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Case Report

Lipoblastomatous Hamartoma in a Newborn

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Abstract

We present a clinical case concerning the diagnosis of Lipoblastomatous Hamartoma (LH) in otherwise healthy newborn. The patient (a three-month old female) was referred to the Federal University of Uberlandia (UFU - Minas Gerais, Brazil) - Department of Pediatric Dentistry with a rapidly-growing lesion on the tongue, which according to the mother, was present at birth and hindering breastfeeding. An intra-oral clinical examination showed a lesion (approximate diameter of 0.5 cm) on the dorsum of the tongue that was pediculated, soft, smooth, and with a color similar to that of the insertion tissue. Complete surgical excision and a subsequent biopsy were chosen given that the lesion was growing and inhibiting breastfeeding. Surgical removal was performed in an outpatient setting under local anesthesia and the specimen was sent to a laboratory for histopathological analysis. The association between clinical and histopathological findings led to a diagnosis of lipoblastomatous hamartoma. Six months of follow-up examinations showed that breastfeeding was adequate and that the surgery had healed completely with no sign of lesion recurrence. To date, the patient has not presented any abnormalities.

Keywords

Breastfeeding; Lipoblastomatous Hamartoma; Tongue

Introduction

Hamartomas are abnormal growths that are composed of a mixture of tissues and cells that are usually endogenous to the area in which the lesion develops [1-4]. These growths do not have an inflammatory or neoplastic nature, presenting self-limiting growth [5] and most of them do not regress spontaneously [6].

Hamartomas are usually diagnosed in the liver, kidneys, lungs, pancreas, spleen and, rarely, in the oral cavity [7]. Within the oral cavity, hamartomas composed of several types of tissue (smooth or skeletal muscle, adipose, vascular, lymphatic, glandular and neural) have been described [6]. These lesions are usually asymptomatic, small (less than 2 cm in diameter [8], polypoid, nodular or pediculed, and tend to occur in the posterior and median region of the tongue [2,4]. However, multiple hamartomas can be found in Cowden syndrome [5], tuberous sclerosis [9], oral-facial-digital syndrome [2] and Crohn's disease [10]. Surgical excision is not always necessary, unless the patient presents dyspnea, dysphagia, or difficulties in sucking and breastfeeding [7].

Lingual hamartomas are rarely reported and usually exist as isolated clinical cases [1-7,11]. Hemangiomas (predominance of blood vessels) and lymphangiomas (predominance of lymphatic vessels) are the most common hamartomas of the mouth [1,2,7]. Lingual hamartomas that consist predominantly

of adipose tissue are even less common. These lesions present aggregates of adipocytes, undifferentiated mesenchymal cells and an absence of inflammatory infiltrate [12]. Because its uncommon presentation in oral tissues, hamartoma composed by lipomatous tissue (adipocytes and/or lipoblasts) is not well known, differential diagnosis for lipomatous benign tumor in childhood and infancy is mandatory [13-15].

The present study reports on a clinical diagnosis of a Lipoblastomatous Hamartoma (LH) an otherwise healthy infant and a review of pertinent literature.

Case Report

A healthy three-month old female was referred to the School of Dentistry of the Federal University of Uberlandia (UFU - Minas Gerais, Brazil) with a lesion located on the dorsum of the tongue, which according to the mother, was present at birth and hindering breastfeeding. The mother claimed that the lesion continues to grow until the time of the initial evaluation.

At birth, the baby showed no signs of anoxia, cyanosis, and hemorrhage, difficulty in sucking or breathing.

At the first dental appointment, the patient was 3 months old, measured 6.125 kg and 61 cm, and had no systemic alterations. An extra-buccal physical examination showed an absence of palpable lymph nodes. However, an intra-oral clinical examination showed a lesion on the dorsum of the tongue (Figure 1) that was the same color of the insertion tissue, soft, smooth and approximately 0.5 cm in diameter. No signs of invasion (base induration), infection or trauma were observed in the surrounding tissue.

