

PREVALENCE, DISTRIBUTION, AND DIFFERENTIAL DIAGNOSIS OF NASOPALATINE DUCT CYSTS

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SUMMARY

Introduction. Nasopalatine duct cysts (NPDCs) are the most common developmental epithelial non-odontogenic cysts of the maxillae. Their origin, however, is still a source of considerable debate.

Aims. The aim of this investigation is to describe and discuss the etiology, differential diagnosis, clinic-pathological characteristics as well as to report the relative frequency and distribution of nasopalatine duct cysts in population (NPDCs) with a literature's review on the topic.

Methods. The retrospective study was carried out using 36 clinical cases, with histopatological confirmation for NPDC, radiographs and oral photographs. Data included age and gender of the patient, radiographic findings, etiological factors, treatment, and prognosis of NPDC. Few surgical consideration are discussed.

Results. The study results report a clear male predilection with a 3:1 ratio. No statistically significant correlation was observed between the size of the lesion and patient's gender. Lesions were usually asymptomatic. All cysts were located in the anterior maxillary midline region. Panoramic X-rays and computed tomography was used to identify the lesion. Surgical treatment was performed under local anesthesia including the dissection and removal of the cyst, adopting a usually palatine approach, with an enveloping flap from 1.4 to 2.4.

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

Key words: jaw cyst, non-odontogenic cyst, nasopalatine duct cyst.

Introduction

A cyst is defined as a pathological cavity lined by a membrane and may contain liquid or semisolid material (3).

Cysts in maxillar, mandibular and perioral regions vary greatly in histogenesis, incidence, behavior and treatment.

Authors have categorized cysts in odontogenic, non-odontogenic cysts, and pseudocysts. Pseudocysts differ from a real cyst because of the absence of epithelial membrane.

The nasopalatine duct cyst (NPDC) is an in-

traosseous developmental cyst of the midline of the anterior palate.

Studies have reported that the Nasopalatine duct cysts (NPDCs) are the most common developmental, epithelial, and non-odontogenic cysts of the oral cavity (1, 2), representing up to 1% of all maxillary cysts (1, 5).

The maximum incidence is between 40 and 60 years of age and these lesions are more frequent in males than in females, with a 3 : 1 (1) ratio. Due to a lack of representative studies, it is not fully clear whether NPDCs are more common in Caucasians, Negroes or Asians (1, 6); these lesions are uncommon in children, especially in Caucasian children (2).

This lesion (NPDC) was first described by Meyer in 1914 (as a paranasal sinus) and is considered to be the most common non-odontogenic cyst, accounting for approximately half of those reported (7-10).

In the past, these lesions, also known as “anterior middle cyst”, “anterior middle palatine cyst”, and “incisor duct cyst”, were regarded as fissural cysts (1, 11). Nowadays according to the classification of the World Health Organization (WHO, Tab. 1), these lesions are regarded as non-odontogenic cysts of the maxillae, along with maxillary midline cyst and nasolabial cyst (4).

They often happen to be in relationship to nasopalatine canal, which connects the nasal cavity with the anterior region of the upper maxilla (7, 10, 12).

NPDCs’ origin is uncertain, many authors suggest a spontaneous proliferation theory (1).

Previously they were considered as fissural cysts, originated from the epithelium trapped during the fusion of embryological process, but this concept has been discarded.

Now many authors suggest that NPDCs develop from the epithelial remnants of the oro-nasal ducts within the incisive canals (2, 6, 7, 13, 14).

Many etiologic factors have been proposed: local trauma during mastication or ill fitting dentures, bacterial infection, and spontaneous proliferation. Blockage of glandular ducts as well as racial or genetic factors are other possible causes mentioned in the literature (7).

Histologically, the type of cystic epithelium varies according to the location involved (palatine, nasal

or intermediate). A squamous cell epithelium is often observed, though a ciliary respiratory type epithelium can be seen when lesion is located higher or nasally (1) (Figs. 8-10).

Lesions are normally asymptomatic (7), constituting casual radiological findings.

Usually early symptoms could appear for the caudal location of the cyst. Inflammatory process, (46% of cases) rarely produces facial asymmetry, because the growth or expansion is intraorally (palatine). Pain and itching in this type of lesion can also occur in advanced cases (1).

In rare cases, (in 17% of cases) patients report pain for compression of structures adjacent to the cyst, when becomes an over-infection (1).

The most commonly reported clinical symptom is swelling on the anterior part of the palate (7, 15).

