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RADIO "LOGICAL" REFLECTIONS

A radiolucent heart

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CASE PRESENTATION

Mrs. G., 47 years old in good health, consults for moderate canine-to-canine crowding, that she wishes to treat. She was prescribed a periapical series of x-rays, and the images taken of the incisors showed lacunae that attracted attention. Additional examinations were requested of her, and pulpal vitality tests were done: 11, 21, and 22 responded positively. 12 had a previous root canal and a ceramo-metal restoration.

DESCRIPTION OF THE RADIOGRAPHIC FILE

The radiographic file, centered on the area of the maxillary incisors is composed of 5 images using the long cone paralleling technique (Fig. 1 a-e) and of multiplanar reconstructions using limited field cone beam computed tomography imaging (CbCt): the axial slice passing through the apical third of the incisors (Fig. 2); frontal distal cut at the anterior nasal spine (Fig. 3), and paramedian dento-axial cuts, passing through the roots of 11 and 12 (Fig. 4 a-e).

From the periapical films:

– a voluminous radiolucent image is projected in the region of the anterior nasal spine, centered on the apical extremity of 11 and 12 and extended to part of the other side up to the apices of 12 and 22 which is its limit, measuring approximately one centimeter higher towards the nasal fossae. Its lateral borders are clearly defined by cortical walls but it is very blurred in its inferior and superior aspects.
– 11 presents a different image on the film 1b and 1c: its root appears short on the distal-eccentric view (1c), and on the contrary respected on the ortho-centered image (1b), with a superimposed lacuna on the root well delimited with respect to the *lamina dura*;
– 12 presents a ceramo-metal crown, with a post and a radiopaque image showing a root canal. A discrete lacuna a few

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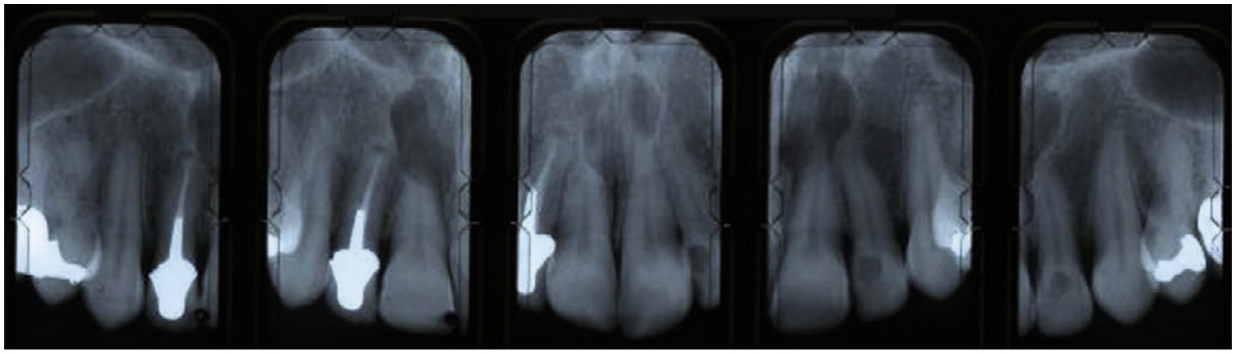


Figure 1
Patient #1: Periapical views obtained using the long cone paralleling technique in the incisor-canine area (Dr. Pasquet and Dr. Cavezian, Paris).

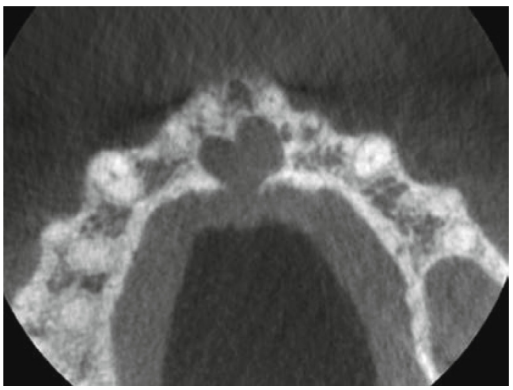


Figure 2
Horizontal (axial) reconstruction of the maxilla passing through the apical 1/3 of the maxillary incisors of patient #1, obtained by a cone beam centered on the incisor area (limited field CbCt, Planmeca Romexis™, Helsinki, Finland).

