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MUCOCELES OF MINOR SALIVARY GLANDS IN CHILDREN. OWN CLINICAL OBSERVATIONS

TORBIELE ŚLUZOWE MAŁYCH GRUCZOŁÓW ŚLINOWYCH U DZIECI. OBSERWACJE WŁASNE

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Abstract

Introduction: Mucoceles are benign lesions associated with the pathology of the oral mucosa of minor salivary glands. Two types of cysts are distinguished depending on their pathogenesis. Most often they occur as a result of mechanical trauma and mucus extravasation into tissues or obstruction of the gland ducts.

Aim: The aim of the study was to present our own experiences regarding mucoceles of minor salivary glands in the oral cavity taking into account how frequently the individual types of cysts occur in children.

Material and methods: The research was carried out based on medical files from the years 2005-2015. These were: medical case records, operating books and the medical registry of patients treated at the Clinic of Maxillofacial Surgery, Frederic Chopin Clinical Regional Hospital in Rzeszów. In that period 64 children and teenagers, 28 girls and 36 boys were treated. What was considered was the age and gender of the patients, the reason for their appointment with a doctor, the location, size and histopathological type of the cysts, as well as the course and results of the diagnostic and therapeutic process.

Results: In the group analyzed, the reasons for referral to the Clinic were: in 25 patients accidental ascertainment of a non-symptomatic tumor in the oral cavity during examination by a dentist, pediatrician or laryngologist which had not caused any discomfort to the children; in 13 patients concern had been raised by a gradually increasing tumor; in 18 cases there was an increased tissue tension surrounding the tumor, while in 3 children red oedema was observed in the oral cavity (suspicion of abscess).

The most frequent mucocele location was the lower lip (34 children). The most frequent size was 2.1-3 cm (28 children). The most frequent histological type was MEP. All the patients were treated at the Clinic in the one-day surgery mode, with good outcome.

Conclusions: Mucocele ascertainment in children's oral cavity could be made accidentally in routine pediatric examination, therefore it is necessary to extend pediatricians' knowledge about small salivary gland mucoceles. The most frequent type of MEP could be related to different types of trauma in the oral mucose.

Key words: Mucus Extravasation Phenomenon (MEP) Mucoceles, mucus retention in minor mucous glands, diagnosis, surgery

Streszczenie

Wstęp: Mucocele należą do łagodnych zmian związanych z patologią małych gruczołów ślinowych wyścielających błonę śluzową jamy ustnej. Wyróżnia się dwa typy tych torbieli. Najczęściej powstają w wyniku urazu mechanicznego i wynaczynienia śluzu do tkanek lub na skutek zaczopowania przewodów gruczołu ślinowego.

Cel: Celem pracy było przedstawienie własnych spostrzeżeń dotyczących torbieli śluzowych gruczołów ślinowych jamy ustnej z uwzględnieniem typów torbieli występujących u dzieci.

Materiał i metody: Badania przeprowadzono na podstawie dokumentacji medycznej z lat 2005-2015. Obejmowała ona historie chorób, księgi operacyjne, księgi rejestru chorych leczonych w Klinice Chirurgii Szczękowo-Twarzowej Klinicznego Szpitala Wojewódzkiego im. Fryderyka Chopina w Rzeszowie. W okresie tym leczono 64 dzieci i młodzieży, w tym 28 dziewcząt i 36 chłopców. Oceniano wiek i płeć pacjentów, przyczyny zgłoszenia się do lekarza, umiejscowienie cysty, wielkość i typ histopatologiczny torbieli śluzowych, a także przebieg i wyniki procesu diagnostycznego i terapeutycznego.

Wyniki: W badanej grupie przyczyną skierowania pacjenta do Kliniki było: u 25 pacjentów stwierdzenie w czasie badania przez stomatologa, pediatrę lub laryngologa, istnienia guzka w jamie ustnej, niepowodującego żadnych dolegliwości, u 13 pacjentów niepokój wzbudził guzek stopniowo powiększający się, u 18 wzmożone napięcie tkanek wokół guzka, u trojga zaczerwienienie i obrzęk (podejrzenie ropnia). Najczęstszym miejscem występowania torbieli była warga dolna (34 dzieci). Wielkość torbieli najczęściej wynosiła 2,1-3 cm (28 dzieci). Najczęstszym typem histologicznym mukoceli było MEP. Wszyscy pacjenci byli leczeni w Klinice w oddziale pracującym w trybie chirurgii jednego dnia, z dobrym wynikiem.