Due to the limited sucking during breastfeeding and the growth of the lesion, as reported by the mother, surgical removal was performed under local anesthesia (Alphacaine 2% ® Lidocaine with Epinephrine 1: 100,000), in an outpatient setting. Hemostasis was achieved with gauze and absorbable sutures (VICRYL®, Ethicon, Inc., SP, Brazil) and the mother was instructed on postoperative procedures.

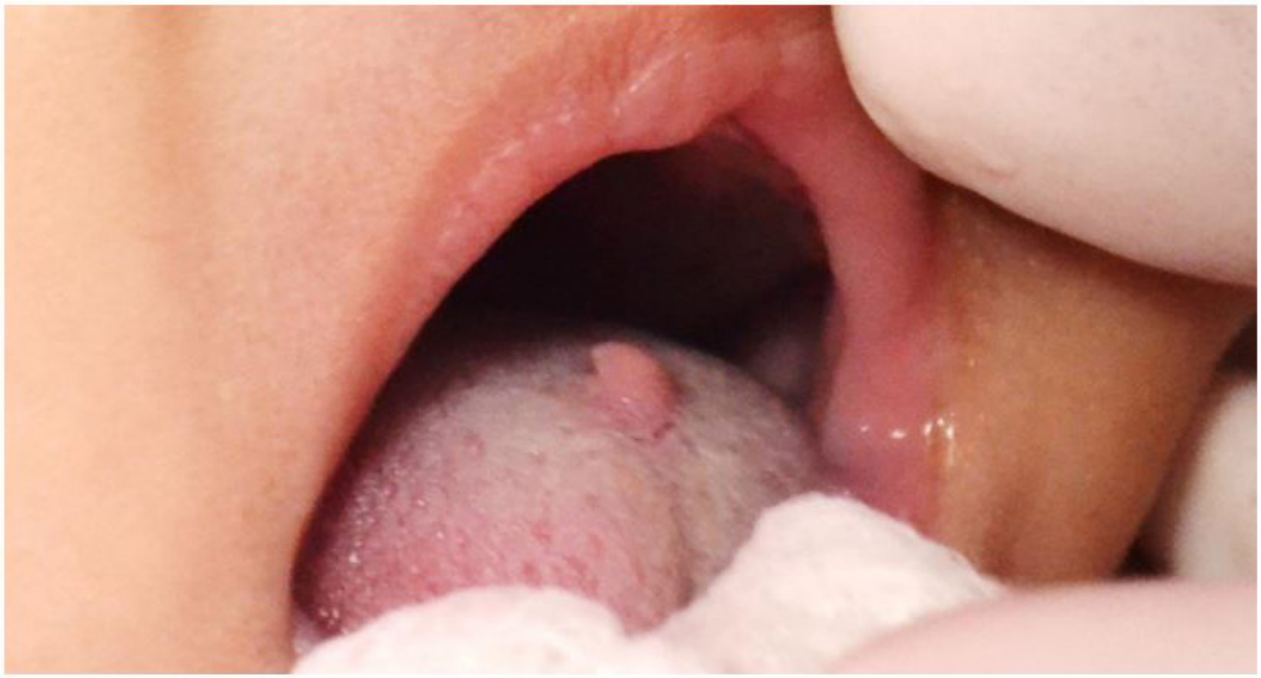

Figure 1: Initial aspect and location of the lesion.

Regarding prenatal medical history, the mother's age at gestation was 23 years and all prenatal consultations and examinations had been performed properly. During gestation, the mother experienced gestational diabetes, a thyroid disorder (hyperthyroidism) and anemia. The patient was delivered at 39 weeks via cesarean section and measured 46 cm and 2.75 kg.

Histopathologically, the tongue mucosa showed a parakeratinized stratified squamous epithelium with normal aspects. The lamina propria was composed by a dense connective tissue, blood vessels, some few thin neural fibers and skeletal muscle fibers. In the central region it was possible to observe circumscribed aggregates of cells presenting ovoid nuclei, with finely granular

chromatin and milky cytoplasm without distinct limits mixed with large cells, with clear well-defined cytoplasm limits, and picnotic eccentric nuclei, resembling adipocytes. It was not possible identify any sign of anaplasia, mitoses and invasion of these tissue or even capsule around the growth (Figure 2).