In rare cases patients have complained a burning sensation in the anterior part of the maxilla that occasionally radiates into the bridge of the nose and the orbits (7, 15).

NPDCs appear as a well delimited, rounded or heart-shaped radiotransparency circumscribed to the upper interincisal midline (Fig. 3). The differential diagnosis is established with the following conditions: an enlarged nasopalatine duct, a central giant granuloma, a central incisor root cyst, an osteitis fistulizing in the palatine direction, or a bucconasal and or buccosinusual communication. Treatment in all cases involves complete surgical removal as soon as possible after diagnosis. A relapse rate of up to 30% has been reported (1). The purpose of this article is to consolidate and organize available information regarding NPDCs with a review of the literature.

Table 1 - Classification of the World Health Organization (WHO) 1998.

| Epithelial cysts | | |
|---|--|--|
| Developmental cysts | | Inflammatory cysts |
| Odontogenic cysts | Non Odontogenic cysts | |
| <ul style="list-style-type: none"> - Newborn gingival cyst - Odontogenic cheratocyst - Dentigera or follicular cyst - Eruptional cyst - Lateral periodontal - Gingival cyst of adult - Scialo-odontogenic cyst | <ul style="list-style-type: none"> - Nasopalatine duct cyst - Naso-alveolar and naso-labial cyst | <ul style="list-style-type: none"> • Radicular cyst <ul style="list-style-type: none"> - Apical - Lateral - Residual • Periodontal cyst <ul style="list-style-type: none"> - Inflammatoty collateral - Infected vestibular mandibular |

Materials and methods

A retrospective observational study was made in a ten-year period (from 1998 to 2008) selecting a series of 36 out of 2098 patients evaluated for surgery treatments of cystic lesion, with histopathological confirmation of NPDC.

The study included patients treated in the Oral Surgery department of "S. Giovanni Calibita" hospital - Fatebenefratelli, IsolaTiberina (University of Rome Tor Vergata).

The following data of the case histories of all the patients were compiled: age, gender, possible etiology, shape of the lesion, symptoms, surgical treatments, radiographic appearances, histology and course.

Panoramic X-rays were obtained in all cases and computed tomography (CT) scans have been requested in some cases (Figs. 3-6).

Surgical treatment was carried out under local anesthesia and comprised the dissection and removal of the cyst usually adopting a palatine approach. An enveloping flap from 1.4 to 2.4 was generally prepared (Figs. 1, 2).

Excision Biopsies submitted to the Laboratory of Oral Pathology for histological study ("S. Giovanni Calibita" hospital - Fatebenefratelli, Isola Tiberina): the samples obtained (excision biopsies) were sent to the pathology laboratory in 10% formalin solution for histological studies after staining with



Figure 1
Intraoral photograph showing raised mucoperiosteal flap exposing the cyst.

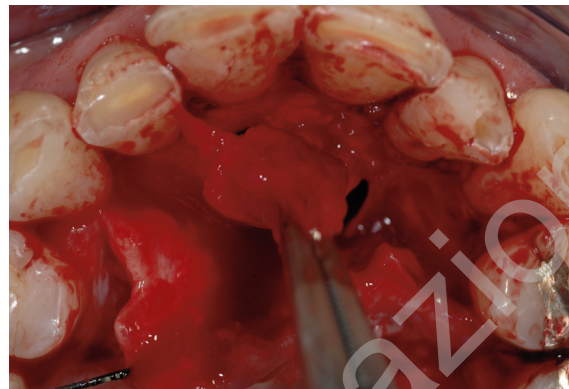


Figure 2
Intraoral photograph: exeresis of the cyst.

hematoxylin-eosin and recorded as nasopalatine duct cysts (2).

In the histological evaluation, the cyst epithelium type and features of the cystic wall were reviewed (Figs. 8-10).

