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DIAGNOSIS AND TREATMENT OF NASOPALATINE DUCT CYST – CASE REPORT

ДИЈАГНОЗА И ТРЕТМАН НА НАЗОПАЛАТИНАЛНА ДУКТАЛНА ЦИСТА – ПРИКАЗ НА СЛУЧАЈ

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Abstract

Nasopalatine duct cyst, called the incisive canal cyst, described in literature as a cystis canalis nasopalatine or cystis canalis incisive, is the most common non-odontogenic cyst occurring in the oral cavity. It develops in the midline palate around the incisive foramen from debris of nasopalatine duct's epithelium. The etiology is unknown, although it is believed that some of the irritants may have an impact on its development. Cyst develops slowly destroying the alveolar bone of the maxilla. Typically, the process is asymptomatic and is detected accidentally on radiograph. Clinical diagnosis is made due to X-rays, but only histopathology can confirm the results. The treatment of choice is total enucleation of pathological changes. The following article presents a case of nasopalatine duct cysts, which is diagnosed and treated at the Faculty of Dentistry in Skopje. The case is characteristic regarding the symptomatology, as well as the need of differentiation from other pathological changes within the region, as well as the need for proper treatment. **Key words:** Nasopalatine duct cyst, non-odontogenic cyst, incisive canal cyst.

Апстракт

Назопалатиналната циста, наречена уште и циста на инцизивниот канал, опишана во литературата како cystis canalis nasopalatine или cystis canalis incisive, е најчестата неодонтогена циста во оралната празнина. Се развива во средишната линија на палатумот околу инцизивниот отвор од епителиумот на назопалатиналниот канал. Етиолигијата за нејзиното создавање е непозната, но се смета дека одредени иритативни фактори можат да имаат влијание врз нејзиниот развиток. Цистата се развива многу бавно вршејќи коскена деструкција на околната коска на максилата. Типично, процесот е асимптоматски и најчесто се детектира случајно на радиографска снимка, а клиничката дијагноза се потврдува со хистопатолошка анализа. Терапијата е хируршка со целосна енуклеација на цистичната промена. Авторите презентираат клинички случај на назопалатинална дуктална циста, која е дијагностицирана и хируршки третирана на Стоматолошкиот факултет во Скопје. Случајот е карактеристичен во однос на симптоматологијата, како и потребата од диференцирање од другите патолошки промени кои се јавуваат во таа регија и потребата од соодветен третман. **Клучни зборови:** Назопалатинална дуктална циста, неодонтогена циста, циста на инцизивен канал.

Introduction

Nasopalatine duct cyst is the most common non – odontogenic cyst occuring in oral cavity. It was first described by Meyer in 1914[1, 2]. In the past, known as the fissured cyst, now according to the WHO classification is defined as a non – odontogenic, developmental, epithelial cyst of maxilla[3]. In most of cases, it develops in the midline of the palate near the incisive foramen[4]. In normal development, the nasopalatine duct canal is converted into incisive canal by the disappearance of epithelium. Other cells in the form of epithelial cord can initiate the development of cysts. Mostly it is located in palatal part of canal or superficially[5]. Depending on whether the cyst was caused by palatal or nasal part of the canal, it is lined by stratified squamous or ciliated epithelium[6]. If it appears above, within the nasal part of the canal, cilliary respiratory epithelium can even be observed[3]. It has a capsule built of dense fibrous tissue containing scattered chronic inflammatory cells[5]. Nasopalatine duct cyst is rare and occurs in 1% of the population. Observations show that it is usually detected among people between 4th and 6th decade of life, although there were cases among children up to 8 years old. It is three times more common among men than women[3–5, 7]. It occurs in both human races: white and black[8, 9]. The etiology of the cyst is unknown, although it is suspected that its development may contribute to nasopalatine duct infections, or retention of mucus[3–5, 10]. Of all the factors most likely theory is spontaneous cystic degeneration of the nasopalatine duct remains. This duct, in the fetal period, is a broad connection between mouth and nose. In the course of normal development, shortly after birth, it becomes obliterated and atrophies. Among some peo-

