Lymphangioma of the Lower Lip Mimicking a Mucocele in Children

Fabrício Kitazono de Carvalho, DDS, PhD[1]
Tiago Novaes Pinheiro, DDS, PhD[2]
Raquel Assed Bezerra da Silva, DDS, PhD[3]
Alexandra Mussolino de Queiroz, DDS, PhD[4]
Lea Assed Bezerra da Silva, DDS, PhD[5]
Paulo Nelson-Filho, DDS, PhD[6]

ABSTRACT

Lymphangioma is a hamartoma resulting from the proliferation of common lymphatic vessels in the head and neck area, rarely appearing in the lower lips. Its clinical presentation is a nodular mass with a pebbly surface and no defined borders formed by a cluster of slow-growing coalescing vesicles. The purpose of this paper is to present two children with a lesion in the lower lip whose clinical characteristics (single papillary lesion with a pediculated base, same color and consistency of the surrounding mucosa, and negative result for diascopy test) suggested an initial diagnosis of mucocele but were later confirmed as cavernous lymphangiomas. The clinical and microscopic characteristics of the lesions are discussed as well as the differential diagnosis and the treatment approach. These cases reinforce the importance of always performing a confirmatory histopathological analysis, even for lesions with typical clinical features. (J Dent Child 2015;82(3):TBD)

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An accurate diagnosis in dentistry is essential for providing an effective clinical treatment, especially when it comes to oral mucosal lesions. In addition to creating parental anxiety, their occurrence in children may cause orofacial functional and esthetic problems of varying severity. The diagnosis of oral mucosal lesions is usually obtained by visual clinical examination, clinical tests, and review of lesion history, leading to a suggestive clinical diagnosis that should be confirmed by biopsy and histopathological examination.[1,2] A study reported moderate concordance (approximately 50 percent) between clinical and definitive histopathologic diagnosis of oral soft tissue lesions, which shows the importance of the histopathological analysis, even for lesions with a typical clinical aspect.[1]

Lymphangiomas are uncommon, benign, hamartomatous tumors of lymphatic vessels and are considered to be developmental malformations rather than true

[1]Dr. Carvalho is an assistant professor; Drs. [3]R.A.B. Silva and [4]Queiroz are associate professors; and Drs. [5]L.A.B. Silva and [6]Filho are professors, all in the Department of Pediatric Dentistry, School of Dentistry, University of São Paulo, Ribeirao Preto, São Paulo, Brazil. [2]Dr. Pinheiro is a professor, School of Dentistry, Amazonas State University, Manaus, Amazonas, all in Brazil.
Correspond with Dr. Carvalho at fabricio_kc@forp.usp.br.

neoplasias.[3,4] They are classified as simple, cavernous, and cystic, according to the diameter and number of lymphatic vessels in the lesion.[2,5] Surgical excision is usually the treatment of choice, but other therapies like cryotherapy, electrocauterization, and laser therapy can be used for removal of large lesions that could represent a risk of damage to adjacent anatomical structures or functional and esthetic impairment.[4] Lymphangiomas grow slowly and have a predilection for the head and neck region. Its occurrence in the lower lip is rare, accounting for less than one percent of the biopsies performed in this region.[6]

The purpose of this paper is to present two children with a lesion in the lower lip whose clinical characteristics suggested an initial diagnosis of mucocele but later were confirmed as cavernous lymphangiomas.

CASE REPORT

A six-year-old boy and a four-year-old girl came for a dental consultation at the pediatric dentistry clinic in the School of Dentistry, Amazonas State University, Manaus, Amazonas, Brazil, each with complaints of a lesion in the lower lip with duration of one and two years, respectively. The patients reported absence of pain or other symptoms, but the girl and her mother reported difficulty with eating and speaking. In both cases, the medical and family histories were noncontributory. In the clinical examination, both lesions presented as a single nodular mass with a polypoid base and the same consistency as the surrounding mucosa. The boy's lesion was pink in color, measuring approximately three mm in diameter and located in the center of the lip (Figure 1A). The girl's lesion had a bluish color, with diameter of approximately one cm, and was located on the right side of the lip (Figure 1B).

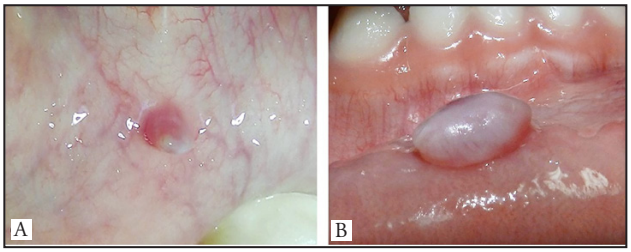

Figure 1. Single papillary lesions with a polypoid base located in the lower lip of young children measuring approximately three mm (A) and one cm (B) in diameter.

Diascopy with a microscope slide was performed in both cases to rule out the diagnosis of hemangioma. This test is used to determine whether a lesion is vascular (inflammatory), nonvascular, or hemorrhagic. A microscope slide is pressed against a lesion (diascopy) to see whether it blanches. Hemorrhagic and nonvascular lesions do not blanch, while vascular and inflammatory lesions do. Both lesions presented a negative result for diascopy. Based on the clinical characteristics, an initial diagnostic hypothesis of oral mucocele was suggested for both lesions.

After obtaining written informed consent from the parents, an excisional biopsy was performed chairside without sedation, using local anesthesia (lidocaine 2% with epinephrine 1:100.000), and behavior guidance only. The clinical impression of mucocele changed during the excisional biopsy procedure because the characteristics and consistency of the nodular mass and absence of associated salivary glands suggested that the lesion could have a different definitive diagnosis after microscopic examination. Dissection of involved salivary glands from surrounding tissues was not necessary because there was no glandular involvement. A single suture was placed and post-operative instructions were given to the children and parents.