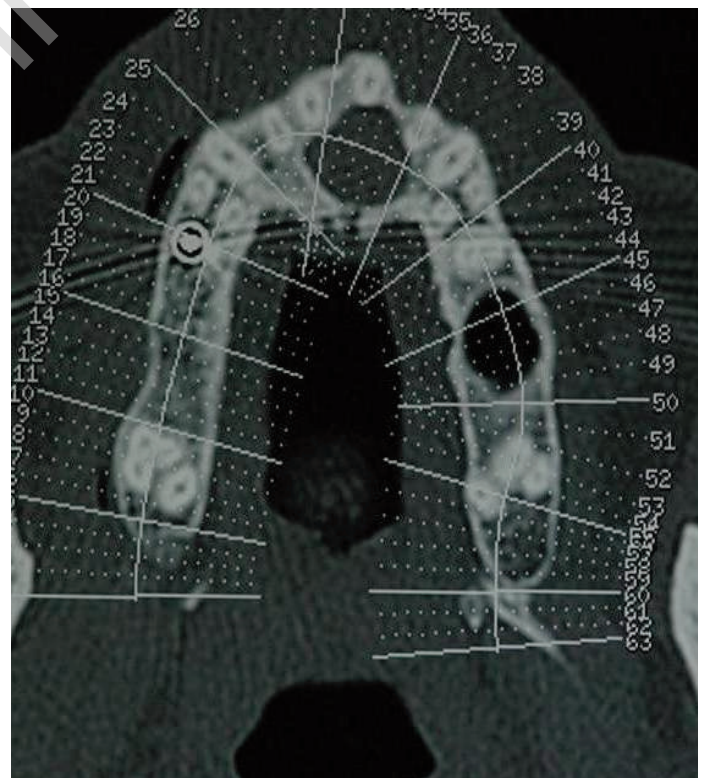


Figure 3
Axial computed tomography scan showing a rounded radiolucency on the upper maxillary midline.

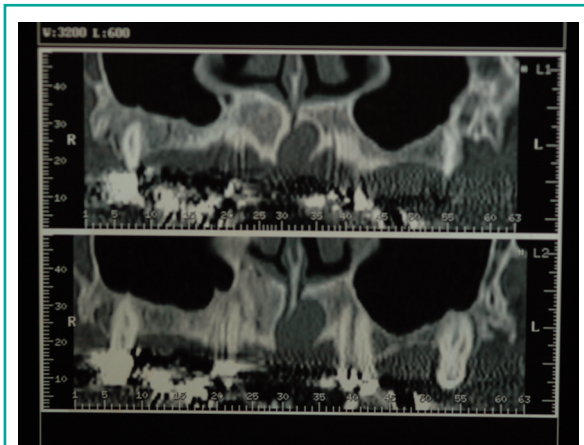


Figure 4
Computed tomography: image shows a communication between the nose and the nasopalatine duct cyst.

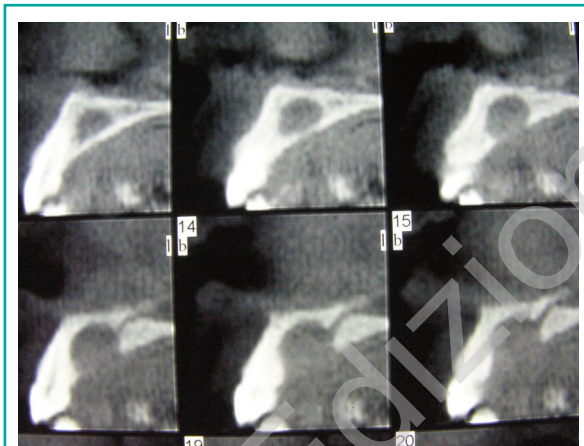


Figure 5
Computed tomography shows a well delimited, rounded radiotransparency.

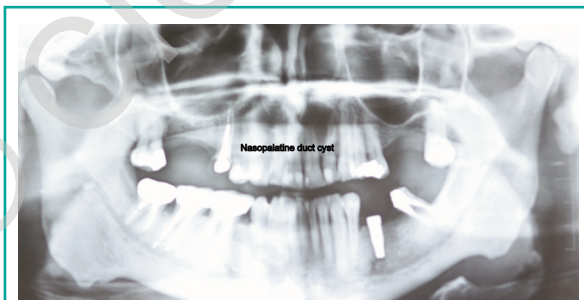


Figure 6
Panoramic X-rays shows a heart shaped radiotransparency; this image is due to the presence of the anterior nasal spine.

Results

Out of 2098 patients evaluated for surgery treatments of cystic lesion in the period of this study (10 years), thirty-six cases were diagnosed as NPDC (52% males, and 48% females). There was no significant age difference at time of diagnosis between males and females. The majority of the patients were males (20 cases, 52%, 1099 unit.), while the NPDC rate in females was 48% (999 unit.). The patient's mean age at the time of diagnosis was 53 years (18), higher in female patients 55 years (19) than in males 52 years (17) ($p=0.001$). The overall age mean is 44,72 years and the SD is 9,50 (Tab. 2). Ten patients presented antecedent diseases of interest: 5 arterial hypertension, 4 diabetes mellitus type II, 3 idiopathic osteoporosis.