ple, however, it does not disappear completely and its remnants, in the form of epithelial cord, could lead to the development of cysts in the incisive canal. However, the cause of this abnormal development is still unknown[8]. Some authors suggest here the similarity to the lower mammals, in which inside the nasopalatine canal is penetrable by air „nasopalatine duct", which is an auxiliary olfactory organ, called Jacobson's organ[8]. Among the suggested causes of incisive canal cysts formation are also some genetic factors. However, the literature does not describe a lot of evidence for an unambiguous confirmation of any of the hypotheses[3,5]. Cyst develops slowly leading to loss of bone in the maxilla. Most of its development is asymptomatic and is detected incidentally on radiograph. It is observed within the median line of the palate as an oval or heart-shaped radiolucency[3, 5, 13, 14]. If the symptoms appear, swelling in the median line of the palate's front part is the most common[5]. It may also manifest itself on the labial side of the alveolar process of the maxilla, causing rarely facial asymmetry[3, 5, 15]. In some cases, pain may occur as a result of pressure of nasopalatine nerve, reported primarily by people using prosthesis or as a result of palpation examination of incisive papilla's area.

The pain can be caused also by the superinfection. The more advanced change is, the symptoms become more pronounced. Also it is observed that symptoms appear earlier when the cyst is located caudal[5]. Surgical total enucleation is the recommended treatment with pathological findings and shows a very low rate of recurrence[5, 12, 13]. Among all types of cysts diagnosed in dental surgery, a nasopalatine duct cyst is rare. Since 1960, the English-language literature has published only 468 case reports[8]. The aim of our study is to present one case documented by the University Dental Clinic „St. Pantheleimon" - Skopje, in 2019.

Case Report

A 24-year-old male patient was referred by his general dentist to the department of oral surgery, our clinic, complaining to pain in the area of the first maxillary incisors, followed by a panoramic x-ray image showing translucency in the region of the apexes of the central maxillary incisors. (Fig. 1)

The main subjective problem reported by the patient is swelling and pain in the midline, on the inside of the upper jaw. Clinical inspection and examination of the region detects swelling in anterior third of medial line of palatum. (Fig. 2)

Palpation in the area of the incisal papilla produces a painful sensation and a strongly elastic fluctuating prominence that is easily elevated above the level of the gingival mucosa. The panoramic x-ray image shows intact central incisors, with no previous carious lesions and no previous conservative or endodontic treatments (Fig.1). On a vertical percussion, there is a mild painful sensation on them. A vitality test of the central and lateral incisors was performed bilaterally, with a positive result.

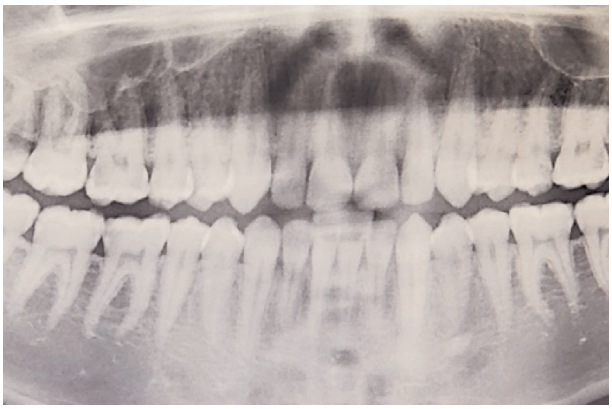

Figure 1. *Panoramic x-ray*

Figure 2. *Intraoral view of the palatum*

By the clinical examinations and differential diagnostic pathways, panoramic X-rays revealed that the diagnosis was a nasopalatinal ductal cyst. The patient is referred for further 3D X-ray imaging, with the aim of precise and accurate localization of the cystic change. We performed a drainage procedure, which produces purulent content and a rubber drain was installed. The 3D x-ray recording clearly reveals a well-restricted radiolucent bone change of non-odontogenic origin, preserving a well-preserved lamellar bone around the roots of all teeth in the frontal region (Fig. 3, 4 and 5). Dimensions of the translucency are 14 mm in craniocaudal direction and 11 mm in transversal direction. After all clinical and paraclinical investigations, the previous differential diagnostic finding confirms that it is a nasopalatinal ductal cyst and that the patient is in need of further surgical treatment, which would result in total enucleation of the cyst.

