CASE REPORT

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Nasal teeth associated with rhinosinusitis*

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SUMMARY

Intranasal ectopic dentition is a rare clinical entity. It may be asymptomatic or can be associated with different symptoms. In 25% of the reported cases an association with rhinosinisitis is suspected. The possible pathophysiology, diagnosis and treatment are discussed. Teeth in the floor of both nasal cavities, associated with chronic rhinosinusitis, are reported for the first time, and a new treatment option of endonasal extraction under microscopic control is presented.

Key words: bilateral ectopic nasal teeth, rhinosinusitis

INTRODUCTION

Nasal teeth are an unusual finding. Until recently only about 50 cases of ectopic nasal dentition have been published. The largest series of 13 consecutive cases was reported by Lee (2001) and another series of 12 cases by Martinson (1972). We are presenting two cases and review of the literature. In both cases, this rare differential diagnosis was not taken into consideration for months to years.

The first case is unique, presenting ectopic teeth in both nasal cavities, associated with chronic rhinosinusitis. Nasal ectopic teeth are examined and extracted using head light or an endoscope (Lee, 2001). In the second case, the use of an operating microscope is reported.

PATIENTS

CASE REPORT 1

A 22-year-old female complained of excessive bilateral nasal watery discharge and bilateral facial pain in the perinasal area with a two-week duration. The patient had recurrent acute rhinosinusitis for five years and has been complaining on episodic bilateral facial pain and headache. Her milk teeth were prematurely replaced with permanent teeth at the age of six. At the age of 11 years both her fourth upper teeth were extirpated for cosmetic reasons.

On examination two yellowish-white masses surrounded by granulation tissue were seen in each inferior nasal meatus. On palpation they were firm and extremely painful even under good local anesthesia. The septum was moderately deviated and the middle turbinates were enlarged. Plane sinus X-rays (Figure 1) showed two bilateral homogeneous high attenuations on the nasal floor. Sinus CT scan (Figure 2) showed mucosal thickening of the ethmoids, bilateral bullose middle conchae and bilateral “rhinoliths”.

During endoscopic operation under general anaesthesia polyps were found within and around the bullous middle turbinates,

Figure 1. Plane X-ray of patient 1. Bilateral high attenuations on the nasal floor.

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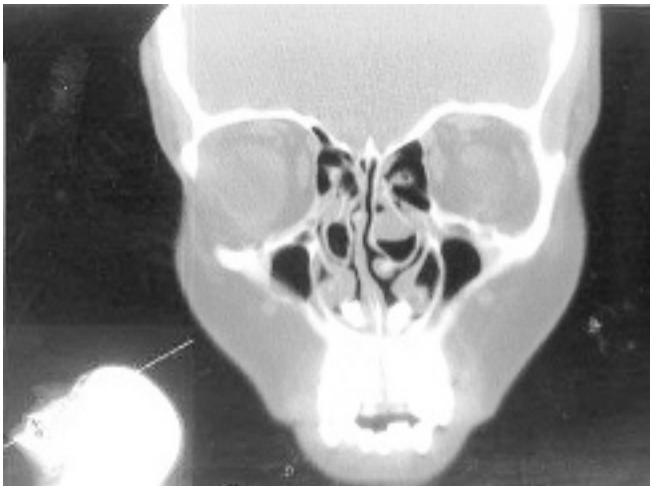

Figure 2. CT scan of patient 1. Bilateral concae bullosae, mucosal thickening and "rhinolits" in the nasal floor.

