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Research Article

Salivary Mucoceles in Children and Adolescents: A Clinicopathological Study

Abstract

Background: Mucoceles are cavities filled with mucous and often occur in the oral mucosa. When they develop in children may cause diagnostic and management concerns.

Objectives: The aim of the present study was to examine retrospectively the clinical and pathological features of salivary mucoceles in children and adolescents.

Methods: 102 cases of salivary mucoceles in children and adolescents aged from 1 to 17 years were retrieved from our archives and examined for their clinical and histological features and frequency of recurrence. All cases were treated surgically under local anesthesia.

Results: Salivary mucoceles most often occurred in patients aged from 11 to 17 years (65,68%) with very weak male predominance (1,04/1). They were located most frequently in lower labial mucosa (87,25%). The incidence of extravasation mucoceles and mucous retention cysts were 99, 01% and 0,98% respectively. Confirmed history of local trauma and recurrence of the lesions were found 12,74% and 3,92% respectively. Histologically, 77.22% of cases of extravasation mucoceles were well-defined cysts surrounded by a capsule of granulation tissue and 22.77% granulation tissue intermixed with mucous. There was not relationship between the duration and histology of the mucous extravasation mucoceles.

Conclusions: Possibly, unnoticed local trauma and/or biting habit are the main etiological factors for the most frequent occurrence of extravasation mucoceles in the lower labial mucosa. The occurrence of mucous retention cysts and superficial mucoceles in children is very rare.

Introduction

Mucoceles are cavities filled with mucous and often occur in the oral mucosa. They are subdivided into extravasation mucocele or mucous escape reaction and mucous retention cyst. Extravasation mucocele represents an accumulation of extravasated mucous that is surrounded by granulation tissue and results secondary to traumatic rupture of minor salivary gland excretory duct [1-5] .The extravasation mucocele that is located in the floor of the mouth that arises from the sublingual gland, either from a torn main duct or from ruptured acini after obstruction, is called ranula [6-8]. The superficial mucocele represents a variant of extravasation mucocele that results from the accumulation of mucous beneath the mucosal epithelium. The mucous retention cyst is an accumulation of mucous surrounded by the lining epithelium of excretory duct and results from obstruction [1-5]. Clinically, extravasation mucoceles and mucous retention cysts appear as dome-shaped, painless swellings that measure from a few millimeters to

several centimeters. The color of lesions ranges from blue to normal pink [4–9]. The superficial mucoceles appear as small painless, single or multiple clear vesicles filled with mucous and measure from 1 to 5 mm [10–12].

Mucoceles most commonly affect children. Nevertheless, there is only two specific published research among the pediatric population [13,14]. Some researchers focus on the general population and then specify the incidence in pediatric patients. On the other hand, other authors refer to isolated clinical cases.

The aim of the present study was to examine in detail the clinical and pathological features of salivary mucoceles in children and adolescents.

Materials and Methods

102 cases of salivary mucoceles in children and adolescents (1to 17 years) were retrieved from the archives of our Department

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and retrospectively examined. All cases were treated surgically under local anesthesia. From the total number of our cases, 101 cases were extravasation mucoceles, and only 1 mucous retention cyst. The age and gender of patients and location, duration, size and recurrence of the lesions were recorded. The histological features of all cases were evaluated by two of the authors (D.A. and A.P.) independently of each other, and when there were differences, re-examination and discussion were necessary to establish uniformity. The relatives of patients had given informed consent and the whole study was performed according to the declaration of Helsinski II.

