Nasopalatine duct cyst: Report of 22 cases and review of the literature

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Abstract

Objectives: Nasopalatine duct cysts (NPDCs) are the most common developmental, epithelial and non-odontogenic cysts of the maxillae. The present study describes the clinicopathological characteristics of 22 NPDCs and discusses their etiology, incidence, treatment and prognosis, with a review of the literature on the subject.

Study design: A retrospective observational study was made comprising a period of 36 years (1970-2006), and yielding a series of 22 patients with histopathological confirmation of NPDC. Surgical treatment was carried out under local anesthesia and comprised the dissection and removal of the cyst adopting a usually palatine approach, with the preparation of an enveloping flap from 1.4 to 2.4.

Results: No statistically significant correlation was observed between the size of the lesion and patient age, although the size of the cyst differed according to patient gender, with a mean NPDC diameter of 16 mm in males and 12 mm in females. In no case did we observe root reabsorption or loss of vitality of the upper incisors following surgery. The X-ray image was rounded in 15 cases and heart-shaped in the remaining 7 cases. In the majority of cases panoramic X-rays and periapical and occlusal X-rays sufficed to identify the lesion, though computed tomography was used in cases of doubt.

Conclusions: The etiology of NPDC is unclear. Simple surgical resection is recommended, followed by clinical and radiological control to ensure correct resolution of the case.

Key words: *Nasopalatine foramen, nasopalatine duct cyst, electroscalpel, squamous cell epithelium, ciliary cylindrical epithelium.*

Introduction

The nasopalatine duct communicates the cavity nasal 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 6 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). These lesions, also known by other names such as anterior middle cyst, maxillary midline cyst, anterior middle palatine cyst, and incisor duct cyst, were regarded as fissural cysts in the past (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 non-odontogenic cysts of the oral cavity, representing up to 1% of all maxillary cysts (5). These lesions are more almost three times more frequent in males than in females (6). The maximum incidence 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, negroes 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).

Histologically, the type of cystic epithelium varies according to the location involved (palatine, nasal or intermediate). A squamous cell epithelium is almost always observed, though a ciliary respiratory-type epithelium can be seen when the lesion is located higher up or nasally (7).

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).

NPDCs appear as a well delimited, rounded or heart-shaped radiotransparency circumscribed to the upper interincisal midline. The differential diagnosis is established with the following conditions: an enlarged nasopalatine duct, central giant cell granuloma, a central incisor root cyst or other maxillary cysts, osteitis fistulizing in the palatine direction, or a bucconasal and/or buccosinusal communication. Treatment in all cases involves complete surgical removal as soon as possible after diagnosis (6). A relapse rate of up to 30% has been reported (10).

The present study describes the clinicopathological characteristics of 22 NPDCs and discusses their possible etiology, incidence, treatment and prognosis, with a comparative review of the literature on the subject.

Patients and Method

A retrospective observational study was made comprising a period of 36 years (1970-2006), and selecting a series of 22 patients with histopathological confirmation of NPDC. The study included patients treated in a number of centers: Vall d'Hebron General Hospital, Granollers General Hospital, the Teknon Medical Center, and in the context of the Master of Oral surgery and Implantology of the University of Barcelona (Spain).

The following data were compiled from the case histories of all the patients: age, gender, race, disease antecedents, toxic habits, possible etiology, size and shape of the lesion, symptoms, surgical treatment provided, histology and course. Panoramic X-rays and computed tomography (CT) scans were obtained in all cases.

Surgical treatment was carried out under local anesthesia and comprised the dissection and removal of the cyst adopting a usually palatine approach. An enveloping flap from 1.4 to 2.4 was generally prepared, though in some cases a vestibular approach was used – in which case a triangular or trapezoidal flap was raised. The electroscalpel was used in most cases, to avoid important bleeding of the nasopalatine neurovascular bundle. Lastly, in 11 patients a palatine plaque was prepared preoperatively, using Visco-Gel® tissue conditioner (Dentsply DeTrey, Konstanz, Germany). The plaque was placed in the immediate postoperative period, to avoid the formation of an important submucosal hematoma and surgical wound dehiscence.

The samples obtained (excision biopsies) were sent to the pathology laboratory in 10% formalin solution for histological study after staining with hematoxylin-eosin. In the present study only those cases with a histologically confirmed diagnosis of NPDC were included.

The descriptive statistical study was made using the SPSS version 12.0 statistical package (SPSS Inc., Chicago, USA; License of the University of Barcelona).

Results

The study comprised a total of 22 cases of NPDC diagnosed in 22 Caucasian patients (12 males and 10 females) with an age range of 16-73 years (mean 46 years) and a peak incidence between 50-60 years of age.