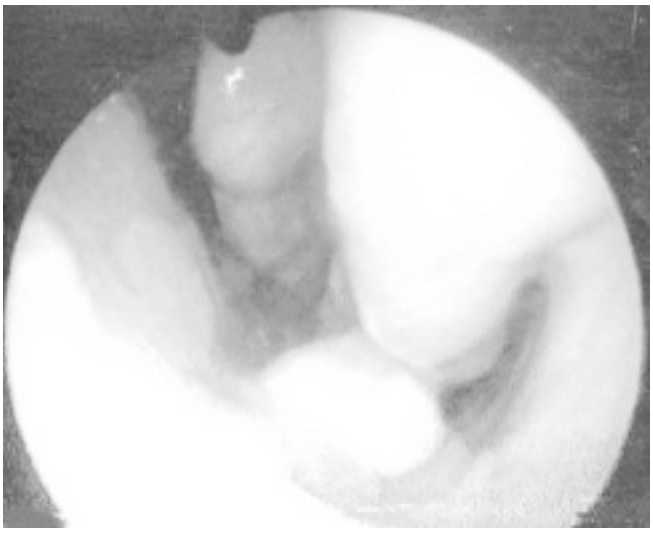

Figure 3. Tooth in the left nasal cavity (intraoperative view).

the anterior ethmoid cells and in both osteomeatal complexes. Teeth (Figure 3) were separated and easily removed from the nasal floor on each side.

CASE REPORT 2

A 36-year-old female complained of unilateral nasal obstruction and discharge for several months. She was otherwise healthy. Rhinoscopy revealed a grey mass, firmly attached to the posterior part of the nasal septum, surrounded by granulation tissue that looked like a rhinolith. Dentition was normal.

CT scan (Figure 4) showed a hyperdense mass in the right inferior meatus. During the operation under general anesthesia, a nasal tooth was removed (Figure 5) with the use of an operating microscope. The postoperative period for both patients was uncomplicated, and on follow up they had fully recovered.

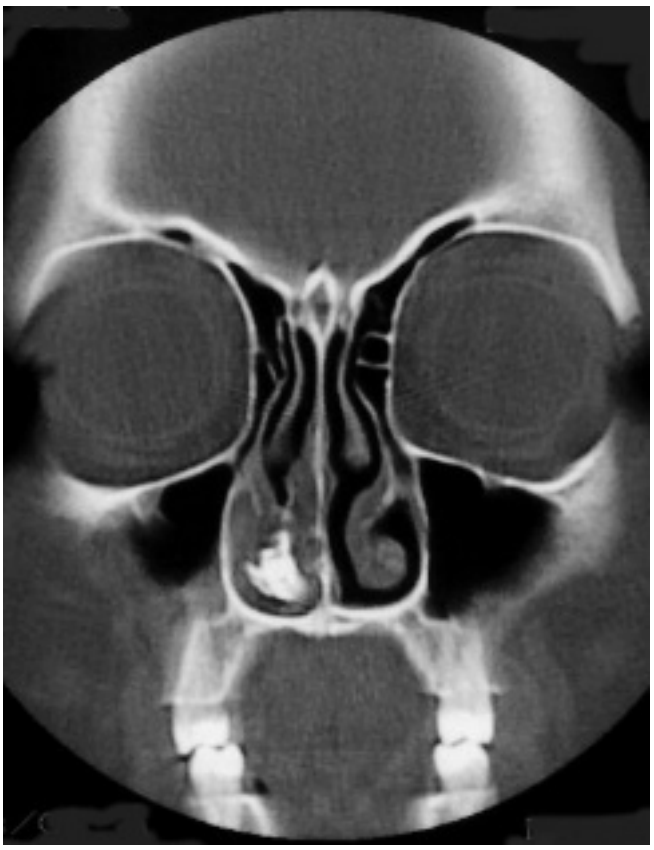

Figure 4. CT scan of patient 2. Tooth in the right nasal cavity.

Figure 5. Patient 2. Tooth with surrounding rhinolith after removal.

DISCUSSION

Nasal teeth are rare. There are only 50 reported cases of intranasal ectopic dentition in the literature (Gupta and Shah, 2001). The location inside the nasal cavity may be different: inferior and medial meatus, subperichondrially in the nasal septum and within the inferior turbinate (Martinson, 1972; Lee, 2001). Other non-nasal sites may be the ethmoid sinus (Weber et al., 1993), maxillary sinus, chin, palate, mandibular condile, coronoid process, orbit and skin (Smith et al., 1979; el-Sayed, 1995; Yehung and Lee, 1996). The incidence of the extranumerary teeth is up to 1% of the population (Thalwey and Ferrier, 1997). Ectopic nasal dentition is obviously very rare, but the incidence was never documented. Medeiros et al. (2000) reported an average 0.48% prevalence of intranasal teeth in children with cleft lip and palate.