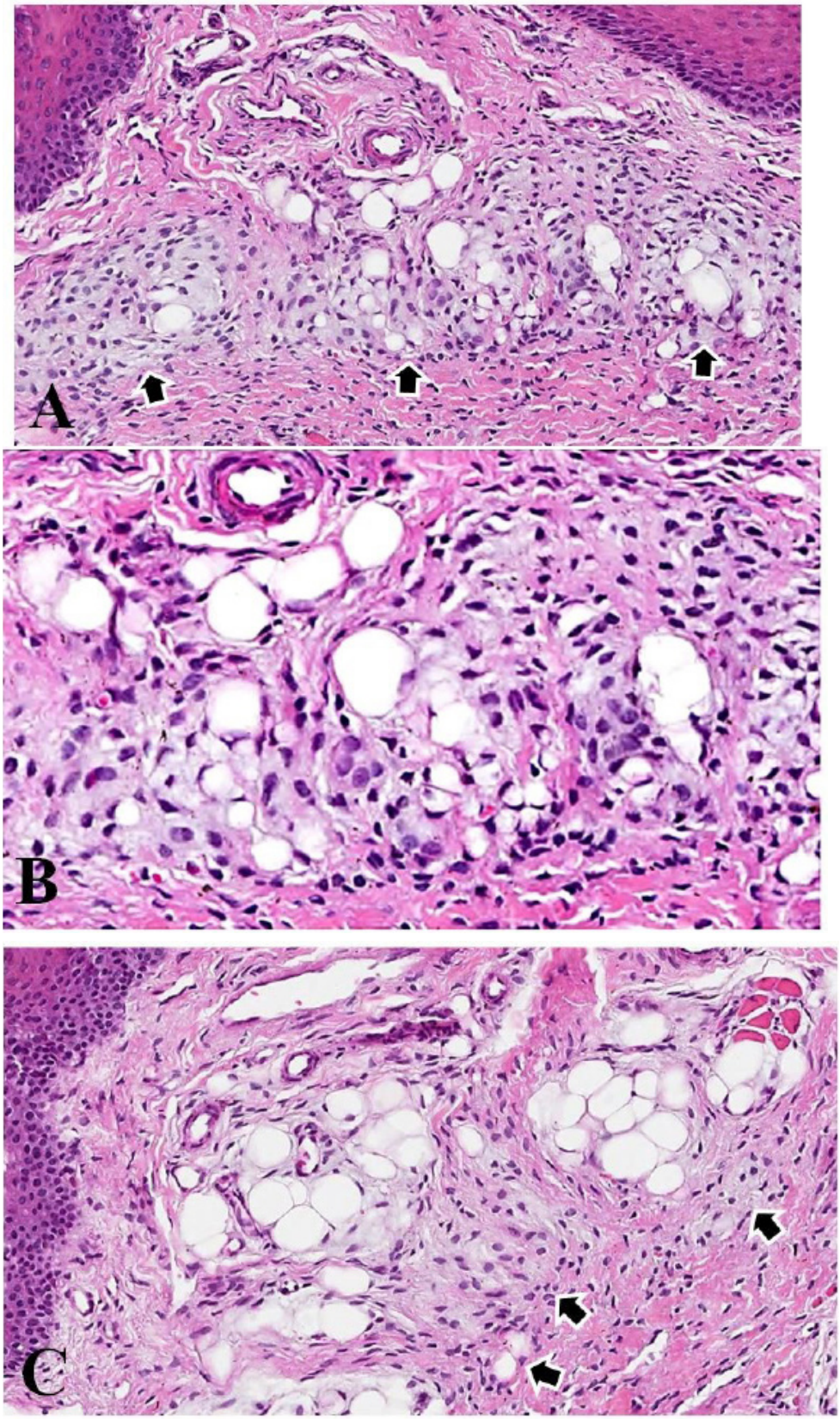

Figure 2: Histologic view of the tongue lipoblastomatous hamartoma. A, B (magnification of A) and C show representative areas of the lesion. In these areas it can be noted relatively well-delimited cellular aggregates composed of two types of cells: small cells, without apparent limits showing milky-like cytoplasm, and large and small adipocytes, surrounding by a fibrous extracellular matrix containing vessels, peripheric neural fibers and squeletic muscle.