Only 7 patients presented disease antecedents of interest: arterial hypertension (n=4), chronic obstructive pulmonary disease (n=2), gastroduodenal ulcer (n=3), hypercholesterolemia (n=2), and a single patient with a history of three strokes. Regarding toxic habits, 6 patients were smokers of approximately 20 cigarettes/day, while 9 smoked between 4 and 10 cigarettes/day. Eight patients (of which 7 were moreover smokers) reported moderate alcohol consumption (1-2 glasses of wine or beer daily). Only 6 patients presented no toxic habits.

As regards the possible etiology of the lesions, four patients showed imbalance of their removable complete upper dentures. In 8 patients the cysts were associated to a history of chronic permanent upper central incisor infection secondary to trauma, a periodontal problem, or failed

endodontic treatment. In two patients the nasopalatine duct could have become infected via the nasal route, since they presented chronic bacterial rhinitis without symptoms at the time of diagnosis. The remaining 8 patients in the series presented no pathology associated to the lesion.

The vitality of the teeth adjacent to the lesion (permanent upper central incisors) was almost always preserved (14 cases, 63.63%). However, in 8 cases the teeth had been subjected to endodontic treatment, or the pulp tissue was necrotic. Percussion was generally negative (18 cases, 81.81%), with the exception of those adjacent teeth which had suffered some periodontal or periapical problem (4 cases, 18.18%). The mean radiological diameter of the lesion was 14 mm (12 mm in females and 16 mm in males). The X-ray image of the lesion was heart-shaped in 10 cases (Figures 1 and 2), and rounded or oval in 12 cases (Figure 3).

Most of the patients were asymptomatic (14 cases), while four showed local inflammation, and the remaining four reported pain and ulceration produced by dentures.

The position of the NPDCs was mostly superficial or palatine (15 cases), while in the remaining 7 cases the lesions were located deep or in the nasal region (Figure 4). A computed tomography scan was requested of all the patients to precisely establish the position of the NPDCs (Figure 5).

In all cases, treatment consisted of complete removal of the lesion using the cold scalpel, with careful dissection of the nasopalatine neurovascular bundle. Lastly, the electroscalpel was used (Figure 6) for correct bleeding control of the surgical bed. In 21 cases (95.45%) a palatine approach was used, with vestibular access in a single case (4.54%). There were no peroperative complications. However, the 11 patients operated upon via the palatine approach with no palatine plaque placement immediately after surgery reported discomfort due to important swelling, pain and tenderness in the anterior region of the hard palate (submucosal hematoma) during the first two postoperative weeks.

From the histological perspective, the lesions presented exclusively squamous cell epithelium in 15 cases (68.18%), while the remaining 7 cases (31.82%) moreover also showed ciliary cylindrical epithelium (Figure 7). The presence of melanin was observed in one case.

The mean duration of follow-up was one year, until correct ossification of the surgical zone was confirmed by X-ray study. Following surgical exeresis, 20 lesions healed completely after the first operation, while the remaining two cases suffered relapse after three years of follow-up, i.e., the relapse rate in our series was 9.09%. This situation required a second intervention (removal of the NPDC with ligation of the nasopalatine neurovascular bundle), after which complete healing was confirmed within two years.

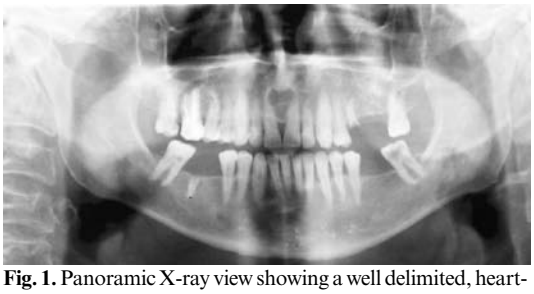

Fig. 1. Panoramic X-ray view showing a well delimited, heart-shaped radiotransparency on the upper maxillary midline.

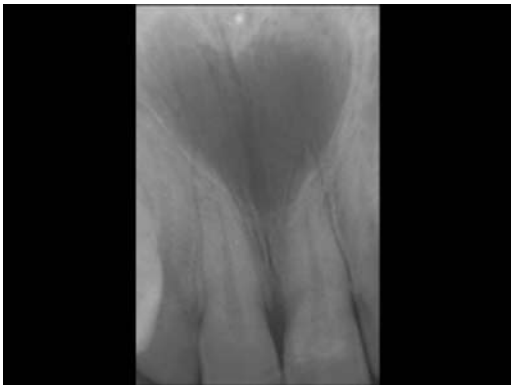

Fig. 2. Periapical X-ray view showing a homogeneous well delimited, heart-shaped radiotransparency, without affecting the roots of the two permanent upper central incisors.

