

## Case Report

# Diagnosis and Management of Lower Labial Mucosa Ulcer in a Hypotonic Infant

Yair Schwimmer<sup>1\*</sup>, Aviv Shmueli<sup>1</sup>, Jawad Abu-Tair<sup>2</sup>, Rinat Schwimmer-Noy<sup>3</sup> and Alexander Maly<sup>4</sup>

<sup>1</sup>Department of Paediatric Dentistry, The Hebrew University, Israel

<sup>2</sup>Department of Oral and Maxillofacial surgery, The Hebrew University, Israel

<sup>3</sup>Department of Oral Medicine, The Hebrew University, Israel

<sup>4</sup>Hadassah-Hebrew University Medical Center Department of Pathology, Israel

\*Corresponding author: Yair Schwimmer, Department of Paediatric Dentistry, The Hebrew University-Hadassah School of Dental Medicine, Jerusalem, Israel

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## Introduction

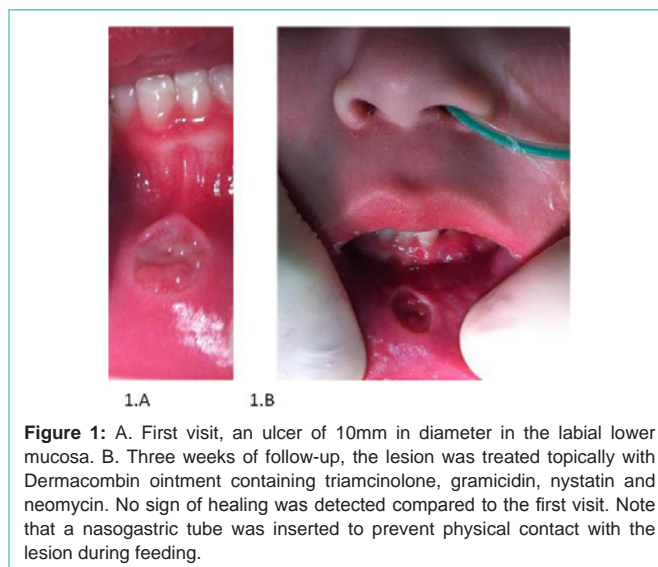
Reactive oral lesions associated with chronic local irritation or trauma may manifest as tumor-like hyperplasia. These proliferations are painless, pedunculated or sessile masses in different colors, from light pink to red. The surface appearance is variable, from non-ulcerated smooth to an ulcerated mass. Lesion size varies from a few millimeters to several centimeters [1].

Most studies on the frequency of oral diseases in paediatric populations examined the presence of dental caries, periodontal diseases or other oral lesions such as tumors or cysts. Zuñiga et al. studied 542 oral biopsy specimens from children (which represented 20.6% of all oral biopsies), with mean patient age of 11.1 years, and female predilection (60.5%). The most common category of oral lesions was inflammatory and reactive (75.8%), followed by tumor/tumor like (16.8%) and cystic (7.4%). The mucocele was the most commonly found lesion, followed by pyogenic granuloma and irritation fibroma. Together, these reactive lesions accounted for 63.8% of all paediatric oral biopsies. The most common site for lesions was the lower lip (50.3%) [2].

Cerebral Palsy (CP) is a condition caused by damage to the brain during or shortly after birth, and during the early years of life. The worldwide incidence is 2-2.5 per 1,000 live births [3]. CP comprises a group of permanent disorders of the development of movement and posture, which limit activity, and which are attributed to non-progressive disturbances that occurred in the developing fetal or infant's brain. The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication and behavior. In some cases, epilepsy and secondary musculoskeletal problems are diagnosed [3]. Patients with CP may show oral clinical manifestations such as lip incompetence, a high rate of Class II malocclusion and an anterior open bite due to hypotonia of the orofacial muscles (poor control of the lips and tongue), which may

## Abstract

We describe a 28-month-old girl with congenital cerebral palsy, uncontrolled epilepsy, failure to thrive, breathing difficulties and secondary diabetes mellitus due to a ketogenic diet. She was brought to our clinic by her parents with the chief complaints of a long lasting ulcer in her lower labial mucosa and feeding difficulties. We describe the diagnosis and treatment of the ulcer over the course of one year. Possible etiological factors for the lesion are discussed.