The etiology of ectopic nasal teeth is unknown. There are many different explanations. According to one of them such a tooth develops from a third tooth bed that arises from the dental lamina near the permanent tooth bud, or, possibly, from splitting of the permanent bud itself (Moreano et al., 1998). According to the same authors the other possibility is that their development is a reversion to the extinct primate dentition (primates have three pairs of incisors).

Other etiopathogenic factors may include genetic predisposition, rhino- or odontogenic infections and post-traumatic displacement. Cyst and cleft palate may also affect this entity, due to incomplete union of the embryonic process or even due to pushing up while unrecognized during surgical repair of an alveolar cleft. Other associated developmental disturbances are obstruction to eruption secondary to crowded dentition, retained primary teeth or exceptionally dense bone (Smith et al,. 1979; Moreano et al., 1998). In our first patient crowded dentition can be proposed as a contributing factor.

The differential diagnosis includes rhinolith, foreign body, granulomatous inflammatory diseases such as found in tuberculosis, syphilis, fungal infection, calcified polyps and dermoids, benign tumors (haemangioma, osteoma) or malignant tumors (chondrosarcoma, osteosarcoma) (Martinson, 1972; Lee, 2001; Chen et al., 2002). Ogisi and Odita (1988) published a very rare association of ectopic teeth (nose and palate) with squamous cell carcinoma of the palate in a 12-year-old Nigerian boy.

Nasal ectopic dentition may be asymptomatic or may be associated with nasal pain, nasal obstruction, epistaxis, headache, nasal discharge, mild fever, crusting of nasal mucosa, localized ulceration, external deviation of the nose, nasal septal abscess, nasal-oral fistulas, signs of maxillary or pansinusitis, epiphora and recurrent dacriocystitis and even speech problems (Martinson, 1972; Srivastava et al., 1977; Pracy et al., 1992; Weber et al., 1993; Moreno et al., 1998; Lee, 2001; Chen et al., 2002; Kim et al., 2003;).

The association of rhinosinusitis with ectopic intraantral teeth has been reported in the literature (Goh, 2001) as well as with tooth occluding the osteomeatal complex (Hasbini et al., 2000). In these cases the etiological relationships are well recognized. However, little is written about the association of an ectopic tooth in the nasal cavity with chronic rhinosinusitis.

Analysis of the literature has revealed 13 cases of intranasal teeth associated with chronic rhinosinusitis. Confirmation of chronic rhinosinusitis was sometimes difficult, due to lack of anamnestic information. An association with chronic rhinosinusitis was therefore established if there was evidence of purulent nasal discharge of more than 3 months, or if there were suggestive radiological findings such as opacification of one or more the sinuses or mucosal swelling. This analysis has demonstrated that 25% of the reported patients with intranasal ectopic dentition have röntgenological signs suggestive of chronic rhinosinusitis. In the majority of the symptomatic patients complete recovery was obtained after teeth removal. In some cases therefore, ectopic intranasal dentition can play an etiologic role in the development of chronic rhinosinusitis.

It is doubtful that in the first patient the ectopic nasal teeth played a pathophysiologic role in the development of the chronic rhinosinusitis. Anatomically all the sinuses were correctly developed. The bilateral conchae bullosa is a frequent finding in normal subjects, and no reports of ectopic teeth associated concha bullosa have been found in the reviewed literature. Perhaps the exceptional findings reflect disembryogenesis, which may play a causative factor of the rhinosinusitis.

Interestingly, only one published report on ectopic nasal teeth in animals was found (Priddy et al., 2001), hence no animal model is available for this condition.

