

SURGICAL MANAGEMENT OF NASOPALATINE DUCT CYST: case report

Tratamento cirúrgico do cisto nasopalatino: relato de caso

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Abstract

OBJECTIVE: To present a case and discuss the clinicopathological characteristics of nasopalatine duct cyst and discuss the etiology, diagnosis, treatment and prognosis, with a review of the literature. **DISCUSSION**: Nasopalatine duct cyst occurs in approximately 1% of the population. Presentation may be asymptomatic or include swelling, pain, and drainage from the hard palate. Surgical treatment was carried out under local anesthesia and comprised the dissection and removal of the cyst adopting a usually palatine approach.

Keywords: Non-odontogenic cysts. Maxillae. Nasopalatine duct cyst.

Resumo

OBJETIVO: Apresentar um caso e discutir as características clínico-patológicas do cisto de ducto nasopalatino, discutinfo a etiologia, diagnóstico, tratamento e prognóstico, revisando a literatura. **DISCUSSÃO**: Os cistos naso-palatinos ocorrem em aproximadamente 1% da população. A apresentação pode ser assintomática ou incluir edema, dor e drenagem purulenta do palato duro. O tratamento cirúrgico foi sob anestesia local e constituiu de dissecção e remoção do cisto, via acesso palatino.

Palavras-chave: Cistos não odontogênicos. Maxila. Cisto de ducto nasopalatino.

INTRODUCTION

The nasopalatine duct communicates the nasal cavity with the anterior region of the upper maxilla. It is located on the midline and palatine to the upper maxilla, above the retroincisor palatal papilla. During fetal development the duct gradually narrows until one or two central clefts are finally formed on the midline of the upper maxilla. The nasopalatine neurovascular bundle is located within the duct, and emerges from its intrabony trajectory through the nasopalatine foramen. There can be as many as six different foramina, though there are usually only two, with independent neurovascular bundles (right and left). The vascular and neuronal elements can emerge separately; in this sense, foramina containing exclusively vascular elements are known as Scarpa's foramina (1).

The nasopalatine duct cyst (NPDC) was first described in 1914 by Meyer (2). It is also known by other names such as anterior middle cyst, maxillary midline cyst, anterior middle palatine cyst, and incisor duct cyst (3). At present, according to the classification of the World Health Organization (WHO), these lesions are regarded as developmental, epithelial and non-odontogenic cysts of the maxillae, along with nasolabial cysts (4).

NPDCs are the most common nonodontogenic cysts of the mouth, representing up to 1% of all maxillary cysts (5). These lesions are almost three times frequent in males than in females (6). The maximum prevalence is between 40 and 60 years of age. Due to a lack of representative studies, it is not fully clear whether NPDCs are more common in caucasians, blacks or asians (7) Often mistaken for an enlarged nasopalatine duct, NPDCs are of uncertain origin. The spontaneous proliferation theory appears to be the most likely explanation (a number of studies have reported cystic degeneration in the incisor duct and on the midline of the palate in human fetuses) (8).

NPDCs are normally asymptomatic, constituting casual radiological findings, though sometimes (in 17% of cases) patients report pain due to the compression of structures adjacent to the cyst, particularly when the latter becomes overinfected, or in patients who wear dentures that compress the zone. The more caudal the location of the cyst, the sooner symptoms appears. These normally manifest as an inflammatory process (46% of cases) that rarely produces facial

asymmetry, since growth or expansion is intraoral (palatine). The more advanced cases are able to cause pain and itching (9).

This developmental cyst forms in the nasopalatine duct, and appears on x-ray above the apices of the maxillary incisors. The cyst may overlap the roots of the teeth. It is usually asymptomatic and discovered on routine dental films where it appears as an oval or heart-shaped radiolucent lesion. Rarely this cyst will expand the overlying mucosa. The nasopalatine duct cyst rarely becomes large enough to destroy bone, therefore, no surgical treatment is necessary for an asymptomatic small cyst. If the cyst shows signs of infection or shows progressive enlargement, then surgical intervention may be warranted. Treatment in majority of cases involves complete surgical removal as soon as possible after diagnosis (6). A relapse rate of up to 30% has been reported (10).