**Figure 1:** A. First visit, an ulcer of 10mm in diameter in the labial lower mucosa. B. Three weeks of follow-up, the lesion was treated topically with Dermacombin ointment containing triamcinolone, gramicidin, nystatin and neomycin. No sign of healing was detected compared to the first visit. Note that a nasogastric tube was inserted to prevent physical contact with the lesion during feeding.

lead to additional disturbances in swallowing, chewing and sucking. Most young CP patients visit special daycare centers, where they receive some of their medical and para-medical treatment [3,4].

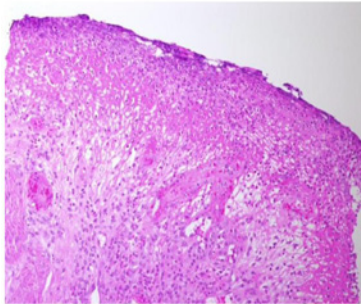
The purpose of this report is to present the diagnostic process and management of a long-lasting ulcerated lesion in the lower labial mucosa in a 28 month old girl with CP.

## Case Report

A twenty-eight month old girl with CP, uncontrolled epilepsy, failure to thrive, breathing difficulties and reactive diabetes (subsequent to a ketogenic diet) arrived at the department of paediatric dentistry due to an unhealed ulcer for 3 weeks. Her paediatrician prescribed Oracort E paste (contains 3% Lidocaine hydrochloride and 0.1%



**Figure 2:** Biopsy under local anaesthesia. The lower labial mucosa immediately after incisional biopsy.



**Figure 3:** Ulcer, base of the ulcer active granulation tissue.

Triamcinolone acetonide; TARO Pharmaceutical Industries Ltd, Israel) to relieve the symptoms and enable proper feeding from a bottle. Oral hygiene was maintained by brushing the teeth twice a day; and due to perioral muscular hypotonus she received physical stimulations of the oral soft tissues a few times a day by caregivers.

A shallow ulcer (1 cm diameter) in the lower labial mucosa was observed (Figure 1a). Self-trauma (biting) was unlikely, because the upper teeth had no contact with the lesion in the occlusion position. After consultation with a maxillo-facial surgeon and oral medicine specialist, it was decided to take viral, fungal and bacterial cultures, to stop the physical stimulation, to clean the ulcer with chlorhexidine gluconate 0.12% alcohol-free solution 4-5 times a day; and, to relieve pain before eating with topical Teejel gel (contains 8.7% choline salicylate and 0.01% cetalkonium chloride; Rafa Laboratories Ltd, Jerusalem). A follow up visit was scheduled after 1 week.

One week later the mother reported that the physical stimuli was stopped, the lesion was cleaned via chlorhexidine gluconate 0.12%, and the area was being treated topically with minocycline ointment for local bacterial infection, as prescribed by the child's paediatrician. At this time, laboratory analysis revealed negative findings of viral infection, but positive findings of gram positive streptococci, and undefined fungus (yeast forms). The clinical picture of the lesion was without change. Therefore, we recommended 2 more weeks of cleaning with CHX, and topical use of Dermacombin (Taro pharmaceutical industries LTD) ointment, which contained a combination of anti-bacterial and anti-fungal medication, 3 times



**Figure 4:** One-year follow-up after biopsy. Note the fibrotic appearance.

a day for two weeks. A decision was made that if the lesion would not heal during this period, an incisional biopsy would be done. Two weeks later a nasogastric tube was inserted for feeding (Figure 1b), and the lesion was biopsied (Figure 2).

A pathological diagnosis of pyogenic granuloma was established (Figure 3).

Two weeks later, partial primary healing was noticed and the nasogastric tube was removed. Excision of the entire lesion was recommended, but the parents refused.

Two months later the patient stopped eating from a bottle; and therefore, the paediatrician inserted the nasogastric tube again. At one year follow up, the lesion had disappeared (Figure 4), and the nasogastric tube was removed. After returning to bottle feeding the lesion returned. Due to unwillingness to stop bottle feeding, the nasogastric tube was inserted again, until the lesion would heal again.

