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# **Case report**

# Unusual growth of a nasopalatine cyst

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# ABSTRACT

The main goal of this work is to report the aggressive potential of the nasopalatine cyst (NPC) in the pre-maxilla, when there's invasion of the medullar spaces.

Bone resorption, soft tissue swelling, and displaced root remnants were clinical and radiological signs of this long-term asymptomatic and slow progression lesion.

Histopathology showed a mixture of stratified non-queratinized with pseudostratified epithelium compatible with NPC.

Careful should be made when screening patients since this entity can easily go unnoticed and the slow painless progression can lead to severe hard tissue defects.

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# Crescimento incaracterístico de um quisto nasopalatino

#### RESUMO

É objectivo deste trabalho reportar o potencial agressivo do quisto nasopalatino (NPC) quando existe invasão dos espaços medulares.

Como sinais clínicos e radiográficos desta lesão assintomática, e de progressão lenta, observamos reabsorção óssea, tumefacção dos tecidos moles e afastamento de restos radiculares.

Ao exame anatomopatológico verificamos uma mistura de epitélio pavimentoso estratificado com epitélio pseudoestratificado compatível com NPC.

Devemos ser cuidadosos nas consultas de rotina uma vez que esta lesão pode facilmente passar despercebida e a sua lenta e indolor progressão pode levar a defeitos ósseos severos.

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# Introduction

The non-odontogenic fissural NPC was first described in 1914 by Meyer, as a result of an inflammatory process resulting in the abnormal growth of the remnant cells, derived from the fusion of the primary palatal of the first branquial arch <sup>1</sup> It is a developmental cyst.

However aetiology is still debatable, as some say it may arise from a vestigial organ (nasal-vomer Jacobson organ) present in some inferior mammals.<sup>2</sup>

The NPC is the most common non-odontogenic cyst in the oral cavity, responsible by  $0.8\%^3$  to  $33\%^4$  overall, predominantly in males, ranging in the literature from 1.1:1 to  $20:1.^{5-7}$ 

It is more common in the fourth and sixth decade of life.<sup>8</sup>

The cyst development is attributed and triggered by several factors: infection (38%), trauma (16%), minor salivary mucus retention, inflammatory stimulus, spontaneous proliferation of epithelial remnants present in the nasopalatine duct and idiopathic origin.<sup>9</sup>

The growth is slow and often asymptomatic, only noticed by palatal swelling or pain in the incisive foramina.

Radiologically it can be unnoticed, since nasal fossae can produce a superimposing image, making the diagnostic very difficult.

When cysts evolve into the pre-maxilla medullar spaces, growth can be both inward to the nasal fossa and outward towards the oral cavity, resorbing bone, and creating large defects.

Malignant transformation is rare, and there are only a few reports in the literature. $^{10}$ 

This case report shows an unusual growth of this entity and his treatment.



Figure 1 - Pre-op photo



Figure 2 - CT scan evaluation

#### Biopsy

Patient made 1-week Hexetidine 0.1% (Hextril<sup>®</sup> 1mg/ml Johnson & Johnson, Lda) rinse twice a day, and was pre-medicated with Amoxicillin 875 mg and Clavulanic-acid 125 mg, two days before the surgery following the regimen for 8 days.

Articain 4% (clorhidrate-articain 1:100000 epinephrine) for buccal infiltration and Mepivacain 3% (clorhidrate-mepivacaine) for palatal anaesthesia were used. Mid-crestal incision was made on sound bone and a split incision on the area of the cyst, for complete enucleation. Two vertical release incisions left and right were made.

The cyst was enucleated and preserved in 10%. formaldehyde (figs. 3 and 4).

The area was sutured with Poliglatin-910 (Vycril 4.0 Ethicon<sup>®</sup>) and the denture rebased with soft material (FITT<sup>®</sup>).

#### Histopathology examination

The histological examination reveals a cyst-like lesion with a well-defined epithelium. Pathological findings reveal squamous or respiratory cell types, or a combination of these, infiltrated by inflammatory cells surrounding a matrix of crystalline and hemorrhagic cells. (fig. 5)

## **Case Report**

Male 59 years old, Caucasian, smoker of 20 cigarettes a day, without known systemic pathology, presented himself to the Oral Surgery Department of the Lisbon College of Dentistry to remove residual root fragments.

At clinical inspection, swelling of the pre-maxilla was noticed, with absence of pain but with displaced root fragments (fig. 1). The patient wore a removable denture that was: unstable, unfit and fractured in multiple sites.

In the radiological (orthopantomography) examination, severe resorption of the pre-maxilla was noticed. The patient was sent for a Computer Tomography Scan (CT-Scan) (fig. 2).

#### Radiological interpretation

At ct-scan interpretation, the resorption was severe and bone replaced by a rounded circumscribed lesion, with no invasion of adjacent structures.

An excisional biopsy was planed and the lesion was sent for histopathology for confirmation of radiologic and clinical provisional diagnose.