The clinical findings and the histopathological analysis led to the final diagnosis of LH.

At six months after surgery, there was no sign of lesion recurrence (Figure 3) and breastfeeding was occurring normally. To date, no abnormalities have been observed.

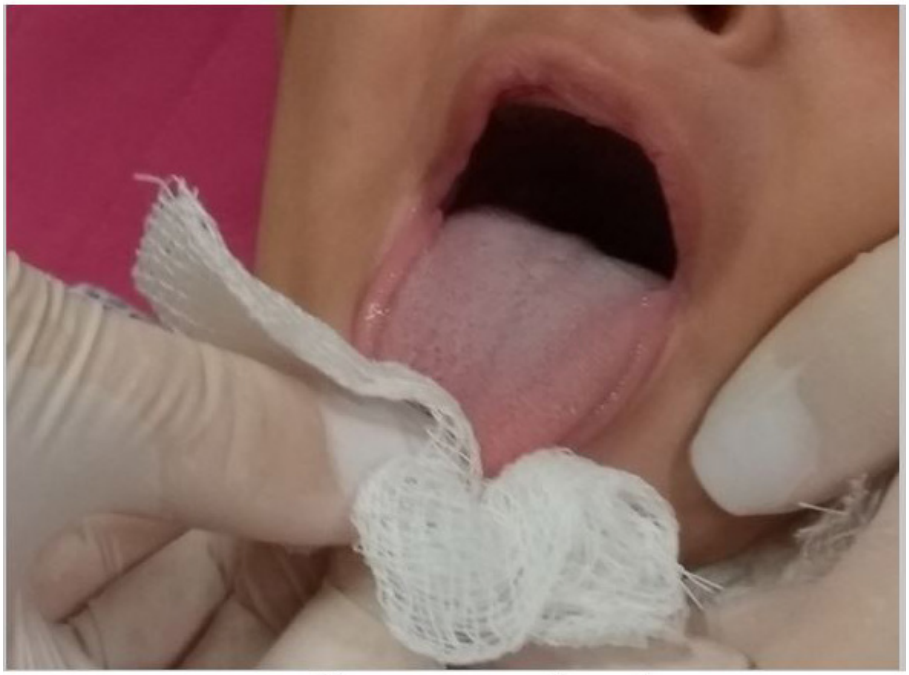

Figure 3: Dorsum of the tongue aspect, six months post-surgery.

Discussion

Distinguishing between benign neoplasms and hamartomas can be difficult. Nevertheless, medical history, clinical and histopathological findings are helpful for diagnosis. Neoplasias tend to develop later, grow autonomously and are often composed of a single tissue [3]. If benign, lesions tend to be circumscribed, frequently encapsulated, without cellular atypia and show a non-invasive growth. On the other hand, although hamartomas show a well-differentiated growth, they have a self-limit congenital growth, generally in midline regions of the body [1].

Because the varied nature of the lipomatous lesions found in childhood and infancy, the present case imposes to make a differential diagnosis to benign circumscribed and diffuse lipoblastomas, myxoid liposarcoma and well-differentiated liposarcoma [16].Tumors that are predominantly composed of adipose tissue are typically found in the superficial subcutaneous tissue of the extremities and rarely found in the orofacial region [17]. These lesions are classified by their delimitations and the cell types identified by histopathological examinations and age of diagnosis [16,18].

