

CLINICAL CASE

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Nasopalatine Duct Cyst – Case Reports

Torbiel przewodu nosowo-podniebiennego – opis przypadków

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Abstract

Nasopalatine duct cyst, called the incisive canal cyst, described in literature as a *cystis canalis nasopalatini*, *cystis canalis incisivi*, is the most common non-odontogenic cyst occurring in the oral cavity. It develops in the mid-line 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 description of three cases of nasopalatine duct cysts, which were diagnosed and treated at the Institute of Dentistry, Poznań University of Medical Sciences in the years 1996–2009. These cases are characteristic for typical exponents reported for incisive cyst, point at the need to diversify to other changes within the region and the need for proper treatment (**Dent. Med. Probl.** 2010, 47, 4, 508–512).

Key words: nasopalatine duct cyst, non-odontogenic cyst, incisive canal cyst.

Streszczenie

Torbiel przewodu nosowo-podniebiennego, zwana także torbielą kanału przysiecznego, opisywana w literaturze jako *cystis canalis nasopalatini*, *cystis canalis incisivi*, jest najczęstszą niezębopochodną torbielą występującą w obrębie jamy ustnej. Rozwija się w linii środkowej podniebienia, w okolicy otworu przysiecznego, z resztek nabłonka przewodu nosowo-podniebiennego. Etiologia jest nieznana, choć uważa się, iż pewne czynniki drażniące mogą mieć wpływ na jej rozwój. Torbiel rozwija się powoli, niszcząc kość wyrostka zębodołowego szczęki. Zwykle jej przebieg jest bezobjawowy i jest wykrywana przypadkowo na zdjęciu radiologicznym. Rozpoznanie kliniczne często ustala się na podstawie obrazu radiologicznego, a jedynym potwierdzeniem są wyniki badania histopatologicznego. Leczeniem z wyboru jest całkowite wyłuszczenie patologicznej zmiany. Praca przedstawia opis 3 przypadków torbieli przewodu nosowo-podniebiennego, które zostały rozpoznane i leczone w Instytucie Stomatologii Uniwersytetu Medycznego w Poznaniu w latach 1996–2009. Przypadki te, charakteryzujące się typowymi wykładnikami opisywanymi dla torbieli kanału nosowo-podniebiennego, zwracają uwagę na potrzebę różnicowania z innymi zmianami w obrębie tego regionu oraz na konieczność podjęcia prawidłowego leczenia (**Dent. Med. Probl.** 2010, 47, 4, 508–512).

Słowa kluczowe: torbiel przewodu nosowo-podniebiennego, torbiel niezębopochodna, torbiel kanału przysiecznego.

Nasopalatine duct cyst is the most common non – odontogenic cyst occurring 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 epi-

thelium [6]. If it appears above, within the nasal part of the canal, ciliary 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 people, 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 three cases documented by the Institute of Dentistry, University of Medical Sciences in the years 1996–2009.

Case reports

Case 1

A 69-year-old man reported in 2004 because for the pain of the left maxillary central incisor. Patient’s main complaint was discomfort caused by hot and cold food. Clinical examination detected a small protuberance in the median line of palate. The medical records showed that there was deep caries in this tooth one year before. Biological treatment of pulp was used as intermediate cover with calcium hydroxide: Life. This information and symptoms described in physical examination suggested that endodontic treatment should be given. Pulp vitality test was performed to confirm diagnosis. The test result was positive. The patient was referred for X-rays, which revealed a well-circumscribed radiolucency in the median line of palate (Fig. 1). Analysis of periodontal space shape aroused suspicion that the reason of this change was not odontogenic. After an examination by a dental surgeon, a presumptive diagnosis was: nasopalatine duct cyst. During the visit, anesthesia with Scandonest of 3% (2 amp.) was made. The cyst was incised, purulent substance was evacuated and the rubber drain was placed. The doctor prescribed antibiotic Dalacin C (300 mg 3 × 1 caps) and the painkiller Ketonal Forte (100 mg 2 × 1 tabl.).