On several subsequent visits, the pathologically changed tissue was washed and cleaned with potassium permanganate and the rubber drain was changed several

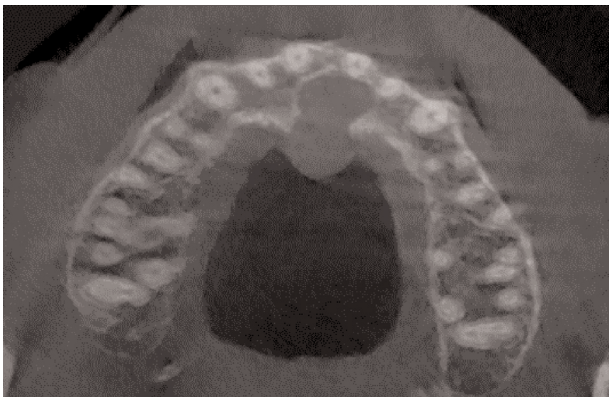

Figure 3. *3D view of the cyst*

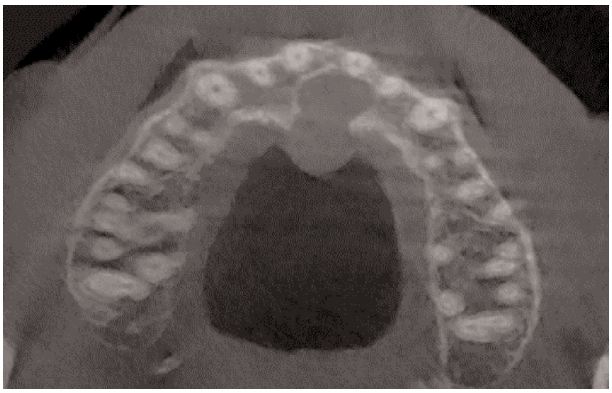

Figure 4. *Transversal view of the cyst*

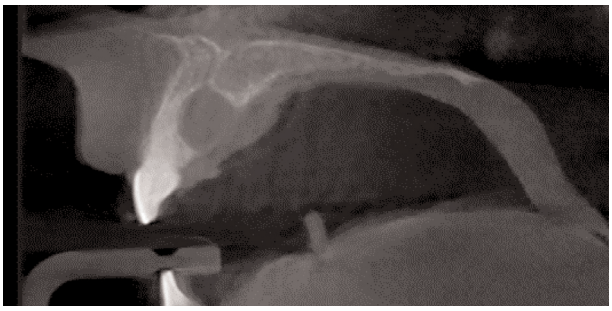

Figure 5. *Sagital view of the cyst*

times every 24 hours. When the subjective problems of the patient disappeared and the purulent content was evacuated, we had complete cystic enuclation. Nasopalatinal ductal nerve block anesthesia was applied, Scandonest 3% in nasopalatine duct and plexus anesthesia in the vestibular support of the upper incisors. We made a marginal, palatal incision in the intercuspid area and raised a mucoperiostal flap with careful prevention of nerve and vascular contents of the nasopalatinal canal (Fig. 6).

After total cyst enucleation, the mucoperiostal flap tissue was returned to its original position and sutured (Fig. 7). A fully enucleated specimen of the cyst is given for

Figure 6. *Intraoral view of the lesion*

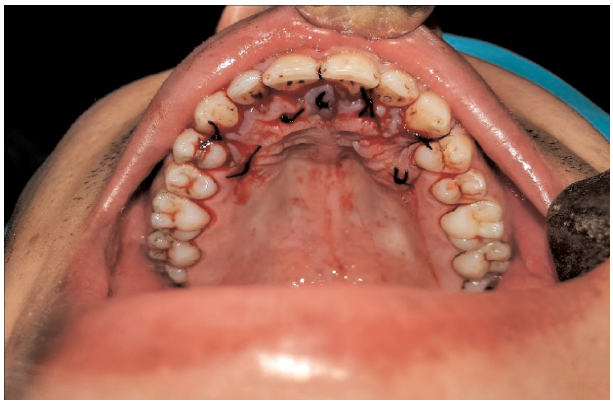

Figure 7. *Postoperative sutured mucoperiostal flap*

pathohistological examination. Postoperatively, oral antibiotic therapy Amoxicillin a 1000 mg twice a day is prescribed, for 5 days after the surgery treatment. In the next few days the patient is followed and advised for rinses with sodium chloride 0.9% for several times in a day.