Wnioski: Stwierdzenie guzka (torbieli) w jamie ustnej u dziecka może być przypadkowe podczas rutynowego badania również przez pediatrów, stąd konieczne jest rozszerzenie wiedzy dotyczącej torbieli śluzowych małych gruczołów ślinowych. Najczęściej występujący typ MEP może mieć związek z różnego typu urazami błony śluzowej jamy ustnej.

Słowa kluczowe: torbiele śluzowe związane z wynaczynieniem śluzu, retencja śluzu w małych gruczołach ślinowych, diagnostyka, leczenie chirurgiczne

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INTRODUCTION

Mucocele is a cyst of the minor salivary glands located in the oral mucosa. These cysts may also develop in the major salivary glands, predominantly in the sublingual salivary gland. Sublingual ranulas are located in the fundus of the oral cavity. Mucoceles may appear anywhere on the mucosa, however, they appear most commonly on the lower lip, or less frequently on the cheek, palate or tongue mucosa [1, 2, 3]. Mucoceles of minor mucous glands are usually present as a single oval or round nodular lesion growing submucosally and often protruding over the mucosa. The nodules are filled with mucus, which shines through the mucous tissue resulting in a characteristic bluish shade. Lesions are usually palpable and painless [9].

The pathogenesis of a mucous retention cyst is usually based on the extravasation and accumulation of mucin in adjacent tissues or the obstruction of the salivary gland ducts [12].

The cysts are filled with mucus, which shines through the mucous membrane giving them a characteristic bluish shade. The distinctive color and consistency are the main clinical features. The differentiation diagnosis of mucoceles should consider disease entities that can easily be differentiated on palpation i.e. lesions like lipomas, hemangiomas, pleomorphic adenomas, nodules of minor mucous glands of different pathogenesis and abscesses [17].

Retention cysts of minor salivary glands are treated only surgically. It involves the enucleation of the cyst or its removal, including the surrounding gland. Incomplete cyst removal may result in relapse [18]. Marsupialization is permitted in the treatment of mucoceles of major mucous glands (sublingual gland), however, constant control during

the healing period must be ensured. Some authors obtained positive results applying other techniques, i.e.: cryotherapy and laser surgery [19, 20, 21].

AIM

The aim of the study was to present our own experience in the diagnostics and treatment procedure in children and teenagers with mucoceles of minor salivary glands.

MATERIAL AND METHODS

Our investigations were of a retrospective character. They were based on the files of 64 children and teenagers treated in the one-day surgery clinic Department of the Maxillo-Facial Surgery in Frederic Chopin Clinical Specialist Hospital in Rzeszów. The documentation included the department register, the medical history, operating records and histopathological findings. Special attention was paid to: the age and gender of patients, the reasons for consultation, the location of the cyst, the character and size of the cysts and the results of the histological examination. Statistical analysis was performed in Statistica 10.0 StatSoft. Two tests were used for statistical analysis. Pearson's Chi-square test evaluated the bilateral relationship of two factors. Quality data were taken into consideration in this test, not numerals. A one-sided significance test of structure indicators was used to compare the number and percentage of answers given to single choice questions in order to determine whether any of the responses occur significantly more or less often than any other. The tests used in the analysis were nonparametric. The level of statistical significance was adopted at p < 0.05.

RESULTS

The results of the research were presented in tables, and descriptively. 64 children including 28 (43.7%) girls and 36 (56.3%) boys were treated in the last decade (2005-2015). Most of the children were over 10 years of age. The youngest child was 5 years old, the oldest 17 years old. Most children [29] were between 11-15 years old, which amounted to 45.3% of the juvenile patients and 22 (34.4%) were over 15 years of age. Table I presents the data on the age and gender of the children treated. No statistically significant differences were found in terms of the distribution of girls and boys in different age ranges (p=0.1769).