CASE REPORT

A 35-year-old woman reported to our department with an asymptomatic, nodular swelling located on the palate between the maxillary right and left central incisors since 1 year. She had noticed that the swelling was growing slowly. The patient's medical history was noncontributory. Clinical examination revealed a round swelling located on the nasopalatal region between the maxillary right and left central incisors (Figure 1) which was almost 1 cm in diameter, and fluctuant on palpation.



Figure 1 - Round swelling, almost 1 cm in diameter located in the nasopalatine region between the upper central incisors

Pulp vitality testing showed that 11 and 21 were vital. Periapical radiograph of the area showed a heart shaped radiolucency with a radiopaque margin located between the roots of 11 and 21 (Figure 2), and Occlusal radiograph of the area showed a rounded radiolucency with a radiopaque margin located between the roots of 11 and 21 (Figure 3).



Figure 2 - Homogeneous well delimited, heart-shaped radiolucency, without affecting the roots of the two permanent upper central incisors



Figure 3 - Well defined rounded radiolucency in the maxillary midline



Figure 4 - Crevicular incision



Figure 5 - Flap is elevated exposing the root surfaces of the upper central incisors

A clinical diagnosis of a nasopalatine duct cyst was made. Therefore, surgical excision of the lesion was proposed to the patient. The surgical intervention was carried out under local anesthesia by infraorbital block injection and infiltration with 2% lidocaine containing 1:100,000 epinephrine. Crevicular incision is given (Figure 4) and the palatal flap elevated (Figure 5).

The cystic lining and contents were removed and the lesion was completely enucleated, exposing areas of the root surfaces of the 11 and 21 (Figure 6). The root surfaces appeared normal without any signs of resorption. Interrupted sutures



Figure 6 - Exposure of the root surfaces and bone after enucleation



Figure 7 - Sutures at place



Figure 8 - Sutures at place

(3-O, silk) were placed (Figure 7 and 8) and a periodontal pack was given. The patient was prescribed antibiotic and analgesic. Sutures and periodontal dressing were removed after one week. The specimens (Figure 9) were sent for microscopic examination Pathological findings revealed



Figure 9 - Figure showing the cystic contents



Figure 10 - Squamous cell epithelium and ciliary cylindrical epithelium (HE, 400 x)

squamous and respiratory cell types, infiltrated by inflammatory cells (Figure 10). The histological diagnosis was naso palatine duct cyst.

The histologic, clinical and radiologic findings were compatible with nasopalatine duct cyst. The postoperative course was uncomplicated and there was no lesion recurrence up to one year follow-up.

DISCUSSION

Nasopalatine duct cysts (NPDCs) are almost three times more common in males than in

females (2, 6, 11). These lesions mainly manifest between the fourth and sixth decades of life (4-6, 10, 12, 13) though there have been reports of NPDCs in pediatric patients up to 8 years of age (5, 9).

The etiology of these lesions is not clear; in addition to the hypothesis of spontaneous proliferation from embryonic tissue remains, other possible etiologies have been proposed - including prior trauma, poorly fitting dentures, the existence of local infection, or the influence of genetic and racial factors (6,9). There have even been exceptional reports such as the casual diagnosis of NPDC nine months after rapid surgical palatal expansion (14) or NPDC associated to the presence of two bilateral mesiodens (15). In our patient the presentation was idiopathic, together with a history of chronic infection of the permanent upper central incisor, secondary to trauma. However, the consideration is an unknown etiology or spontaneous proliferation to be the most plausible explanation, based on studies reporting cystic degeneration phenomena in the incisor duct and on the midline of the palate in human fetuses, in which the above mentioned circumstances are unable to have occurred (4).