## Discussion

CP is a permanent non progressive neuromuscular disorder caused by damage to the brain, prenatally or a few years post-natal. In our patient, hypotonia of the perioral muscles was diagnosed, which is characterized by drooling and uncoordinated or involuntary movements [5]. The CP was accompanied by seizures. This may cause clenching and powerful closure of the jaws, which may lead to trauma of the soft tissues [6].

For our patient, the recommendation to stop the topical usage of the minocycline ointment was due to young age, since this drug may be swallowed and may cause the developing teeth to change their color [7].

The girl described had perioral muscular hypotony. She was fed by a bottle. Her caregivers performed physical stimulation by moving their fingers intraorally and peri orally to strengthen the muscular tonus. We assume that consequent to the muscular hypotony, the irregular swallowing movements of the muscles were the initial cause of the lesion. Both the bottle feeding and the physical stimulation were suspected to have effects. However, when the physical stimulation was stopped, there was no clinical improvement in the lesion. Immediately after the incisional biopsy the girl was fed only by a nasogastric tube, and healing of the lesion was seen gradually. Therefore, we believe that the bottle feeding, in combination with irregular swallowing activity, was the stimuli for the lesion.

Incisional biopsy was performed to elucidate the nature and pathophysiology of the lesion. The differential diagnosis includes pyogenic granuloma, leishmaniasis and Traumatic Ulcerative Granuloma with Stromal Eosinophilia (TUGSE).

Pyogenic granuloma is one of the most common entities responsible for soft tissue enlargements [8]. Females are slightly more affected than males. Gingiva is the predominant site, followed by lips, the tongue, the buccal mucosa and the hard plate. Size varies from a few millimeters to several centimeters and growth is usually slow, asymptomatic and painless, but at times can be rapid. The clinical picture varies, and includes a wide array of clinical appearances, ranging from a sessile lesion to an elevated mass, or may even be ulcerated. Some investigators consider pyogenic granuloma as a "reactive" or "reparative" tumor process, which is caused by several factors such as microorganism infections (staphylococci, streptococci, botryomycosis, and fungus), hormones and minor trauma to tissues. The treatment of choice is surgical excision; and extension of the healthy tissue, curettage and removal of the suspected cause of the lesion [8,9]. The ulcerative form of pyogenic granuloma is quite rare. Therefore, we considered two other entities that are clinically more characteristic of ulcers.

Cutaneous leishmaniasis is endemic in Israel, but mucosal involvement is rare and considered as metastatic dissemination. Mucosal leishmaniasis is caused by the parasite *leishmania major*, *leishmania tropica*. It is commonly seen in immune-suppressive patients, and the diagnosis is by local biopsy of the lesion and blood testing [10,11].

Another differential diagnosis is TUGSE, which exhibits a deep pseudo invasive inflammatory reaction and is typically slow to resolve. Interestingly, many of these traumatic granulomas undergo resolution after incisional biopsy [12].

In our patient, the histological exam revealed ulcerated tissue with partially squamous epithelial coverage. Active granulation tissue in pyogenic granuloma form, along with inflammatory infiltrate with mucinous salivary glands, without signs of leishmania bodies or eosinophilic cells.

We think that the improper swallowing, in combination with bottle feeding, may have irritated the soft tissue of the mouth and caused pyogenic granuloma.

Breast and bottle feeding are known to cause various oral lesions such as Riga Fede and Bednar's aphthae, due to local irritation, high temperature of the liquid or other reasons [13]. Following the biopsy and initiation of feeding through a nasogastric tube, the lesion disappeared. This led to the presumption that bottle feeding contributed to the formation of the lesion. Unfortunately, the girl does not feed from anything other than a bottle. Therefore, she should be fed by a nasogastric tube until the lesion disappears, since the lesion reappeared and disappeared again and again during the previous year.

Our recommendation is that feeding babies with muscular perioral weakness should be performed gently. Parents, paediatricians and dentists must be aware and check the health of the oral mucosa on a regular basis.

Why this clinical report is important to paediatric dentists.

1. The report raises awareness to hypotonia of the peri oral muscles.
2. This should encourage paediatric dentists to carefully examine the oral soft tissues of hypotonic children.
3. A novel etiology is presented for lesions of the lower lip in the hypotonic child.

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