The majority of the children. i.e. 39, were referred for treatment by dentists, 12 by pediatricians, 11 by GP's and 2 by otolaryngologists. Most cases of admission to the clinic were scheduled. The lesions in young children were most often found accidentally by their parents; older children reported the presence of lesions in the oral cavity themselves. The reasons for reporting for treatment are presented in table II. An asymptomatic lump accidentally detected by a dentist or a parent was the most common cause of reporting for treatment – it was diagnosed in 25 patients, representing 39.1%. 18 (28.1%) of the children reported for treatment after a period of observation by parents; a gradually enlarging nodule raised concern at some stage. Sudden redness and a swelling of tissues accompanied by inflammation was found in 13 children (20.35%). A burning sensation in the mucous membrane limited to the site of the injury was the reason for starting treatment in 5 (7.8%) of the patients, and only 3 (4.6%) reported pain. The analysis showed that none of the reasons for reporting for treatment existed significantly more frequently than any other, as confirmed by a non-parametric, one sided test – a comparison of structure indicators.

Mucoceles presented in this paper were located in various anatomical parts of the mouth. The data on their location are presented in table III. Analysis showed that in 34 cases, which accounted for 53.1% of cysts, they were located in the mucosa of the lower lip, while in 15 (23.5%) of the children, they were related to the upper

lip. Buccal mucoceles were found in 7 (10.5%) of the patients, mainly in the occlusal area and near the corner of the mouth, which may confirm the traumatic nature of their creation. Cysts located around the bottom of the oral cavity and the palate tissue were present in individual cases. The cysts were located at the bottom of the oral cavity in 3 (4.5%) of the patients, while on the palate, especially the soft palate, in 5 (7.8%) children. The structure test confirmed the presence of a statistically significant difference between the incidence of cysts in the area of the lower lip and the incidence of cysts in the area of the upper lip (p=0.0272), cheek (p=0.0207) and the floor of the oral cavity (p=0.0291).

Data obtained by interviewing parents and older children made it possible to determine the causes of mucous cysts in the oral cavity. Biting the buccal mucosa with molars was found to be the reason in 29 cases, while in 17 children compulsive sucking of the mucosa of the lip corner was observed during inspection. A single mechanical injury with a toothpick, a pencil or a toothbrush was found in 11 patients. In 7 cases, the cause of cyst formation failed to be determined.

The size of the lesions, clinically known as mucous cysts, found on admission, ranged from 0.5 cm to more than 3.5 cm in diameter. The smallest diameter of a nodule was about 0.5 cm and was located in the mucosa of the upper lip. The largest was 3.5 cm in diameter and was observed in the buccal mucosa. Cystic nodules with a diameter between 2.1 and 3 cm were the most numerous and were found in 28 (43.7%) patients. In 20 (31.3%) of the patients, the size of the nodule ranged from 1.1 to 2 cm. The nodules had a diameter of less than 1 cm only in 4 children, which represented 6.3% of all the patients. No statistically significant differences were found in the frequency of mucous cysts and their size in the comparison of structure indicators by means of a one-sided test. Data on the size of the cysts is presented in table IV.

The most common histological type of mucocele in the group of children was the Mucus Extravasation Phenomen (MEP), diagnosed in 37 (57.8%) of the patients. Mucus Retention Phenomenon (MRP) was found in 8 (12.5%) cases.

Table I. Classification of the respondents defined by age and sex. *Tabela I. Klasyfikacja badanych ze względu na wiek i płeć.*

Sex		Significance				
Płeć	<6	6-10	10-15	>15	Total	Istotność (p)
Girls Dziewczynki	1 100.0%	8 66.7%	10 34.5%	9 40.9%	28 43.7%	
Boys Chłopcy	0 0.0%	4 33.3%	19 65.5%	13 59.1%	36 56.3%	χ²(3)=4,93 p=0,1769
Total Razem	1 100.0%	12 100.0%	29 100.0%	22 100.0%	64 100.0%	

 $[\]chi^2$ – Pearson chi-square test; p- level of probability

χ²– wynik testu chi-kwadrat Pearsona; p-poziom prawdopodobieństwa

Table II. The reasons for reporting for treatment. Tabela II. Przyczyny zgłoszenia się do lekarza.