Results

Salivary mucoceles most often occurred in patients aged from 11 to 17 years (65,68%) whereas 34, 31% of our patients were under the age of 10 years, in addition was observed a very weak male predilection (ratio : 1,04/1). They were located most frequently in lower labial mucosa (87,25%), followed by floor of the mouth (6,86%) and ventral surface of the tongue (4,90%) . The incidence of extravasation mucoceles and mucous retention cysts were 99,01% and 0,98% respectively. The duration of mucoceles extended from 1 week to 12 months (mean value 4,5 months) and their size ranged from 0,2-1,7 cm (mean value 0,7cm). The clinical appearance of a typical extravasation mucocele is presented in Figure 1 and the clinical features of all salivary mucoceles are summarized in Table 1. Only 13 patients had a history of confirmed local trauma (12,74%). The mean value of follow-up after treatment was 6.5 months and recurrence was observed in 4 cases (3,92%). The above mentioned cases were surgically retreated and follow-up for one year did not showed any recurrence.

The histological examination of 101 cases of extravasation mucoceles showed that 78 of cases (77,22%) were welldefined mucous cysts surrounded by a capsule of granulation tissue that contained macrophages, sometimes neutrophils and occasionally lymphocytes (Figure 2A). 18 consisted of granulation tissue intermixed with mucous (Figure 2B). 14 of the above mentioned 18 cases, in many regions were separated from the attenuated mucosal epithelium by a strip of granulation tissue. 5 of the total 101 extravasation mucoceles were well defined cysts associated with granulation tissue intermixed with mucous. The one case of mucous retention cyst was occurred in a 15-year-old boy and located in the floor of the mouth with duration of one month. Histologically the cyst was unilocular and its lining epithelium consisted of cuboical or atrophic squamous epithelium (Figure 2C). The incidence of mucous retention cyst was 0,98% of cases. Examination of the clinical and histological results did not showed a relationship between the duration and histology of extravasation mucoceles.

Discussion

Salivary mucoceles seem to be common benign lesions on pediatric patients. Large series reported that salivary mucoceles represent 16%-17,2/% of all oral and maxillofacial lesions in pediatric patients [15,16]. Our results showed prevalence (65,68%) in patients aged from 11 to 17 years and are in agreement with previous studies [17,18,19]. Whereas 34,31% of our patients were under the age of 10 years and this finding may have some importance in preparing these patients for surgery: younger patients may require additional measures in terms of management/approach, anesthesia and sedation. Furthermore, Nico et al [13] reported prevalence (65,7%) in the first decade of life. According to the literature [1,13,14,19] salivary mucoceles show female predominance whereas in our study was found a very weak male predilection. However, Jani et al [18] reported a slight male predominance (1:1,2) which is very near to our finding. Salivary mucoceles can appear at any site of the oral mucosa where salivary glands are present. In general population, the most common site of salivary mucoceles is the lower labial mucosa [4,9,20] and this site seems to be repeated in children and adolescents according to our study (87,25%). This finding is similar to those reported by Nico et al [13] and Minguez-Martinez et al [14] (83,3% and 79,8% respectively). Although local trauma is considered to be the main etiological factor of extravasation mucoceles, in

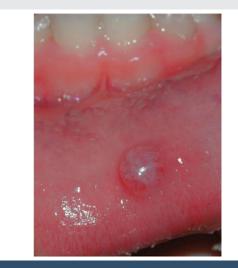
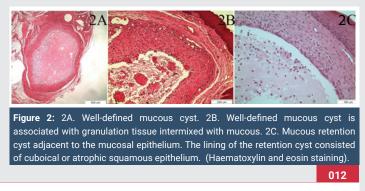


Figure 1: Clinical appearance of extravasation mucocele in a child 7 years old

Table 1: Clinical features of 102 cases of salivary mucoceles in children and adolescents.

Age: n/%	Gender: n/%	Location: n/%	Size	Duration
1-10y:	Male:	Lower labial mucosa :	Range	Range
35/34,31%	52/50,98%	89/87,25%	0.2-	1w-12mo
11-17y:	Female:	Floor of the mouth: 7/ 6,86%	1.7cm	MV
67/65,68%	50/49,01%	Ventral surface of tongue:	MV	4.5 mo
	Ratio :	5/4,90%	0.7 cm	
	1,04/1	Buccal mucosa:1/0,98%		