Figure 3 - Intraoperatory photo



Figure 5 - Histopathology Exam



Figure 4 - Cyst enucleated

## Follow-up

The patient wore a removable denture for 6 months (healing time) and after that period he was sent for implant-supported overdenture.

At 6-month postoperative follow-up the defect made by the NPC was visible. (fig. 6)



Figure 6 - 6 Month Follow up

# Discussion

NPC are benign lesions that if left untreated can lead to abnormal growth. Diagnose is often difficult and can easily be unnoticed at early stages.

The NPC can be divided into two types: cyst of the incisive canal and cyst of the incisive papilla, both showing the same pattern of growth.

The pattern of development is slow and rarely malignant but the potential of damaging is great when medullary cortex is penetrated.<sup>11</sup>

In literature there are reports of several cases of abnormal growth when left untreated.  $^{\rm 12-14}$ 

Differential diagnose is made with: radicular cyst, residual cyst, keratocyst, and central bone tumours.

The probability of recurring is low, ranging from 0-2%.<sup>15,16</sup> In this case the progression was slow which lead to cortex/medullary spaces invasion, resorbing bone in a crater-like lesion.

The greater the lesion more difficult the surgery is, which in this case was due to the presence of the nasopalatine artery and the surrounding branches.

The patient was asymptomatic for years, and the chief complaint was the presence of residual root fragments.

In NPC pain and swelling are not initial features of this entity that bears unnoticed in the initial stages, the lesion is often misdiagnosed by the superimposing nasal fossae.

It is only clinically relevant at later stages of the disease.

The types of epithelia that line the nasopalatine duct are highly variable, depending on the relative proximity of the nasal and oral cavities.

Typically the most superior part of the ducts is characterized by a respiratory-type epithelial lining. Moving downward to the oral cavity the lining changes to cuboidal epithelium. In the most inferior portion, squamous epithelium is the usual type.

In this case we found both respiratory and squamous type of epithelial cells.

#### Conclusions

Careful must be take when screening patients and, panoramic radiograph and ct-scans should be mandatory to evaluate cyst like lesions.

The diagnosis is based on the clinical history, the clinical exploration, and radiological exams (particularly CT Scan).

Early surgical removal is advised in order to avoid bone defects and 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.

#### REFERENCES

- 1. Abrams AM, Howell FV, Bullock WK. Nasopalatine cysts. Oral Surg Oral Med Oral Pathol. 1963;16:306-32.
- Escoda FJ, Almendros MN, Berini AL, Gay Escoda C. Nasopalatine duct cyst: report of 22 cases and review of the literature. Med Oral Patol Oral Cir Bucal. 2008;1;13 E438-43

- Killey HC, Kay LW. Benign Cystic Lesions of the Jaws, Their Diagnosis and Treatment. 2<sup>nd</sup> ed. New York, NY: Churchill Livingstone; 1977.
- Burket LW. Nasopalatine duct structures and peculiar bony pattern observed in the anterior maxillary region. Arch Path. 1937;23:793-800.
- Regezi JA, Sciubba JJ. Oral Pathology: Clinical Pathologic Correlations. 3<sup>rd</sup> ed. Philadelphia, Pa: WB Saunders; 1999.
- 6. Cabrini RL, Barros RE, Albano H. Cysts of the jaws: a statistical analysis. J Oral Surg. 1970;28:485-9.
- Vasconcelos R, de Aguiar MF, Castro W, de Araújo VC, Mesquita R. Retrospective analysis of 31 cases of nasopalatine duct cyst. Oral Dis. 1999;5:325-8.
- Elliott KA, Franzese CB, Pitman KT. Diagnosis and surgical management of nasopalatine duct cysts. Laryngoscope. 2004: 114:1336-40.
- Takagi R, Ohashi Y, Suzuki M. Squamous cell carcinoma in the maxilla probably originating from a nasopalatine duct cyst: report of case. J Oral Maxillofac Surg. 1996;54:112-5.
- Curtin HD, Wolfe P, Gallia L, May M. Unusually large nasopalatine cyst: CT findings. JComput Assist Tomogr. 1984; 8:139-42.
- Tanaka S, Iida S, Murakami S, Kishino M, Yamada C, Okura M. Extensive nasopalatine duct cyst causing nasolabial protrusion. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2008;106:e46
- 12. Torres LM, Benito JI, Morais D, Fernández A. Nasopalatine duct cyst: case report Acta Otorrinolaringol Esp. 2008;59:250-1.
- Swanson KS, Kaugars GE, Gunsolley JC. Nasopalatine duct cyst: an analysis of 334 cases. J Oral Maxillofac Surg. 1991; 49:268-71.
- 14. Hertzanu Y, Cohen M, Mendelsohn DB. Nasopalatine duct cyst. Clin Radiol. 1985;36:153-8
- Harris IR, Brown JE. Application of cross-sectional imaging in the differential diagnosis of apical radiolucency. Int Endod J. 1997;30:288-90.
- Hisatomi M, Asaumi J, Konouchi H, Shigehara H, Yanagi Y, Kishi K. MR imaging of epithelial cysts of the oral and maxillofacial region. Eur J Radiol. 2003;48:178-82.