Figure 3
Frontal reconstruction, distal to the anterior nasal spine of patient #1 (Planmeca Romexis 3D Viewer™, Helsinki, Finland).

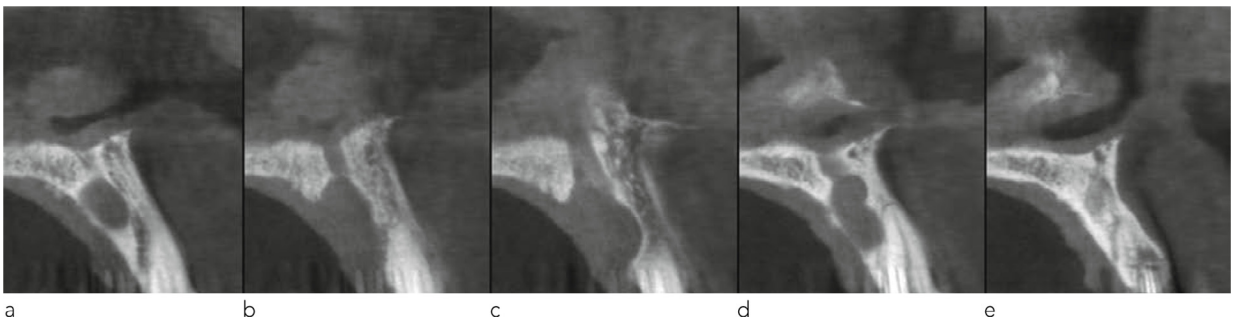


Figure 4a-e
Dental axis reconstructed cuts in the right incisor region (11, 12) of patient #1 (Planmeca Romexis 3D Viewer™, Helsinki, Finland).

millimeters in size, is projected on its apical extremity;
– 22 presents an image with radiolucency at the cingulum, rounded and well delimited, resembling an old composite restoration, that is non-radiopaque (Fig. 1 d and e).

From the sectional images

– in the horizontal plane, one notes in the median anterior region of the hard palate an image of the density of water, in the form of a heart found on a playing card, asymmetrical with a dominance on the left side, whose lobes are equidistant from the roots of maxillary central incisors, and the tip is mixed with the palatal fibro-mucosa. Its borders are well delimited, and its expanse is on the order of a centimeter (Fig. 2);
– on the frontal reconstruction (Fig. 3), one finds the image of an asymmetrical heart, where the left lobe is dominant, and where the tip is mixed with the palatal mucosa, and the lobes extend to the mid-height of the hard palate, without reaching the nasal fossae;
– on the dento-axial cuts of Figure 4 (sagittal and para-median), one notes an oval aspect well defined by the lacuna of corresponding to the right lobe (Fig. 3a), of which the axis follows the alveolar bone; the left lobe appears doubled (Fig. 4d). The cystic image is projected over the incisive canal and is delicately corticalized in its anterior portion (juxtaposed over the teeth) on all the images. The cyst does not seem to be continuous with the incisive canal but rather superimposed on it, because one can distinguish the rounded borders of the cyst from the walls of the canals (Fig. 4c and 4d). It seems to push the incisors buccally;
– on the 4e cut, at the level of tooth 12, we were able to show an apical radiolucency evoking a granuloma of endodontic origin, independent of the neighboring cyst.

WHAT DIAGNOSIS COMES TO MIND?

The naso-palatine cyst, or cyst of the incisive canal, is part of the non-odontogenic epithelial cysts of the jaws, namely, arising from epithelial structures other than embryological dental structures[10]. It could be from a malformative origin, derived from epithelial remnants in the naso-palatine canal, a structure found only in the fetus[7]. The hypothesis for the formation of the naso-palatine cyst based of epithelial remnants in the fusion of the primary and secondary palate is not retained today.