According to the macroscopic finding of the pathohistological examination, a soft tissue fragment was presented with an aspect of a cyst wall of 0.1 cm thickness. The luminal and outer surface of the cyst wall are smooth, and the overall dimensions of the fragment are 1 cm x 0.7 cm. All material is embedded in paraffin block for analysis.

According to the microscopic finding, the analysis showed connective tissue with areas of edema that contained a cystic formation coated with prominent squamous epithelium without any atypical cells signs. Peripheral to the epithelium is a rich mononuclear inflammatory infiltrate and local areas of fresh bleeding.

Discussion

Diagnosing and differentiation of the nasopalatine duct cyst has to be made very carefully before making the final diagnosis. A presumptive diagnosis suggested

on the base of the anamnesis and precise clinical examination has to be completed with the radiological examination in minimum two projections: periapical and occlusal. However, the only certain confirmation is a result of the histopathological examination. During the clinical examination, the differential diagnosing is very important. Mainly the nasopalatine duct cyst has to be differenced with the radicular cyst to avoid unnecessary endodontic treatment of vital and healthy teeth[5]. For this purpose, the pulp vitality test, the percussion test and the analysis of shape and width of the periodontal space has to be performed[11]. In case of the nasopalatine duct cyst, pulp of the neighboring teeth remains vital and the lamina dura of the periodontal fissure does not lose continuity. All the mentioned tests and analyses have to be performed when periapical granuloma of the upper incisors is suspected. It is very important according to the modern endodontics, which says that most of the periapical granulomas remain 'not infected' because they are caused by the bacteria which are present only in the root canal. That fact implicates the treatment of these lesions, which is based on the antiseptic endodontic treatment without obligatory surgical removal of the periapical granulomas. During the interpretation of the X-ray, those pathological periapical lesions are most frequently suggested as the presumptive diagnosis. In differential diagnosis the rare lesion – median palatal cyst, similar on the X-rays, should be also concerned. However, its etiology is connected with an inappropriate fusion of the maxillary processes and with an injection of the epithelial cells between them[16]. As far as Francoli and Torres claim, diagnostic problems appear also during examination of smaller lesions (average size of the nasopalatine duct cyst is between 6 mm to 17 mm)[3, 8] because they can be similar to anatomical structures like the incisive foramen or widen to 6–8 mm incisive canal. On the other hand, the cyst can reach the size overcrossing 50 mm[8, 11]. The differential diagnosis should concern the supernumerary tooth appearing in this area – the mesiodens in the follicular cyst and also it should concern the primary cyst, the giant-cell granuloma, the osteitis with the palatal fistula and also naso – palatine and palatal – sinus connections[3]. As soon as the final diagnosis of the nasopalatine duct cyst is made, the lesion has to be surgically removed, as the literature recommends, not only because it is destroying the bone, but also a few malignant transformations are known[3]. In English literature, there are a few cases of the squamous cell carcinoma which have developed as a result of the metaplasia of the epithelium which lines the cyst[17]. Gardner observed characteristic symptoms of the cancer developing from the cyst. As an example, it can be a dynamic growth of the lesion, also a resorption of the roots which can be observed on the X-rays and changes in the sensitivity to touch of the upper lip[18]. According to literature, a marsupialization is also a possible alternative treatment for some patients[5, 10]. It is recommended in the cases when the capsule of the cyst shows adhesions with the surrounding area which make it difficult to enucleate. Rounded incision should be made on the biggest circumference and then the cyst's lower wall with surrounding mucosa has to be removed. The procedure should be finished with inserting the sutures between the lining of the cyst and the mucosa of the oral cavity. According to some theories, if the lesion is asymptomatic and does not reach larger sizes, the surgical treatment is not necessary. But they also emphasize that the cyst has to be removed before prosthetic treatment in this area, because the chronic irritation can lead to inflammatory reaction. Considering all those facts, it seems that surgical treatment is the best way of treatment of the nasopalatine duct cyst[3, 16].