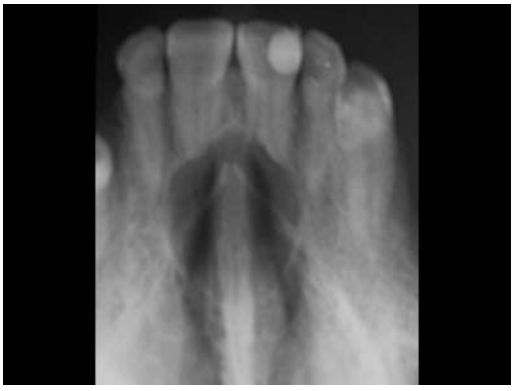

Fig. 3. Occlusal X-ray view showing a well delimited, rounded radiotransparency on the upper maxillary midline.

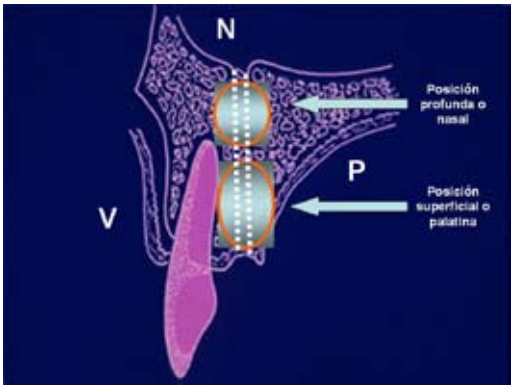

Fig. 4. Schematic representation showing the two most frequent locations of nasopalatine duct cysts.

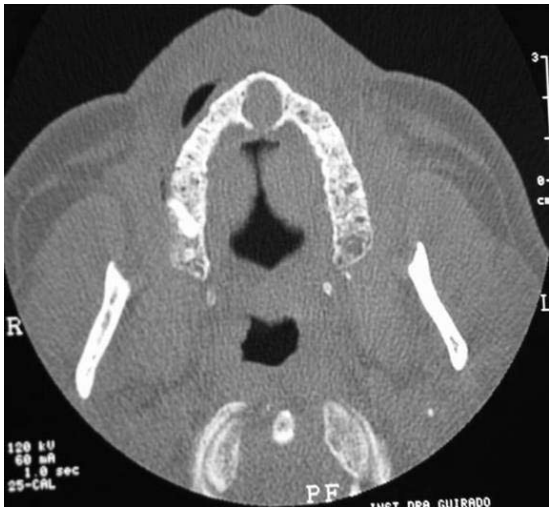

Fig. 5. Axial computed tomography scan showing a rounded radiotransparency on the upper maxillary midline. The lesion is located in a deep or nasal position, and apparently perforates the palatal cortical layer.

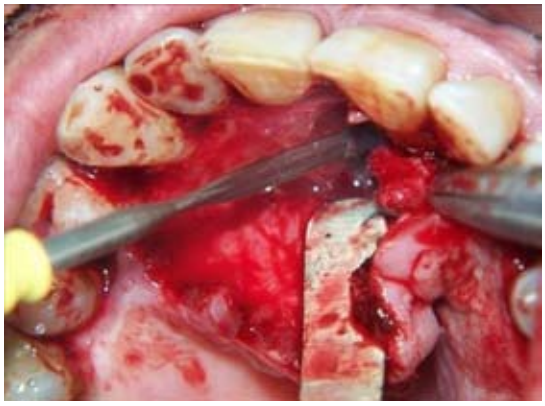

Fig. 6. Peroperative view showing use of the electroscalpel during removal of the cyst, grasped with mosquito forceps and a periostotome.

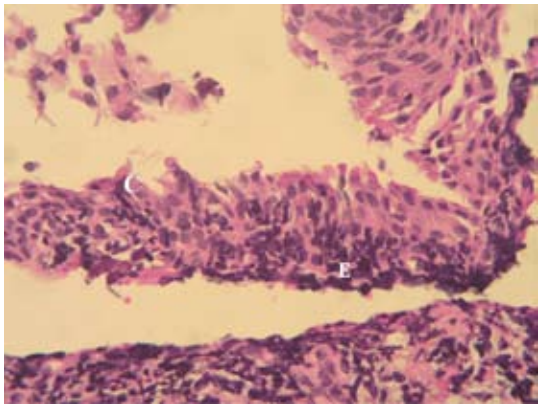

Fig. 7. Histological section of a nasopalatine duct cyst following hematoxylin-eosin staining (x 400). Squamous cell epithelium (E) and ciliary cylindrical epithelium is observed (C).