The frequency of naso-palatine cysts are in the fourth position among cysts of the jaws, after radicular, follicular cysts and odontogenic keratocysts, with a prevalence in the order of 1.5% within the general population. As these naso-palatine cysts are generally asymptomatic (except for secondary infection), and discovered fortuitously during a radiological ex-

amination it is estimated that true incidence is under-estimated.

Most of the studies conclude that there is a greater incidence among men than women (ratio men/women of 1.7/1 to 3/1)[6,15], especially around 50. Some cases have been described in infants[12,18].

The lesion formed by the naso-palatine cyst is radiolucent, round, ovoid, pear or heart shaped, and located in the midline, below or between the roots of the maxillary central incisors. The dental roots can be displaced . The heart shape, well known by clinicians, results in super-imposition of the anterior nasal spine or as well as the nasal septum on that of the cyst; but the typical image is not always found. The differential diagnosis[6,15], in the most frequent cases, can be with the normal image of the incisive foramen, which can be quite large, that of an apical cyst of endodontic origin, odontogenic keratocyst, or that of a dentigerous (follicular) cyst derived from a mesiodens.

The normal image of the incisive canal is shown in Figures 5 and 6, CT images obtained in this case for the localization of an impacted maxillary right canine. On the maxillary axial cut (Fig. 5), parallel to the palatal plane and passing through the apical third of the incisors, on can visualize the 2 incisive foramina (stoma of the incisive canal), either side of the median sagittal suture. On the dento-axial cuts (Figs. 6a, b, c, from the most medial to the most lateral, Dentascanner™), the canal appeared like a regular image, linear, extended from the palatine fibro-mucosa to the homolateral nasal fossa, with a diameter in millimeters. Its density is that of water, the canal is

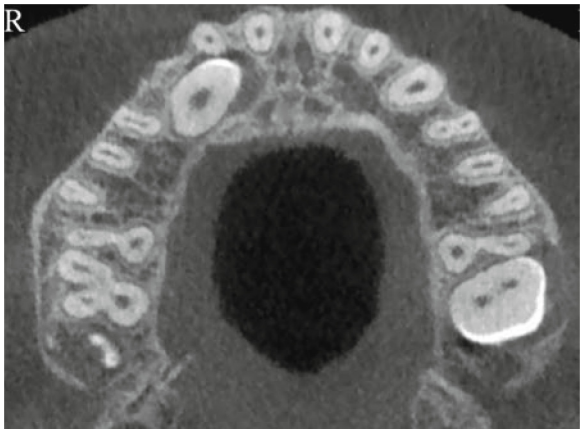


Figure 5
Axial scanner cut of the maxilla of patient #2, presenting an impacted maxillary canine. Normal appearance of the incisive canal (two foramina).

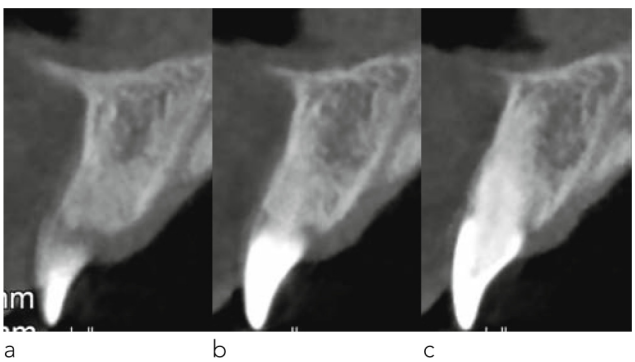


Figure 6
Reconstructed dento-axial cuts (Dentascan™).

well defined by a dense cortical edging.

– In the case of the apical lesion of endodontic origin, we find the loss of vitality of the adjoining incisors. In the present case, all the incisors are vital with the exception of 12. This last one presents an apical image evoking a granuloma visible on the periapical images (Fig. 1a and b), but which do not communicate with the voluminous cystic image (Fig. 4e).

