccal cortex were resorbed and floors of the nose were exposed, none of the patients showed root resorptions. There was also slight protrusion of the upper lip and widening of the nasal alar bases in case 1. One interesting sign of both of the cases was the deviation of nasal septum and to our knowledge only 2 cases have been reported in the literature.[6,7] This sign can be the cause of the difficulty of breathing.

Despite the fact that both of the patients we presented are female, generally NPDCs are seen more frequently in men.[2-5] They have a tendecy to

develop between the fourth and sixth decades of life.[4,5,9] In our presentation, one of the patients was in her second, and the other one was in her sixth decade of age. The more aggresive form was seen in younger patient but a recent study reported that there was no statistically significant difference in cyst dimensions between the patients older than or equal to 50 years of old and younger than 50 years of old.[17]

Accuracy of plain radiographs to exclude the diagnosis of odontogenic lesions adjacent to roots of teeth is troublesome. Superimposition of the non-odontogenic lesions over the apex of teeth have often led to misdiagnoses.[10,18-25] Therefore NPDCs are one of the most wrongly diagnosed entities owing to location of incisive foramen.[10,23,24] Since they are radiographically superimposed over the apices of the maxillary central incisors, these radiolucent images complicate the diagnosis and they are wrongly regarded as apical lesions or radicular cysts. Therefore the pulp vitality tests should be performed on the teeth that are radiographically evaluated and supposed to have apical odontogenic lesions. Positive results should direct the dentist's attention to the nonodontogenic entities. If the teeth are endodontically treated, differential diagnosis becomes ambiguous.[23,26,27] In such circumstances, conventional radiographs are not capable of distinguishing between odontogenic and non-odontogenic lesions.[6,28,29]

A cone beam computed tomography scan is worth mentioning at this point. Several studies reported that the CBCT has the ability to show early stages of bony lesions more than periapical and panoramic radiographs.[28,30] In early stages, odontogenic lesions like apical periodontitis or radicular cyst have their typical features and they are adjacent to the apex of the pathologic teeth. Similarly, if it is the case of an initial stage NPDC, widening of the nasopalatin canal beyond the mean anatomical sizes will be suggestive of an non-odontogenic entities.[10] But the more lesions enlarge, the closer they get to one another's original growing site (NPDC is to the apex of tooth, and radicular cyst is to the incisive foramen). At this situation CBCT scan fails to show actual site of the pathoses. Faitaroni et al. emphasized this point in their case series and stated that CBCT imaging did not provide the information of differential diagnosis between NPDCs and odontogenic lesions like periapical cyst or dental granuloma. But they also reported that CBCT scan is still valuable for plannig the surgery.[23] In our cases, both of the lesions are so extensive that differential diagnosis based on only CBCT imaging, was impossible.

One another way to assess the probable diagnosis of NPDC is magnetic resonance imaging (MRI). It is an non-invasive scanning method and it provides excellent tissue contrast.[31] In a study comparing the MRI features of different cysts of oral and maxillofacial region. Hisatomi et al., found out that NPDCs have high signal intensity on T1-weighted images (T1WI) whereas most cystic lesions in the maxillofacial region have low to intermediate signal intensity on T1WI.[32,33] According to the authors, it is possible to distinguish NPDCs from radicular cysts and odontogenic keratocysts.[33] But we think that further studies with large numbers of cases are needed to statistically confirm these findings.

Histopathologic features of NPDC is variable and the most common epithelial lining is stratified squamous epithelium. Pseudostratified columnar epithelium, simple columnar or cuboidal epithelium and even ciliated columnar epithelium can also be seen at the same time.[34] However, distinguishing NPDCs from radicular cysts can be troublesome especially when the NPDC is composed of only stratified squamous epithelium. Therefore subepithelial stroma should also be evaluated, since NPDCs generally have nerve bundles, muscular walled vessels and lymphatic infiltration in cystic wall. These findings are regarded as characteristic diagnostic feature of NPDCs.[10] In our study, cyst lined by ciliated pseudostratified columnar epithelium. Mild chronic inflammation was seen on the cyst wall.

Consequently, nasopalatine duct cysts are the most common nonodontogenic cyst of the oral cavity seen in the general population. NPDCs must be distinguished from other maxillary ante-

rior radiolucencies. Vitality testing of teeth adjacent to or involved with a cyst-like lesion is mandatory and the final diagnosis could only be performed after histological analysis. It is important that practitioners are aware of the features of the NPDC.

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