

# 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

01 in 1914 by Meyer (2). These lesions, also known by other  
02 names such as anterior middle cyst, maxillary midline  
03 cyst, anterior middle palatine cyst, and incisor duct cyst,  
04 were regarded as fissural cysts in the past (3). At present,  
05 according to the classification of the World Health Or-  
06 ganization (WHO), these lesions are regarded as deve-  
07 lopmental, epithelial and non-odontogenic cysts of the  
08 maxillae, along with nasolabial cysts (4).

09 NPDCs are the most common non-odontogenic cysts  
10 of the oral cavity, representing up to 1% of all maxillary  
11 cysts (5). These lesions are more almost three times more  
12 frequent in males than in females (6). The maximum inci-  
13 dence is between 40 and 60 years of age. Due to a lack of  
14 representative studies, it is not fully clear whether NPDCs  
15 are more common in Caucasians, negroes or Asians (7).  
16 Often mistaken for an enlarged nasopalatine duct, NPDCs  
17 are of uncertain origin. The spontaneous proliferation  
18 theory appears to be the most likely explanation (a num-  
19 ber of studies have reported cystic degeneration in the  
20 incisor duct and on the midline of the palate in human  
21 fetuses)(8).

22 Histologically, the type of cystic epithelium varies accor-  
23 ding to the location involved (palatine, nasal or interme-  
24 diate). A squamous cell epithelium is almost always ob-  
25 served, though a ciliary respiratory-type epithelium can be  
26 seen when the lesion is located higher up or nasally (7).

27 NPDCs are normally asymptomatic, constituting casual  
28 radiological findings, though sometimes (in 17% of cases)  
29 patients report pain due to the compression of structures  
30 adjacent to the cyst, particularly when the latter becomes  
31 overinfected, or in patients who wear dentures that com-  
32 press the zone. The more caudal the location of the cyst,  
33 the sooner symptoms appears. These normally manifest  
34 as an inflammatory process (46% of cases) that rarely  
35 produces facial asymmetry, since growth or expansion is  
36 intraoral (palatine). The more advanced cases are able to  
37 cause pain and itching (9).

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

48 The present study describes the clinicopathological charac-  
49 teristics of 22 NPDCs and discusses their possible etiology,  
50 incidence, treatment and prognosis, with a comparative  
51 review of the literature on the subject.

## 52 53 **Patients and Method**

54 A retrospective observational study was made comprising  
55 a period of 36 years (1970-2006), and selecting a series  
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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 electrosca-  
pel 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 im-  
mediate 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 diag-  
nosed 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 in-  
terest: 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 pa-  
tients showed imbalance of their removable complete  
upper dentures. In 8 patients the cysts were associated to  
a history of chronic permanent upper central incisor infec-  
tion secondary to trauma, a periodontal problem, or failed

01 endodontic treatment. In two patients the nasopalatine  
 02 duct could have become infected via the nasal route, since  
 03 they presented chronic bacterial rhinitis without symptoms  
 04 at the time of diagnosis. The remaining 8 patients in the  
 05 series presented no pathology associated to the lesion.  
 06 The vitality of the teeth adjacent to the lesion (permanent  
 07 upper central incisors) was almost always preserved (14  
 08 cases, 63.63%). However, in 8 cases the teeth had been  
 09 subjected to endodontic treatment, or the pulp tissue  
 10 was necrotic. Percussion was generally negative (18 cases,  
 11 81.81%), with the exception of those adjacent teeth which  
 12 had suffered some periodontal or periapical problem  
 13 (4 cases, 18.18%). The mean radiological diameter of  
 14 the lesion was 14 mm (12 mm in females and 16 mm in  
 15 males). The X-ray image of the lesion was heart-shaped  
 16 in 10 cases (Figures 1 and 2), and rounded or oval in 12  
 17 cases (Figure 3).  
 18 Most of the patients were asymptomatic (14 cases), while  
 19 four showed local inflammation, and the remaining four  
 20 reported pain and ulceration produced by dentures.  
 21 The position of the NPDCs was mostly superficial or  
 22 palatine (15 cases), while in the remaining 7 cases the  
 23 lesions were located deep or in the nasal region (Figure  
 24 4). A computed tomography scan was requested of all the  
 25 patients to precisely establish the position of the NPDCs  
 26 (Figure 5).  
 27 In all cases, treatment consisted of complete removal of  
 28 the lesion using the cold scalpel, with careful dissection of  
 29 the nasopalatine neurovascular bundle. Lastly, the electro-  
 30 scalpel was used (Figure 6) for correct bleeding control of  
 31 the surgical bed. In 21 cases (95.45%) a palatine approach  
 32 was used, with vestibular access in a single case (4.54%).  
 33 There were no peroperative complications. However, the  
 34 11 patients operated upon via the palatine approach with  
 35 no palatine plaque placement immediately after surgery  
 36 reported discomfort due to important swelling, pain and  
 37 tenderness in the anterior region of the hard palate (sub-  
 38 mucosal hematoma) during the first two postoperative  
 39 weeks.  
 40 From the histological perspective, the lesions presented  
 41 exclusively squamous cell epithelium in 15 cases (68.18%),  
 42 while the remaining 7 cases (31.82%) moreover also  
 43 showed ciliary cylindrical epithelium (Figure 7). The  
 44 presence of melanin was observed in one case.  
 45 The mean duration of follow-up was one year, until co-  
 46 rrect ossification of the surgical zone was confirmed by  
 47 X-ray study. Following surgical exeresis, 20 lesions healed  
 48 completely after the first operation, while the remaining  
 49 two cases suffered relapse after three years of follow-up,  
 50 i.e., the relapse rate in our series was 9.09%. This situation  
 51 required a second intervention (removal of the NPDC with  
 52 ligation of the nasopalatine neurovascular bundle), after  
 53 which complete healing was confirmed within two years.  
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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.