Discussion

Nasopalatine duct cysts (NPDCs) are almost three times more common in males than in females (2, 6, 11), and show a predilection for Caucasian individuals (7). However, in our study, NPDCs affected both males and females equally, with only a slight predominance among the former. This could be because women typically seek dental help sooner than men (7). As regards race, the findings are controversial, since some studies report the same incidence in both negroes and whites (2, 4, 12), and any difference between them may be attributable to lesser economical resources among the former (7). In our series, and since all patients were Caucasian, no conclusions regarding the racial predilection of NPDCs can be drawn. 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 maximum incidence in our series was between the fifth and sixth decades of life, in coincidence with the observations of most studies in the literature (4-6, 10, 12, 13).

The etiology underlying these lesions is not clear, though 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 patients the most frequent presentations were idiopathic, together with a history of chronic infection of the permanent upper central incisor, secondary to trauma. However, we consider 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 (14 of the 22 patients with NPDC in our study), 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 (15 of the 22 patients with NPDC in our study). 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 (12 of our 22 cases), with a lesser prevalence of heart-shaped images (8)(10 of our 22 patients). The latter image is explained by the presence of the anterior nasal spine. Asymptomatic radiotransparencies measuring un-

der 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 dental vitality testing and negative percussion findings of the permanent upper central incisors, provided these teeth do not have pulp or periodontal problems (6). Radiological exploration is essential for diagnosing NPDCs, and in addition to panoramic X-rays, other complementary techniques are advised, such as periapical and occlusal X-rays and computed tomography. The latter technique (requested in all 22 cases of our series) offers maximum 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 (normally palatine, as in 21 of our 22 cases)(8, 16).

The mean radiographic size of the lesion tends to vary according to patient gender (4). In our study, the inter-gender difference in mean diameter was 4 mm (12 mm in females and 16 mm in males).

The differential diagnosis must be established with other conditions such as an enlarged nasopalatine duct (less than 6 mm in diameter), central giant cell granuloma, a root cyst associated to the upper central incisors, a supernumerary tooth follicular cyst (normally 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 in addition to exposing the patient to lesser radiation doses 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 highly reliable in diagnosing NPDCs, discarding root cysts or any other cysts of odontogenic origin (3, 18, 19).

The treatment of choice is surgical exeresis 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. We therefore consider that the electroscalpel offers adequate safety in such surgical procedures, and used it in all 22 NPDCs in our series. 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). In our study we recorded two cases of palatal paresthesia after repeat surgery of the two cases that relapsed.

The histological study of NPDCs normally only reveals squamous cell epithelium (in 40% of cases; 68.18% in our study), though in some cases the latter is combined with other types of epithelium such as ciliary cylindrical cells (31.82% in our study, in deep-lying or nasal cysts) (11). In addition, in our study, one NPDC was seen to contain melanin remains. The cyst lumen usually contains an abundant inflammatory infiltrate with a great variety of polymorphonuclear leukocytes, secondary to chronic inflammation (11).

On one hand, we consider that odontogenic keratocysts are not specific cysts; rather, they correspond to a histological concept characterizing any maxillary epithelial cyst with keratin coating of one degree or other. This hyperkeratotic feature can also be found in NPDCs, and is associated with a poorer prognosis, since the relapse rate is higher (close to 30%)(10). Moreover, the possibility of malignization is greater if early resection is not performed. On the other hand, there have been reports of a cartilaginous component in the NPDC wall, which could be attributable to fibrous connective tissue metaplasia in response to chronic irritation (20).

Squamous cell carcinomas originating in maxillary bone are mainly due to the metaplasia experienced by the epithelial wall of a cyst or of the epithelial remains that participated in odontogenesis. Therefore, there are cases in which NPDC gives rise to squamous cell carcinoma in the anterior zone of the upper maxilla (21). The treatment in these cases is of course less conservative, with greater morbidity and a poorer prognosis in advanced cases, compared with those lesions identified in early stages. In effect, radical en bloc resection of the anterior region of the upper maxilla is required in such situations (10). This explains the need for early removal of the NPDC, with the purpose of minimizing the risks and pre-, per- and postoperative complications (9).

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

Conclusions

Nasopalatine duct cysts (NPDCs) are of uncertain origin, and show a peak incidence between the fifth and sixth decades of life. In our series no particular male predilection was recorded, though the literature reports a clear male predominance. No racial predilections have been established. In the absence of overinfection, 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 malignization. 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.

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