Several case reports have reported naso-palatine cysts treated erroneously as inflammatory lesions of endodontic origin[8,13]. . .
- Odontogenic keratocysts are difficult to distinguish radiographically and clinically from naso-palatine cysts, and only the anatomopathological examination makes it possible to establish a definitive diagnosis.
- A dentigerous (follicular) cyst presents as a homogenous radiolucent shape, singular, regular, well defined with a dense border, associated with the crown of a tooth included or during eruption (in this locality, a mesiodens). No image of enamel-dentinal density was found, that allowed us to exclude this hypothesis.
- Finally, the possibility of a tumor cannot be excluded, benign or malignant.
- Although rare, chondrosarcomas can be found in this region[15].

HOW SHOULD WE PROCEED AND WHAT IS THE POTENTIAL IMPACT ON ORTHODONTIC TREATMENT[1,2,10,15-17]?

When there is suspicion of a naso-palatine cyst, it is recommended to take a radiographic exam at least two different beam orientations (standard radiograph and occlusal film)[9,15]. The largest lesions will require sectional images by an CTscan or CbCT, allowing the determination of the involvement of the neighboring structures (respect of dental roots, thinning and repression of the cortical walls); if they are not diagnosed early, the cysts can not only extend toward the buccal and palatine cortical walls, but also towards the nasal fossae, complicating surgical treatment[11]. It is important to note that the absence of symptoms is not correlated with the dimensions of the naso-palatine cyst; the larger they are, the more the risk of surgical complications increases[14].

Treatment depends of the size of the cyst and of its three dimensional expanse. Marsupializtion (or cystotomy or Partsch I), under local anesthesia, can be chosen faced with large cysts to avoid the formation of a persistent fistula or the loss of vitality of the nearby dental organs. The designation under the term "marsupialization" is due to the fact that the procedure forms a pocket that communicates with the exterior producing an opening in the cystic cavity (comparable to those of marsupials) and in suturing the periphery of this opening to the lips of the cutaneous incision. The placing of a drainage tube permits the reductions of the intracystic pressure, and is accompanied, in parallel by the regression in the volume of the cyst, with spontaneous peripheral ossification.

Surgical enucleation (Partsch II) by a palatine track is the usual recommended treatment, often associated with the reconstruction of the osseous defect by appositional or biomaterial grafts. A proportion of patients presents with spontaneous (complete

in 82% of the patients operated on in the study of Anneroth *et al.* (1986)[3], by some present persistent osseous defects with the absence of regeneration.

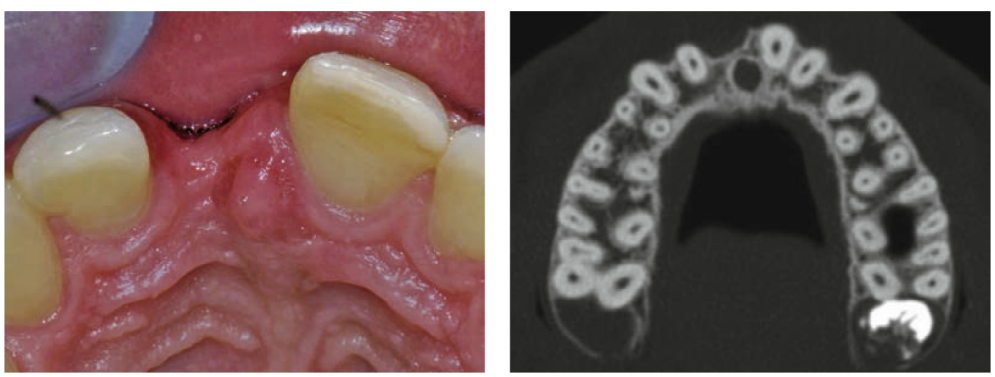
Enucleation must be complete in order to avoid relapse, which can manifest itself beyond the 5th year post surgery[4]. The surgical complications of these procedures are known, such as hemorrhagic or infectious, but also with the creation of persistent fistulas (oro-nasal or oro-antral), the loss of vitality in teeth or anterior palatal paresthesia.