Benign tumor of adipose tissues have been described in infancy as early as three months-old in two distinct variants: circumscribed lipoblastoma and diffuse lipoblastomatosis. In both tumors, lipoblasts similar to those found in the embrionary adipose tissue have been recognized as the main neoplastic cell. Circumscribed lipoblastoma, the most common tumor, tends to involve superficial soft tissue and is well-encapsulated, while diffuse lipoblastomatosis have been described as an invasive form, with greater tendency to recur [19]. In head and neck region, this infiltrative growth pattern has been associated to airway obstruction and respiratory insufficiency [20]. Although the present case have lipoblast and a somewhat lobular growth pattern, we didn't find either capsule, a distinct aspect of lipoblastoma or deep subcutaneous invasion of the neoplastic cells. Moreover, the fibroblasts are not a proliferative component of the lesions since they are imersed in a mature collagenic matrix. Besides this, no histological aspect of malignant lesions as cellular atipia, mitoses, for example, could also be detected. Taking in account clinical and histopathological aspects, and because adipose tissue can be found in tongue, we believe that the present case represents, in fact, a hamartomatous swelling, in a similar presentation of that described by Zalzal et al. [12].

Most tongue hamartomas has been found in pediatric patients under two years of age being slightly frequent in males than females [4,15]. Horn et al., [21] evaluated 10 years of hospital medical records on tongue lesions in pediatric patients and found a prevalence of solid, macroglossia tumors, and only

one hamartoma. Conversely, Kaplan et al., [22] evaluated 50 hamartomas diagnosed over 12 years and inferred that these lesions may not be as rare as they are misdiagnosed. These lesions are usually located along the midline of the tongue [1], since this region is where structures are fused during embryonic development [4,7,10].

An examination of tongue lesion diagnoses from 1987 to 2005 showed that only one of 18 hamartomas was composed primarily of adipose tissue with blood vessels, mature adipocytes and salivary glands, very similar to our study [23]. Zalzal et al., [12] reported a series of cases of congenital lesions on the back of the tongue, which showed a predominance of adipose tissue and related histopathological characteristics. Although rare, it appears that hamartomatous lesions with fat containing have a limited potential of growth [12]. It presents as a sessil, asymptomatic and well-limited swelling, which is generally seen in other regions such as lungs [24], mitral valve [25,26], palm [27], perineal area [28] and pancreas [29]. Besides the rareness of this type of lesion in head and neck region, case reports with midline of tongue [12,23,30,31], buccal mucosa [32,33], palatine tonsil [34] has been already described.

No matter their indolent benign behavior, LH is a rare and poorly understood lesion. No studies were found on the etiology of this type of lesion. However, given the mother's health conditions during pregnancy (anemia, thyroid disorder and gestational diabetes), future studies should evaluate correlations between prenatal factors and the presence of oral hamartomas.

Despite of its rareness, LH should take into consideration as a differential diagnosis in fat tumors in children under 3 years old. Further, other benign and malignant lesions, congenital or not, should also be included such as other kinds of hamartomas and choristomas, benign and malignant soft tissue tumors of smooth, skeletal, vascular, lymphatic and perineural differentiation such as hemangioma, lymphangioma, cellular and cavernous hemangioma, capillary lobular hemangioma (pyogenic granuloma), reactive, pseudocystic and cystic lesions [5]. Because of this, histopathological diagnosis is obligatory, since there is no pathognomonic symptomatology associated with LH.

Congenital tongue lesions are rare as well and may hinder breastfeeding and obstruct airways [12]. In general, a conservative enucleation is sufficient and no recurrences have been reported [12,23,30,31]. The patient in the present study had difficulty sucking during breastfeeding. This limitation provided the impetus for the surgical excision of the lesion, which in turn allowed a return to normal breastfeeding. No sign of recurrence was observed.

Conclusion

A lesion was diagnosed as lipoblastomatous hamartoma on the dorsum of the tongue, which had been present since birth. The lesion was surgically removed because it was hindering the suction needed for breastfeeding. The removal of the lesion was shown to be effective in resolving the case.

Conflicts of Interest

All authors declare no conflicts of interest in this article.

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