A total enucleation should be a surgical treatment of choice because there are some evidenced cases of recurrence reaching from 0% up to 11%. According to Kimberly, in all collected by him 334 cases, only 7 recurrences were observed (2%). Hedin recommends regular control visits, including X-ray examination and pulp vitality tests of neighboring teeth, after the surgery during 3 years[2, 19]. As presented in this study case, the four-week observation period after surgery was performed and then the treatment was considered as completed. However, the total enucleation was performed in our case, the patient was informed that he was obligated to self-control and if any symptoms of recurrence would appear he had to come to clinic as soon as possible. Another rare complication after a surgery which can be observed only in 10% of cases is the paresthesia of the frontal part of the palate. It is caused by removing the part of the cyst's wall which can be connected with the endings of the naso-palatine nerve[3]. Although this does not let us to forget about the proper examination and also if it is necessary to use all the diagnostic sources and make a differential diagnosis to start in the right time, a good treatment to avoid dangerous complications.

Reference

1. Meyer A.W. A unique supernumerary paranasal sinus directly above the superior incisors. J. Anat. 1914; (48): 118–129.
2. Terry B.R., Bolanos O.R. A diagnostic case involving an incisive canal cyst. J. Endod. 1989, 11, 559–562.
3. Francoli J.E., Marques N.A., Aytes l.B., Escoda C.G. Nasopalatine duct cyst: Report of 22 cases and review of literature. Med. Oral Patol. Oral Cir. Bucal. 2008, 13, 438–443.
4. Hegde R.J., Shetty R. Nasopalatine duct cyst. J. indian Soc. Pedod. Prev. Dent. 2006, 24, 31–32.
5. Cicciu M., Grossi G.B., Borgonovo S., Santoro G., Pallotti F., Maiorana C. Rare bilateral nasopalatine duct cysts: A case Report. Open Dent. J. 2010, 4, 8–12.

6. Panaś M.: Torbiele. In: Chirurgia szczękowo-twarzowa. eds. Bartkowski S.B., collegium Medicum UJ ages, Kraków 1996, 155–168.
7. Neville B.W., Damm D.D., Brock T. Odontogenic keratocysts of the midline maxillary region. J. Oral Maxillofac. surg. 1997, 55, 340–344.
8. Torres l.M., Benito J.I., Morais D, Fernandez A. Nasopalatine duct cyst: case report. acta Otorrinolaringol. esp. [8] 2008, 59, 250–251.
9. Righini C.A., Boubagra K., Verougstreate G., Reyt E. Kyste du canal nasopalatin. a propos de 4 cas, revue de la literature. ann. Otolaryngol. chir. cervicofac. 2004, 121, 115–119.
10. Swanson K.s., Kaugars G.E., Gunsolley J.C. Nasopalatine duct cyst: an analysis of 334 cases. J. Oral Maxillofac. surg. 1991, 49, 266–271.
11. Gnanasekhar J.D., Walvekar S.V., Al-Kandari A.M., Al-Duwairi Y. Misdiagnosis and mismanagement of a nasopalatine duct cyst and its corrective therapy. Oral surg. Oral Med. Oral Pathol. Radiol. endod. 1995, 80, 465–470.
12. Hisatomi M., Asaumi J., Konouchi H., Matsuzaki H., Kishi K. MR imaging of nasopalatine duct cysts. eur. J. Radiol. 2001, 39, 73–76.
13. Tanaka S., Iida S., Murakami S., Kishino M., Yamada C., Okura M. Extensive nasopaltine duct cyst causing nasolabial protrusion. Oral surg. Oral Med. Oral Pathol. Radiol. endod. 2008, 106, 46–50.
14. Elliott K.A., Franzese C.B., Pitman K.T. Diagnosis and surgical management of nasopaltine duct cysts. laryngoscope 2004, 114, 1336–1340.
15. Lipski M., Gala A., Smółka B. Chirurgiczno-protetyczne leczenie torbieli jamy ustnej. Poradnik stomatol. 2005, 6, 5, 40–46.
16. Allmendinger A., Gabe M., Destian S. Median Palatine cyst. J. Radiol. 2009, 7, 7–10.
17. 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. 1966, 54, 112–115.
18. Gardner A. The odontogenic cyst as a potential carcinoma: A clinicopathologic appraisal. J. am. Dent. assoc. 1969, 78, 746–755.
19. Hedin M., Klamfeldt A., Persson G. Surgical treatment of nasopalatine duct cysts. int. Oral surg. 1978, 7, 427–433.