In the cases illustrated in Figures 7 to 11, a naso-palatine cyst was discovered fortuitously during a pre-implant appraisal following the traumatic avulsion of a central incisor in a young adult:

– clinically, we observed a collapse of the alveolar bone in the bucco-lingual direction, and perhaps at the level of the soft tissues a relief a little marked of the retro-incisive papilla, without a symptom (Fig. 7 a);
– on the native image of the corresponding CT scan (Fig. 7 b), one remarks on the buccal bone loss with regard to the missing 11, and the round image of the cyst, of which the walls appear to be clean and corticalized with a diameter of 6-7 mm. Its borders are in almost contact with the buccal cortical bone, thin, contra-indicating the placing of an implant in that location;
– on the sagittal reconstruction (Fig. 8), one visualizes the enlarged image of the incisive canal, from the palatal to the nasal portions.

Given the wish of the patient to benefit from an implant supported reconstruction, it was decided to use an osseous reconstruction with an appositional graft, in parallel with the enucleation of the cyst; the prior information available concerning the patient stressed the cumulative risks of different procedures, complicated by the presence of the cyst[5].

On Figure 9, the amplitude of the looming buccal osseous defect and



a b

Figure 7a and b
Occlusal intraoral view before treatment of patient number 3; b: Corresponding Scanner RX axial cut.

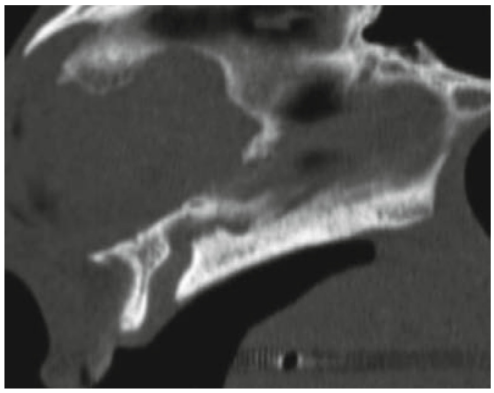


Figure 8
Scanner reconstruction passing through the anterior nasal spine region (patient number 3).

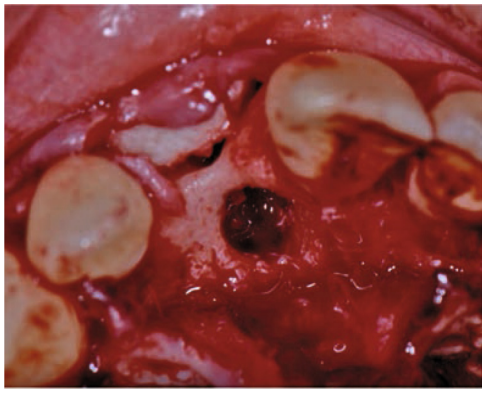


Figure 9
Intraoral view during the surgical procedure, one reflected flap showing the cystic cavity and the vestibular osseous defect.

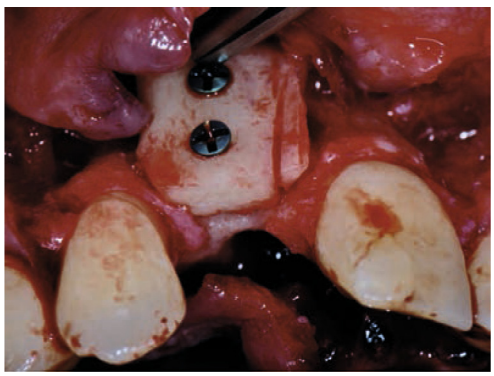


Figure 10
Appositional graft (ceramic) screwed to the osseous vestibular wall.

Figure 11
Intraoral view of the bone cicatrization and mucosa of the inserted implant.

the cystic cavity required an appositional bone graft of cortical bone (Fig. 10); a filling using biomaterial made of osseous filling (Bio-oss™) was utilized for the cystic cavity once the naso-palatine cyst was excised.

After bone healing, an implant was inserted.

A mandatory anatomo-pathological analysis confirmed the diagnosis.