In most reported cases only one tooth is found in the nasal cavity (as in our second patient), but multiple ectopic nasal teeth were also reported. Alexandrakis (2000) reported two teeth in the right inferior meatus compressing the nasolacrimal duct. Teeth in both nasal cavities were reported in a patient with cleft palate (Srivastava et al., 1997), and Martinson and Cockshott (1972) have reported a very unusual case of 5 nasal teeth in a 6-year-old boy: 4 subperiostally in the nasal septum (2 in each side of the septum) and 1 in an inferior turbinate. In our first patient, who had a normal palate, we found two teeth, one in each nasal cavity. To the best of our knowledge this is unique.

Radiological examination (preferably CT scan) may be helpful in preoperative assessment of the patients, assisting in differential diagnosis and in the evaluation of the exact depth of the eruption site (Chen et al., 2002).

In some cases, where a tooth lies deep inside the underlining bone, removal may require special equipment (i.e. drill). Endoscopic or microscopic approaches can both be employed, as demonstrated in the present report. Surgical removal of ectopic intranasal dentition is recommended by most authors, even in asymptomatic patients, because of its potential associated morbidity. If the tooth is not removed, close clinical follow-up is recommended (Thalwey et al., 1997; Medeiros et al., 2000; Kim et al., 2003).

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ERRATUM

In the article "Nasal polyps and middle turbinates epithelial cells sensitivity to amphotericin B" by L. Jornot, T. Rochat, and J.S. Lacroix that was published in Rhinology 41, 201-205, 2003, a low-resolution picture of Figure 2 was printed. The article also did not contain references throughout the text.

Below is a high resolution of Figure 2. Subscribers to Rhinology have access to a new pdf file of this article containing the revised text on our website: www.rhinologyjournal.com/article/member_download.php?rtcl_id=&inhoud_filenaam=jornot.pdf

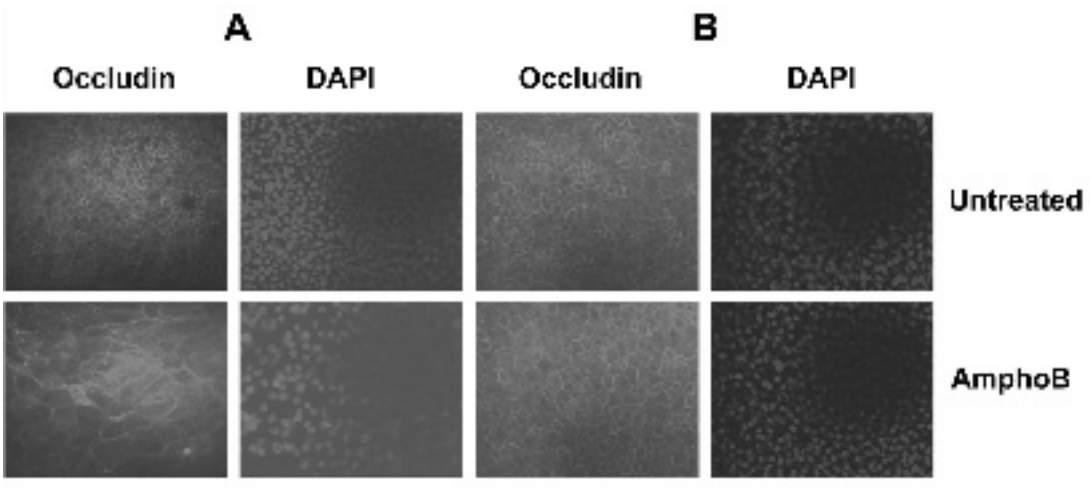

Figure 2. Expression of the tight junction protein occludin in untreated and AmphoB-treated nasal epithelial cells. Immunocytochemistry localization of occludin was performed in cell cultures derived from polyps (A) or middle turbinates (B), left untreated or treated with AmphoB for one month. Cell nuclei were stained with DAPI to assess the number of adherent cells.