Table 2 - Patients diagnosed with naso palatine duct cyst (n = 36). SD* = standard deviation.

| Gender | Age mean and SD* |
|-----------------------|------------------|
| Male | 46,4 (years) |
| 20 | S.D.: 6,68 |
| Female | 42,62 (years) |
| 16 | S.D.: 10,78 |
| Total subjects | 44,72 (years) |
| 36 | S.D.: 9,50 |

Regarding toxic habit: only four patients was smokers, nobody reported alcohol consumption. For one patient 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 vitality of the teeth adjacent to the lesion (permanent upper central incisors) was almost always preserved (19 cases). Most of the patients were asymptomatic (30 cases), while six showed local inflammation, and the remaining four reported pain and ulceration produced by dentures.

The position of the NPDCs was mostly superficial or palatine (30 cases), while in the remaining 6 cases the lesions were located deep or in the nasal region, a computed tomography scan of all those patients was requested to precisely establish the NPDCs position.

In all cases, treatment consisted in completely removing the lesion through a combination of cold scalpel and piezosurgery (PIEZOSURGERY® touch starter Mectron), with careful dissection of the neurovascular bundle.

In 35 cases (95.45%) a palatine approach was used, with vestibular access in a single case (4.54%).

The patients operated upon the palatine approach, 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.

The follow-up mean duration was one year, until correct ossification of the surgical zone was confirmed by X-ray study.

Following surgical exeresis, 33 lesions healed completely after the first operation, while the remaining three cases suffered relapse after three years of follow-up.

This situation required a second intervention (removal of the NPDC with ligation of the nasopalatine neurovascular bundle), after which complete healing was confirmed within for years.

Discussion

Nasopalatine duct cyst (NPDC) is of uncertain origin and shows a peak incidence between the fifth and the sixth decades of life.

The differential diagnosis must be established with other conditions such as an enlarged nasopalatine duct, 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 buccosinusual communication (1). The literature reports a male predilection, that was confirmed in our study. Thus far no racial predilections have been established (1).

In accord with the majority of studies so far reported in literature, these lesions appear mainly between the fourth and sixth decades of life.

Although the etiology underlying these lesions is not clear, in addition to the hypothesis of spontaneous proliferation from embryonic tissue remains, other possible etiologies have been proposed - such as prior trauma, poorly fitting dentures, local infection, genetic and racial factors.

Most of the cysts are asymptomatic (30 of the 36 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).

The diagnosis is based on clinical history, clinical exploration and complementary tests.

Radiological exams (panoramic X-rays, periapical and occlusal X-rays and computed tomography) are necessary to correctly diagnose NPDCs.

Radiologically, the lesions appear as a well delimited radiotransparency, measuring often 1-2 cm in diameter. The X-ray image is predominantly rounded, ovoid or heart-shaped (Fig. 3).

The nasopalatine neurovascular bundle is a delicate and highly vascularized structure giving rise to profuse bleeding if inadvertently sectioned during surgery.

We therefore consider that the pizosurgery inserts offer adequate safety in such surgical procedures, and used in our 20 NPDCs exeresis, reduced postoperative pain and edema.

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.

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).

Conclusion

Nasopalatine duct cyst are the most common non-odontogenic cyst of the oral cavity seen in the general population.

NPDCs must be distinguished from other maxillary anterior radiolucencies. Vitality testing of teeth adjacent to or involved in a cyst-like lesion is mandatory and the final diagnosis could only be performed after histological analysis.

The treatment of choice is surgical exeresis of the cyst, although some authors propose marsupialization of large NPDCs (1).

The definitive diagnosis is established only by histological study of the lesion.

Therefore, in 2098 subjects evaluated in the period of the study, the incidence of the NPDC is 1,71%. In conclusion, in accord with the literature on the subject, we can consider NPDC a rare pathology and simple surgical resection is recommended, followed by clinical and radiological control to ensure correct resolution of the case (Fig. 7). The cystic epithelium varied considerably both among different cases and within the same cyst. Respiratory-type epithelium was presented in our histological sections (Figs. 8-10).