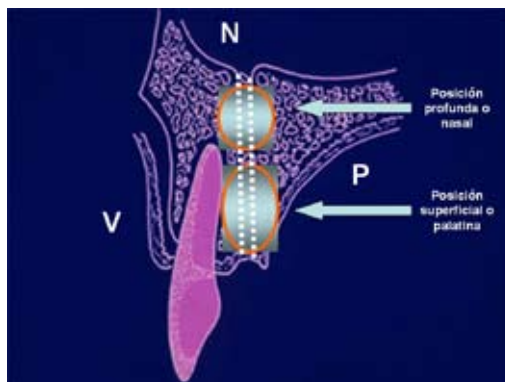


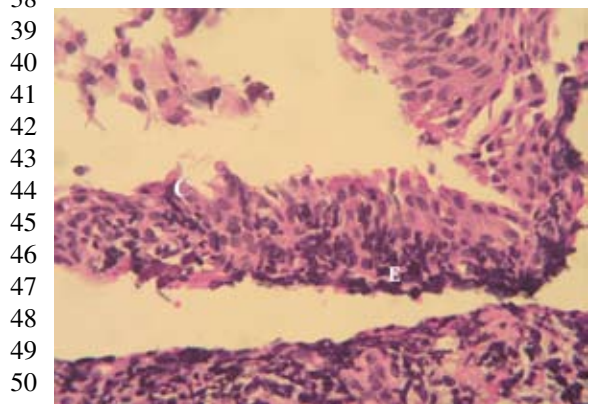
Fig. 4. Schematic representation showing the two most frequent locations of nasopalatine duct cysts.



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17 **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.



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



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51 **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-

01 der 6 mm in size are regarded as enlarged incisor ducts of  
 02 a non-pathological nature (2).  
 03 A thorough differential diagnosis must be established in  
 04 order to avoid unnecessary treatments such as endodontic  
 05 procedures in vital permanent upper central incisors (1, 6).  
 06 A correct tentative diagnosis should be based on positive  
 07 dental vitality testing and negative percussion findings of  
 08 the permanent upper central incisors, provided these teeth  
 09 do not have pulp or periodontal problems (6). Radiolo-  
 10 gical exploration is essential for diagnosing NPDCs, and  
 11 in addition to panoramic X-rays, other complementary  
 12 techniques are advised, such as periapical and occlusal  
 13 X-rays and computed tomography. The latter technique  
 14 (requested in all 22 cases of our series) offers maximum  
 15 guarantees in establishing a tentative diagnosis, since it  
 16 generates great detail of the structures (normally intact)  
 17 adjacent to the lesion. Computed tomography easily  
 18 visualizes the radiotransparency on the midline, with  
 19 well defined sclerotic margins, and informs of the exact  
 20 location of the lesion. In addition, it facilitates planning  
 21 of the best surgical approach (normally palatine, as in 21  
 22 of our 22 cases)(8, 16).  
 23 The mean radiographic size of the lesion tends to vary  
 24 according to patient gender (4). In our study, the inter-  
 25 gender difference in mean diameter was 4 mm (12 mm in  
 26 females and 16 mm in males).  
 27 The differential diagnosis must be established with other  
 28 conditions such as an enlarged nasopalatine duct (less than  
 29 6 mm in diameter), central giant cell granuloma, a root cyst  
 30 associated to the upper central incisors, a supernumerary  
 31 tooth follicular cyst (normally mesiodens), primordial  
 32 cyst, nasoalveolar cyst, osteitis with palatal fistulization,  
 33 and bucconasal and/or buccosinusual communication (1).  
 34 Other diagnostic techniques can be used to radiologically  
 35 assess lesions of this kind, such as multimodal tomogra-  
 36 phy, which in addition to exposing the patient to lesser  
 37 radiation doses employs crossed and sectional tomo-  
 38 graphic acquisitions in the sagittal plane to yield three-  
 39 dimensional images (17). Magnetic resonance imaging  
 40 (MRI) may also prove useful in establishing the diagnosis,  
 41 and particularly contrast the interior of the NPDC with a  
 42 high signal intensity. Specific axial T1-weighted imaging  
 43 reflects the presence of fluid, viscous and protein mate-  
 44 rial within the cyst, and abundant keratin at superficial  
 45 level. Thus, MRI is highly reliable in diagnosing NPDCs,  
 46 discarding root cysts or any other cysts of odontogenic  
 47 origin (3, 18, 19).  
 48 The treatment of choice is surgical exeresis of the cyst,  
 49 although some authors propose marsupialization of large  
 50 NPDCs (2, 11). The nasopalatine neurovascular bundle is  
 51 a delicate and highly vascularized structure giving rise to  
 52 profuse bleeding if inadvertently sectioned during surgery.  
 53 Electrocoagulation is required in such cases. We therefore  
 54 consider that the electroscaquel offers adequate safety in  
 55 such surgical procedures, and used it in all 22 NPDCs in  
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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

01 asymptomatic. The tentative diagnosis is based on the cli-  
 02 nical history, the clinical exploration, and complementary  
 03 tests (particularly computed tomography). Where present,  
 04 irritative factors should be eliminated, and early surgical  
 05 removal is advised in order to avoid possible maligniza-  
 06 tion. The definitive diagnosis is established by histological  
 07 study of the lesion. Following resection, relapse is unlikely,  
 08 though a postoperative follow-up of at least one year is  
 09 indicated in all cases.

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