n=number, % =percentage, y=year ,MV=mean value, w=weeks, mo=months



the present study we found very few patients (12,74%) with confirmed history of trauma. The most frequent occurrence of extravasation mucocles in the lower labial mucosa in patients aged from 11 to 17 years, seen in our study, possibly is due to the development of lip habit as reaction to psychological/ emotional stress [18]. Mucoceles arising on the ventral surface of the tongue are known as mucocele of the glands of Blandin-Nuhn. These glands are located near the ventral tip of the tongue and are arranged in a mass with a horseshoe shape. They are embedded in the muscles of the ventral aspect near the midline. Mucoceles of the glands of Blandin-Nuhn are considered to be rare in adults whereas seem to be more prevalent in young people [13].

Only one mucocele (0,98%) was located on the floor of the mouth and was finally diagnosed as a ranula. These lesions acquire importance in the pediatric population because they have to be differentiated from deeper and more serious conditions such as dermoid cyst, hemangioma, and cystic higroma which may arise in the floor of the mouth.

Mucous retention cysts are rare in the oral cavity. The incidence of mucous retention cysts in general population ranges from 2,8% to 16,6% [12,17,19]. They occur most prevalent in patients older than 20 years and most often are located on the floor of the mouth, buccal mucosa, and palate [1,12,21,22]. In the current study we found only one case of mucous retention cyst (0,98% of cases) in a 15-year-old adolescent and this result points to the rarity of this variant of salivary mucoceles in children and adolescents. However, previous studies did not reported the occurrence of mucous retention cysts [13,14].

Occurrence of superficial mucoceles was not seen in the present study and this finding concurs with previous reports [13,14]. Searching in the literature revealed a review of 27 well documented cases of superficial mucoceles and a report of 4 additional cases [23,24]. Three of these total 31 cases of superficial mucoceles occurred in patients aged from 10 to 14 years and were associated graft-versus-host-disease [25]. The etiology of superficial mucoceles is controversial and remains unclear. Possible etiological factors include local trauma and various foods and liquids [26-28]. Also superficial mucoceles have been associated with lichen planus, mucous membrane pemphigoid and graft-versus-host-disease [25,26]. The histological results of the present study showed that most extravasation mucoceles (78/101) were well-defined mucous cysts and this finding concurs with previous studies [1,13]. Also our results showed that there was not relationship between the duration and histology of the lesions and agree with previous study [29]. The presence of granulation tissue for long time seems to represent a local unsuccessful attempt to repair the lesion [5].

Treatment of salivary mucoceles is surgical, followed by careful dissection of the affected minor salivary gland, although some authors reported spontaneous resolution of superficial mucoceles and no need for treatment [26,30]. Mucoceles can be treated by common scalpel, electrosurgery, cryosurgery, or CO2 laser vaporization [31]. Regardless of the chosen technique, it is important to reach the muscle layer. One research group observed complete recovery without reappearance when treating lesions with high corticosteroid [32]. In the current study all cases were treated by excision under local anesthesia, followed by careful dissection of the affected minor salivary glands. Recurrence was seen in 3,92% of cases. This result is not in agreement with the reported recurrence 8% of cases in previous study in children [14]. The disagreement is possibly due to the different surgical procedures which were performed.

Finally the majority of the recent studies are in agreement that salivary mucoceles should be considered as the most frequent oral benign lesion encountered in children [33-35] and a group was mentioned that there was recorded a significant difference between the study pediatric and adult populations [34]. Usually diagnosis can be made during routine pediatric intraoral examination, consequently it is required to expand medical and dental personnel's awareness about salivary mucoceles.

Conclusions

It is possible that unnoticed local trauma and/or biting habit to represent the main etiological factors for the most frequent occurrence of extravasation mucoceles in the lower labial mucosa. The occurrence of mucous retention cysts and superficial mucoceles is very rare in children. Treatment of choice for the salivary mucoceles is surgical, followed by careful dissection of the affected minor salivary gland. The younger patients may require additional measures in terms of management/approach, anesthesia and sedation.

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