Most of the cysts are asymptomatic and constitute casual findings. Any clinical manifestations that may appear are attributable to inflammation, in which case pain, itching, ulceration, local infection and/or fistulization are observed (2, 7). Palatal or superficial locations are more common than nasal or deep-lying locations. Radiologically, the lesions manifest as a well delimited radiotransparency measuring 1-2 cm in diameter, and located on or close to the midline of the upper maxilla. The X-ray image is predominantly rounded or ovoid, with a lesser prevalence of heart-shaped images (8). The latter image is explained by the presence of the anterior nasal spine. Asymptomatic radiotransparencies measuring less than 6 mm in size are regarded as enlarged incisor ducts of a non-pathological nature(2).

A thorough differential diagnosis must be established in order to avoid unnecessary treatments such as endodontic procedures in vital permanent upper central incisors (1, 6). A correct tentative diagnosis should be based on positive vitality testing and negative percussion findings of the permanent upper central incisors, provided these teeth do not have pulp or periodontal problems (6). In addition to panoramic X-rays, other complementary techniques are advised, such as periapical and occlusal X-rays and computed tomography. The latter technique guarantees in establishing a tentative diagnosis, since it generates great detail of the structures (normally intact) adjacent to the lesion. Computed tomography easily visualizes the radiotransparency on the midline, with well defined sclerotic margins, and informs of the exact location of the lesion. In addition, it facilitates planning of the best surgical approach (8, 16).

The differential diagnosis may include an enlarged nasopalatine duct (less than 6 mm in diameter), central giant cell granuloma, a radicular cyst associated to the upper central incisors, follicular cyst associated with mesiodens, primordial cyst, nasoalveolar cyst, osteitis with palatal fistulization, and bucconasal and/or buccosinusal communication (1). Other diagnostic techniques can be used to radiologically assess lesions of this kind, such as multimodal tomography. which employs crossed and sectional tomographic acquisitions in the sagittal plane to yield three dimensional images (17). Magnetic resonance imaging (MRI) may also prove useful in establishing the diagnosis, and particularly contrast the interior of the NPDC with a high signal intensity. Specific axial T1-weighted imaging reflects the presence of fluid, viscous and protein material within the cyst and abundant keratin at superficial level. Thus, MRI is reliable in diagnosing NPDCs, discarding radicular cysts or any other cysts of odontogenic origin (3, 18, 19).

The treatment of choice is surgical excision of the cyst, although some authors propose marsupialization of large NPDCs (2, 11). The nasopalatine neurovascular bundle is a delicate and highly vascularized structure giving rise to profuse bleeding if inadvertently sectioned during surgery. Electrocoagulation is required in such cases. Paresthesia of the anterior palatal zone is a rare complication found in 10% of the cases on removing nerve endings of the nasopalatine nerve along with the membrane of the cyst (2).

The histological study of NPDCs normally only reveals squamous cell epithelium (in 40% of cases), though in some cases the latter is combined with other types of epithelium such as ciliary cylindrical cells (11). The cyst lumen usually contains an abundant inflammatory infiltrate with a great variety of polymorphonuclear leukocytes, secondary to chronic inflammation (11).

Cystic lesions of the maxillae require exhaustive study and precise treatment and histological diagnosis, since some of them may be aggressive and incorrect diagnosis and treatment can give rise to recurrences (12).

CONCLUSION

Nasopalatine duct cysts (NPDCs) are of uncertain origin, and show a peak incidence between the fourth and sixth decades of life. No racial predilections have been established. In the absence of over infection, NPDCs are asymptomatic. The tentative diagnosis is based on the clinical history, the clinical exploration, and complementary tests (particularly computed tomography). Where present, irritative factors should be eliminated, and early surgical removal is advised in order to avoid possible recurrence. The definitive diagnosis is established by histological study of the lesion. Following resection, relapse is unlikely, though a postoperative follow-up of at least one year is indicated in all cases.

CONFLICT OF INTEREST STATEMENT

The authors declared no conflict of interest in the present manuscript.

INFORMED CONSENT STATEMENT

The patients signed an informed consent, kept in the records, in the archives of the Navodaya Dental College.

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