Redness and swelling, suspected abscess Zaczerwienienie obrzęk, podejrzenie ropnia	3 4.7%	0.1196	0.2602	0.1923	0.4323	ı
Burning and itching of the mucosa Pieczenie i swędzenie błony śluzowej	5 7.8%	0.0885	0.2627	0.3449	-	0.4323
Increased tension in tissue Wzmożone napięcie tkanek	18 28.1%	0.2270	0.3099	-	0.3449	0.1923
Gradually growing lump in the mouth Stopniowo powiększający się guzek w jamie ustnej	13 20.3%	0.1205	,	0.3099	0.2627	0.2602
Coincidentally suddenly detected lump without discomfort Przypadkowo nagle stwierdzony guzek bez dolegliwości	25 39.1%	1	0.2270	0.0885	0.1196	0.1196
8 0		25 39.1%	13 20.3%	18 28.1%	5 7.8%	3 4.7%
Reasons for reporting to treatment Przyczyny zgłoszenia Pr	Number = 64 <i>Liczba = 64</i>	Coincidentally suddenly suddenly detected lump without discomfort <i>Przypadkowo, nagle stwierdzony guzek bez dolegliwości</i>	Gradually growing lump in the mouth <i>Stopniowo</i> <i>powiększający się</i> <i>guzek w jamie ustnej</i>	Increased tension in tissue <i>Wzmożone</i> napięcie tkanek	Burning and itching of the mucosa <i>Pieczenie i swędzenie błony śluzowej</i>	Redness and swelling, suspected abscess Zaczerwienienie obrzęk, podejrzenie ropnia

Table III. The location of a cyst in the oral cavity. Tabela III. Umiejscowienie torbieli w jamie ustnej.

Cyst location mucosa <i>Umiejscowienie torbieli</i> <i>błona śluzowa</i> Number = 64 <i>Liczba = 64</i>		Lower lip Warga dolna	Upper lip Warga górna	Cheek <i>Policzek</i>	Fundi oral cavity Dno jamy ustnej	Soft palate Podniebienie miękkie
		34 53.1%	15 23.5%	7 10.9%	5 7.8%	3 4.7%
Lower lip Warga dolna	34 53.1%	-	0.0272	0.0207	0.0291	0.0540
Upper lip Warga górna	15 23.5%	0.0272	-	0.2436	0.2218	0.2302
Cheek <i>Policzek</i>	7 10.9%	0.0207	0.2436	-	0.4287	0.3770
Fundi oral cavity Dno jamy ustnej	5 7.8%	0.0291	0.2218	0.4287	-	0.4323
Soft palate Podniebienie miękkie	3 4.7%	0.0540	0.2302	0.3770	0.4323	-

Table IV. Data on the size of cysts. Tabela IV. Dane dotyczące wielkości torbieli.

>3 cm

Size of cysts <i>Wielkość torbieli</i> Number = 64 <i>Liczba = 64</i>		<1 cm	1,1-2 cm	2,1-3 cm	>3 cm
		4 6.3%	20 31.3%	28 43.7%	12 18.7%
<1 cm	4 6.3%	-	0.1523	0.0757	0.2770
1,1-2 cm	20 31.3%	0.1523	-	0.1921	0.2174
2,1-3 cm	28 43.7%	0.0757	0.1921	-	0.0658
	4.2				

0.2174

0.2770

18.7% In the remaining 19 (29.7%) patients, it was failed to clearly determine whether the cysts resulted from extravasation of mucus or from retention. The presence of statistically significant differences between the incidence of MEP cysts and the MRP type of cysts (p=0.0101) and types difficult to diagnose (p=0.0232) was demonstrated. Information

study are presented in table V. Table VI provides data comparing different types of cysts and the age of the patients.

on the histological types of mucoceles occurring in the

Statistical analysis showed no statistically significant differences in the incidence of cyst types depending on the age of the children.

The incidence of different types of cysts depended on their location, as confirmed by Pearson's chi-squared test (p=0.0000). MEP cysts were reported most frequently on the mucosa of the lower lip and a cheek, while MRP cysts were most frequently reported on the upper lip, or there were cysts whose manner of creation was difficult to determine. Cyst types depending on their location are presented the table VII.