The histopathological analysis of the smaller lesion revealed numerous delicate lymphatic vessels in the submucous conjunctive tissue without salivary glandular tissue and structures, leading to a diagnosis of capillary lymphangioma (Figure 2A). The examination of the larger lesion showed countless large lymphatic vessels in the submucous conjunctive glandular tissue with plasma-rich areas and eventual erythrocytes (Figure 2B), resulting in a diagnosis of cavernous lymphangioma. The patients were followed up for 12 months with no lesion recurrence.

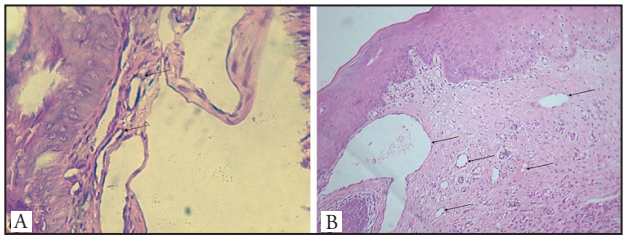

Figure 2. Histopathological analysis of the excised lesions. (A) The smaller lesion exhibited numerous delicate lymphatic vessels (arrows) in the submucous conjunctive tissue without salivary glandular tissue and structures. (B) The larger lesion exhibited countless large lymphatic vessels (arrows) in the submucous conjunctive glandular tissue with plasma-rich areas and erythrocytes.

DISCUSSION

Several cases of lesions that mimicked the clinical appearance of mucocele but had definite histological diagnoses of other lesions, such as canalicular adenoma,[7] collagenoma,[8] sialolithiasis,[9] and schwannoma,[10] have been reported. To the best of our knowledge, there are no reports of lesions with clinical characteristics of mucocele and final diagnosis of lymphangioma of the lower lip confirmed histologically.

Mucoceles are lesions resulting from the rupture of salivary gland ducts and consequent accumulation of mucus into the soft tissue around this gland. They are

most frequently found in the oral cavity of children and adolescents.[11,12] In pediatric dentistry, mucoceles correspond to approximately 12 to 56 percent of the biopsies.[13-15] Clinically, they are nodular, exophytic, and asymptomatic lesions with bluish color or a color similar to the surrounding mucosa and a size rarely exceeding 1.5 cm in diameter.[12,16] They may be found in any site of the oral mucosa with salivary glands but usually occur in the lower lip, followed by the tongue, cheek mucosa, and palate.[17] In our cases, the lesions had the aforementioned clinical characteristics and were present in the lower lip, which is the most common location of mucoceles.

The diagnosis of mucocele is based on the clinical aspect of the lesion and the patient's report. There is usually a history of trauma to the region and variations in lesion size. Differential diagnosis is made with other lesions that may have exophytic characteristics, such as fibrous epithelial hyperplasias, lipomas, hemangiomas, and some minor salivary gland tumors and cysts.[1,12,17] Excisional biopsy is the most common treatment, as performed in our patients, with low recurrence rates. Marsupialization and micromarsupialization are indicated for very young children who are unable to cooperate with a surgical removal or for larger lesions. Other treatment options, such as homeopathy, injection of sclerosing agents, and watchful waiting, have also been described.[18] The concordance between the clinical diagnostic hypothesis and the results of the histopathology exam ranges from approximately 69 to 87 percent for mucoceles.[1,17]

On the other hand, lymphangiomas are rare lesions, appearing in less than one percent of biopsies performed in routine pediatric dental practice.[13,15] The vast majority of lymphangiomas is congenital and seen at birth or evident up to two years of age. The most common sites of occurrence are the submandibular area (close to the parotid gland) and the anterior two thirds of the tongue. They may also occur less frequently on the palate, gingiva, cheek mucosa, and alveolar ridges and are rarely seen in the lips.[3]

Clinically, oral lymphangiomas are usually superficial lesions formed by a cluster of transparent vesicles resembling the appearance of tapioca pudding or frog eggs. These lesions have a slow growth, a red-purple coloration, and a size varying from one to five cm in diameter.[5] Differential diagnoses of lymphangiomas from other lesions include hemangiomas, dermoid cysts, thyroglossal duct cysts, pyogenic granulomas, and granular cell tumors.[4,5,19] Neither lesion of the present cases had the classical clinical aspect of a lymphangioma, mimicking a mucocele, which was the initial impression in both cases. It is worth mentioning that diascopy and palpation were performed as additional clinical tests to discard the diagnosis of hemangioma in both cases.

Oral lymphangioma can cause enlargement of the tongue (macroglossia), affecting speech, mastication, and oral hygiene, making its surgical excision difficult.[19] In one of the cases, difficulty in speaking and eating was reported, despite the lesion being located in the lower lip. Histopathologically, lymphangiomas can be classified into three subtypes: (1) capillary; (2) cavernous; and (3) cystic (cystic hygroma). The cavernous type, diagnosed in case two, is more frequent in the mouth.[3-5] Lymphangiomas occurring in the lip or buccal mucosa are the most clinically similar to mucoceles, but they are not usually considered for differential diagnosis because they are rare lesions.

There is a 27 percent risk of recurrence after complete excision of lymphangiomas, and this incidence may increase according to the characteristics and size of the lesion, often exceeding 50 percent in cases of partial excision.[20,21] In the present study, there was no recurrence of the lymphangiomas after one year.

As seen in our cases, it is important that, even for lesions with typical clinical characteristics, additional clinical investigations (e.g., diascopy, palpation, needle puncture) and complementary examinations, such as biopsy and histopathological analysis, are always recommended to confirm the diagnosis and establish the adequate treatment.

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