REFERENCES

1. Aldelaimi TN, Khalil AA. Diagnosis and surgical management of nasopalatine duct cysts. J Craniofac Surg. 2012;23(5):e472-4. doi: 10.1097/SCS.0b013e318258764b.
2. Allard RH. Naso-palatine duct cyst. Int J Oral Surg 1981;10(Suppl 1):131-3.
3. Anneroth G, Hall G, Stuge U. Nasopalatine duct cyst. Int J Oral Maxillofac Surg. 1986 Oct;15(5):572-80.
4. Ben Slama L, Zoghbani A, Hidaya S. [Nasopalatine duct cyst]. Rev Stomatol Chir Maxillofac 2009;110(5):284-6. doi: 10.1016/j.stomax.2009.07.006. Epub 2009 Oct 13.
5. Casado PL, Donner M, Pascarelli B, Derocy C, Duarte ME, Barboza EP. Immediate dental implant failure associated with nasopalatine duct cyst. Implant Dent 2008;17(2):169-75. doi: 10.1097/ID.0b013e3181776c52.
6. Cecchetti F, Ottria L, Bartuli F, Bramanti NE, Arcuri C. Prevalence, distribution, and differential diagnosis of nasopalatine duct cysts. Oral Implantol (Rome). 2012;5(2-3):47-53. Epub 2012 Nov 16.
7. Falci SG, Verli FD, Consolaro A, Santos CR. Morphological characterization of the nasopalatine region in human fetuses and its association to pathologies. J Appl Oral Sci 2013;21(3). pii: S1678-77572013000300250. doi: 10.1590/1679-775720130008.
8. Gnanasekhar JD, Walvekar SV, al-Kandari AM, al-Duwairi Y. Misdiagnosis and mismanagement of a nasopalatine duct cyst and its corrective therapy. A case report. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1995;80(4):465-70.
9. Harris IR, Brown JE. Application of cross-sectional imaging in the differential diagnosis of apical radiolucency. Int Endod J 1997;30(4):288-90.
10. Kramer IRH, Pindbord JJ, Shear M. Histological Classification of Odontogenic Tumours. World Health Organization 1992:7-91.
11. Pevsner PH, Bast WG, Lumerman H, Pivawer G. CT analysis of a complicated nasopalatine duct cyst. N Y State Dent J 2000;66(6):18-20.
12. Scolozzi P, Martinez A, Richter M, Lombardi T. A nasopalatine duct cyst in a 7-year-old child. Pediatr Dent 2008;30(6):530-4.
13. Suter VG, Büttner M, Altermatt HJ, Reichart PA, Bornstein MM. Expansive nasopalatine duct cysts with nasal involvement mimicking apical lesions of endodontic origin: a report of two cases. J Endod 2011;37(9):1320-6. doi: 10.1016/j.joen.2011.05.0 J
14. Suter VG, Sendi P, Reichart PA, Bornstein MM. The nasopalatine duct cyst: an analysis of the relation between clinical symptoms, cyst dimensions, and involvement of neighboring anatomical structures using cone beam computed tomography. Oral Maxillofac Surg 2011;69(10):2595-603. doi: 10.1016/j.joms.2010.11.032. Epub 2011 Mar 12.
15. Suter VG, Altermatt HJ, Voegelin TC, Bornstein MM. [The nasopalatine duct cyst–epidemiology, diagnosis and therapy]. Schweiz Monatsschr Zahnmed. 2007;117(8):824-39.
16. Swanson KS, Kaugars GE, Gunsolley JC. Nasopalatine duct cyst: an analysis of 334 cases. J Oral Maxillofacial Surg 1991;49:268-271. doi: 10.1016/0278-2391(91)90217-A
17. Vasconcelos R, Aguiar MF, Castro W, Ara°jo VC, Mesquita R. Retrospective analysis of 31 cases of nasopalatine duct cyst. Oral Dis 1999;5(4):325-328
18. Velasquez-Smith MT, Mason C, Coonar H, Bennett J. A nasopalatine cyst in an 8-year-old child. Int J Paediatr Dent 1999;9(2):123-7.