0.0658

All the patients were treated in the one-day surgery mode, most of them - i.e. 42 (65, 6%), were administered general anesthesia, while 22 patients were operated under local anesthetic with sedation. The latter were older children over 15 years of age. The cyst was evacuated without damage

Table V. The incidence of histological types of mucoceles. *Tabela V. Występowanie typów histologicznych mucoceli.*

Histological types of mucoceles Występowanie typów histologicznych mucoceli Number = 64 Liczba = 64		Mucus Extravasation Phenomenon MEP	Extravasation Phenomenon Phenomenon Phenomenon Phenomenon Phenomenon	
		37 (57.8%)	8 (12.5%)	19 (29.7%)
Mucus Extravasation Phenomen MRP	37 (57.8%)	-	0.0101	0.1717
Mucus Retention Phenomen MEP	8 (12.5%)	0.0101	-	0.0232
Difficult to determine Trudne do określenia	19 (29.7%)	0.1717	0.0232	-

Table VI. The incidence of mucocele types by age. Tabela VI. Występowanie typów torbieli w zależności od wieku.

Age range						
Przedział wieku	Mucus Extravasation Phenomenon MEP	Mucus Retention Phenomenon MRP	Difficult to determine Trudne do określenia	Total Razem	Significance (p) Istotność (p)	
<6	0 0.0%	0 0.0%	1 100.0%	1 100.0%		
6-10	10 83.3%	0 0.0%	2 16.7%	12 100.0%		
10-15	18 62.1%	3 10.3%	8 27.6%	29 100.0%	χ²(6)=9,41 p=0,1520	
>15	9 40.9%	5 22.7%	8 36.4%	22 100.0%		
Total <i>Razem</i>	37 57.8%	8 12.5%	19 26.7%	64 100.0%		

 $[\]chi^2$ – Pearson chi-square test; p- level of probability

to the bag in 39 (60.9%) of the patients and in 25 cases the bag was broken during surgery and the liquid content was spilled in the operational site.

In these cases, surgery was extended and the gland connected with the cyst was evacuated. The children felt well after the operation. The postoperative course was generally uneventful. Most children did not report pain, only in 19 (29.7%) cases painkillers, mainly paracetamol and pargyline, were administered. Minor complications in the healing were observed in 14 patients, the area surrounding the wound was reddened in 2 children, hematoma and petechiae on the mucosa were present in 4 cases and 3

patients had inflammatory infiltration initially followed by wound abscess. In 5 patients the wound after the cysts' evacuation spread and was followed by granulation. In 59 (92.2%) cases the stitches were removed on the 8th postoperative day and in 5 patients it was on the 10th day after surgery. After cyst evacuation, the children were monitored in ambulatory mode until complete healing and received the result of the histopathological examination with the recommendation to report to the medical provider if any symptoms occurred after treatment.

The follow-up was performed in the period of 29-48 months after cyst evacuation. 35 patients, which accounted

χ² – wynik testu chi-kwadrat Pearsona; p-poziom prawdopodobieństwa

Table VII. Types of cysts by location.

Tabela VII. Typy torbieli w zależności od umiejscowienia.

Location Umiejscowienie	Mucus Extravasation Phenomenon MEP	Mucus Retention Phenomenon MRP	Difficult to determine Trudne do określenia	Total Razem	Significance (p) Istotność (p)
Lower lip	3	1	11	15	
Warga górna	20.0%	6.7%	73.3%	100.0%	
Upper lip	27	1	6	34	
Warga dolna	79.4%	2.9%	17.7%	100.0%	
Cheek	5	0	2	7	
<i>Policzek</i>	71.4%	0.0%	28.6%	100.0%	
Bottom of the oral cavity Dno jamy ustnej	2 40.0%	3 60.0%	0 0.0%	5 100.0%	χ²(8)=53.52 p=0.0000
Soft palate Podniebienie miękkie	0 0.0%	3 100.0%	0 0.0%	3 100.0%	
Total	37	8	19	64	
<i>Razem</i>	57.81%	12.5%	29.7%	100.0%	

χ² – Pearson chi-square test; p- level of probability

for 54.7% of all patients operated due to mucoceles, reported for follow-up. No recurrence was observed in any cases. The scar after cyst evacuation was invisible in most cases, 6 children had a slight thickening of the mucosa of the lip at the wound, caused by granulation. The results observed during the follow-up after a long time, were in line with the report by the authors who conducted their inspection over a shorter period of time.