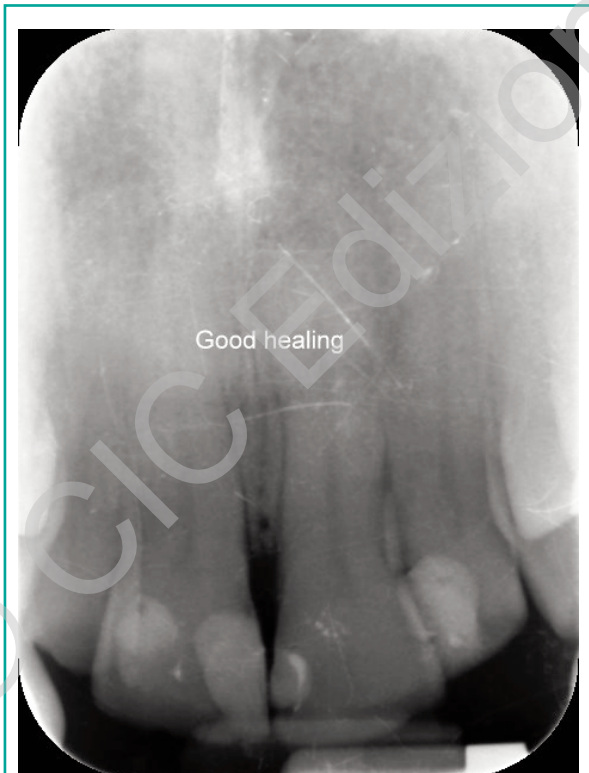


Figure 7
Periapical X-ray 1-year after surgery shows good healing with complete bone fill evident.

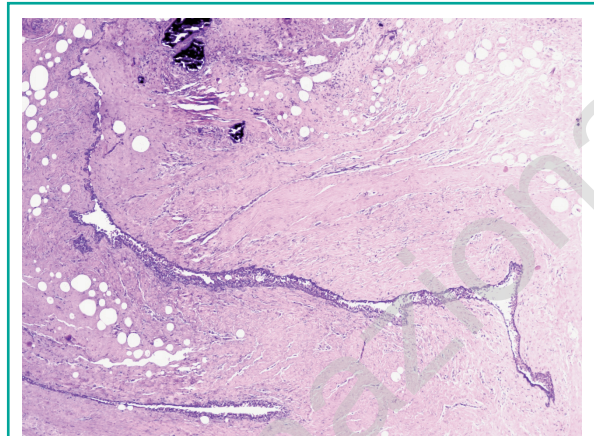


Figure 8
Histological section of a nasopalatine duct cyst following hematoxylin-eosin staining, ciliary respiratory-type epithelium with calcifications and adipose cells is observed (H&E 4x).

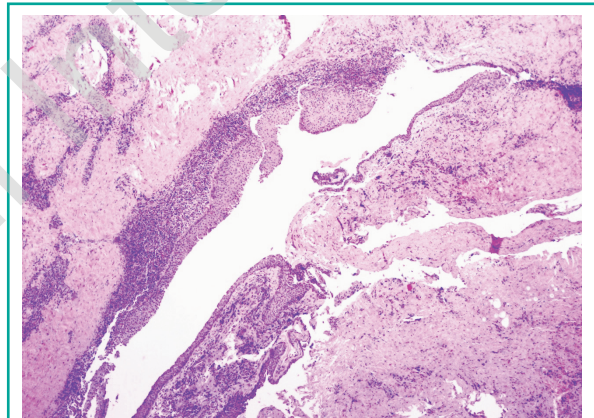


Figure 9
A particular of the ciliary respiratory-type epithelium (H&E 4x).

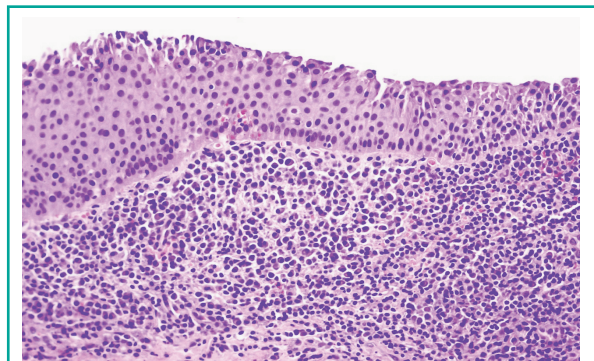


Figure 10
Histological section of a nasopalatine duct cyst following ematoxylin-eosin staining: a respiratory-type epithelium with squamous metaplasia is evident. The fibrous tissue and the stroma. The presence of a basal membrane (H&E 10x).

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