Fig. 1. Case 1. Well-circumscribed radiolucency in the median line of palate

Ryc. 1.
Przypadek 1.
Dobrze
odgraniczone
przejaśnienie
w linii
pośrodkowej
podniebienia

On the next several visits, the change was rinsed by the solution of potassium permanganate and the drain was exchanged. When the pain disappeared and purulent substance was evacuated, the doctor performed the cyst enucleation. Infiltration anesthesia was made with Ubistesin 4%. Marginal incision was made and mucoperiosteal flap was prepared. After removal of the cyst sutures Safil 4.0 were laid. The uptaken samples were given for a histopathological examination, the result described the change as: „Fragments of fibrous wall of the cyst with markers of increased, chronic inflammation, covered with paraepidermal epithelium with the features of focal dysplasia”. Thus, it is confirmed that microscopic image corresponds to a clinical diagnosis. After removal of sutures and the four-week observation period after surgery, the treatment was considered as completed.

Case 2

A 44-year-old man came to the Department of Oral Surgery in year 2000 in order to extract the right third molar in mandible. His major complaint was an intense and lounded pain. Based on interview and clinical examination, the difficult eruption of wisdom tooth 48 with pericoronal inflammation (*dentitio difficile dentis sapienti inferiori dextra, pericoronitis*) was diagnosed. In order to illustrate the exact location and construction of the tooth roots and their relation to the lower alveolar nerve canal patient was referred to an X-ray laboratory for pantomographic radiograph. On the basis of radiograph, extraction was scheduled at the next visit as primarily an acute pericoronal inflammation should have been eliminated by rinsing solution of potassium permanganate and inserting a gauze with kamfenol and iodoform. Examiner, however concerned radiolucency on the radiograph around the roots of maxillary central incisors. To exclude a possible artifact occlusal radiograph was commissioned, which confirmed the presence of the lesion (Fig. 2). In the front part of palate, further tests were performed to rule out causes of odontogenic. Results of the pulp testing of the central incisors were consistent with vital pulp, which confirmed nonodontogenic origin of cyst. The patient was scheduled for surgery to excise the pathological lesion. Because of the hypertension, the patient was anesthetized with Scandonest 3% (2 amp.). After removal of the cyst, material has been taken for histopathological examination. The result is described as: “Fragments of fibrous connective tissue covered by (pavement) cuboidal dual-layer epithelium (cyst wall). This was consistent with a nasopalatine duct cyst diagnosis”. After removal of sutures, and the further



Fig. 2. Case 2. Occlusal radiograph. Radiolucency around the roots of maxillary central incisors

Ryc. 2. Przypadek 2. Zdjęcie zgryzu. Przejaśnienie w okolicy korzeni środkowych zębów siecznych w szczęcie

three-week period of clinical observation of treatment due to the radicalness, performed surgery was considered terminated.

Case 3

A 60-year-old woman was referred to Department of Oral Surgery in 2009 because of swelling in the palate. The region of incisal papilla was tender to palpation. The mucous membrane of the normal color did not report the characteristics of inflammation. About 5 months earlier, the patient noticed a soft swelling in the palate that gradually become larger. The clinical examination estimated the size of swelling at 3 cm in diameter. On the basis of radiograph (Fig. 3) and results of the pulp testing, the palatal lesion was diagnosed as a naso-



Fig. 3. Case 3. Radiolucence around the roots of maxillary central incisors

Ryc. 3. Przypadek 3. Przejaśnienie w okolicy szczytów korzeni przyśrodkowych zębów siecznych górnych

palatine duct cyst. The patient was scheduled for surgery. Palatal anesthesia was obtained using 4% Ubistesin (2 amp.). The lesion was enucleated in total and the specimen sent for histological examination, which described it as: "Cyst covered by stratified squamous non-cornifying epithelium. In the cyst wall, there was non-specific inflammatory infiltration. Microscopic image corresponds to a clinical diagnosis: nasopalatine duct cyst". At the control visit, three weeks after sutures removal, treatment was considered as completed.

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 differentiated 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 has to be performed when periapical granuloma of the upper incisors is suspected. It is very important accordingly 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 this 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. Situation like that is presented in described case 1.

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 otitis 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 the 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 also they 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 recurrence 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]. In presented in this study cases, 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 all cases, the patients were informed that they

were obligated to self-control and if any symptoms of recurrence would appear they 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].

Presented in this study cases confirm that the

nasopalatine duct cyst can be asymptomatic (case 2) as well as can have typical symptoms described in the literature. There were only 3 patients with diagnosed nasopalatine duct cyst during 14 years, what shows how rarely this pathology appears. 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.

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