DISCUSSION

Mucocele formation has not been entirely explained. The majority of authors stress the importance of mechanical and traumatic factors, such as biting of the lip mucosa, biting pencils and nails, repetitive inflammatory factors, irritation by braces [4, 5]. A review of the literature indicates that stimulation of the mucosa with metal piercing, other decorations and so-called jewelry within the oral cavity also predisposes to mucocele formation. Strzałkowska et al. [6] also confirmed the traumatic mechanism of these lesions' formation as a result of the irritation by brackets of orthodontic braces. Congenital mucoceles were also reported in the literature [7, 8]. Cysts located within the oral and lip mucosa usually do not exceed 2 cm in diameter. An example of a large-size mucocele is a ranula affecting sublingual salivary glands, which can even exceed 5 cm in diameter. The ranula may cause difficulty in speaking, swallowing or even breathing [10]. The diagnosis of a mucocele is usually not difficult in most cases and based on characteristic clinical symptoms which rarely require other specialized diagnostic tests [11].

Due to the pathogenesis, construction and contents of mucoceles, Yamasoba et al. [13] distinguished two types of mucocele etiology: the result of Mucus Extravasation Phenomenon (MEP) and Mucus Retention Phenomenon (MRP) in the ducts. In the extravasation type of cyst what occurs are: acute and chronic inflammation cells, granulation tissue, fibroblasts and elements of fibrous tissue forming the so-called pseudo capsule surrounding the mucus [14]. Increased activity of amylase, alkaline phosphatase is observed, which can be a sign of increased fibroblast activity. In the retention type of cyst, fluid is surrounded by a bag of epithelial tissue consisting of a layer of cylindrical cells, salivary ducts or cubic cells, which is a combination of a gland and tubule [15]. MRP cysts, in contrast to MEP cysts, do not exhibit inflammatory reaction and possess a real bag [16]. Koszowski et al. [3] based on 30 cases expressed the belief that clinical symptoms of mucocele correspond to a wide range of histopathological pictures. A review of the literature revealed that the extravasation type of cyst is more common in children. Mucoceles are diagnosed mainly based on the clinical picture and medical history, in which trauma or chronic mechanical irritation are significant.

Mucoceles are lesions occurring on the mucous membrane in the mouth. They occur both in adults and children, but most frequently in people in their twenties [1, 4, 11]. They are rarely observed before the age of 1. The incidence is not related to gender, which is also confirmed by the authors' own observations. The diagnosis of oral mucosal cysts is mainly based on a carefully taken history and clinical examination. In

χ² – wynik testu chi-kwadrat Pearsona; p-poziom prawdopodobieństwa

the material presented in this study, extravasation cysts were found significantly more frequently compared to retention cysts. This fact is confirmed by Bagan et al. [2] who found extravasation cysts in 95% patients. The remaining 5% were retention cysts formed due to the accumulation and obstruction of mucous gland ducts with thick mucus. At first the ducts became enlarged, then outflow obstruction occurred. They are referred to as the so-called pseudocysts and are etiologically related to mechanical trauma. Extravasation cysts do not have a clear bag of tissue and mucus accumulated in the tissue is surrounded by connective tissue cells, fibroblasts and granulation tissue. They are referred to as the so-called pseudocysts. Retention cysts have a bag, and inflammation is not observed [12, 15]. Most authors recommended a differentiation diagnosis of mucous cysts and lipomas, fibromas, angioma and benign and malignant tumors of minor mucous glands located in the oral cavity [2, 7, 11].

The treatment of choice is surgical. However, publications can be found which recommend cryotherapy and the cryodestruction of nodules, and laser surgery [19, 20, 21]. In our own material, mucoceles were primarily enucleated, and in the case of mucous content discharge related to breaking the bag in the operating field, cyst fragments were evacuated in the acute mode with a fragment of the gland. To summarize, the following conclusions can be drawn on the basis of our own observations.

CONCLUSIONS

- 1. Mucocele ascertainment in children's oral cavity could be accidentally made in routine pediatric examination, therefore it is necessary to extend the knowledge of doctors about small salivary gland mucoceles.
- 2. The most frequently appearing type, i.e. MEP, could be related to different types of trauma in the oral mucose.

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