



Giant hemangioma of the tongue: a case report of a potential complication with a real risk of death in a child

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Abstract

Objective: Hemangiomas are considered benign vascular tumors characterized by rapid proliferation of endothelial cells, comprising a space containing blood that is coated with a layer of endothelium. Hemangiomas are considered potentially serious lesions due to the risk of bleeding, airway obstruction, infection and thrombocytopenia – however reports of these complications are poorly described in the literature. The authors report a case of cavernous hemangioma of the tongue in a two-year-old child.

Case description: A child was admitted with a large volume on the tongue due to internal bleeding after trauma caused by biting the tongue, placing the patient in a life-threatening condition. Initially, a life-support treatment was performed until the child was transferred to a specialized hospital. There, she died three days after glossectomy surgery.

Conclusion: The authors report a case of a serious complication of hemangioma of the tongue that put the patient at risk of death by one of the potential complications such as airway obstruction, hypovolemic shock and compartment syndrome, infection and necrosis. The authors emphasize the importance of early diagnosis and treatment of cavernous hemangiomas in areas susceptible to trauma in order to prevent serious and potentially fatal complications.

Keywords: Hemangioma; oral pathology; stomatology; oral lesion

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Hemangioma gigante de língua: relato de caso de uma complicação potencial para um risco real de morte em uma criança

Resumo

Objetivo: Hemangiomas são considerados neoplasias vasculares benignas caracterizados por uma rápida proliferação de células endoteliais, constituídos de um espaço contendo sangue revestidos por uma camada de endotélio. Os hemangiomas são considerados lesões potencialmente graves devido ao risco de sangramento, obstrução de vias aéreas, trombocitopenia e infecção, porém relatos destas complicações são raramente descritos na literatura. Os autores relatam um caso de hemangioma cavernoso de língua em uma criança de 2 anos de idade.

Descrição do caso: Uma criança foi admitida com um grande volume na língua decorrente de sangramento interno após um trauma por mordedura da língua, colocando a paciente em um risco potencial de morte. Inicialmente foi realizado um tratamento de suporte de vida até que a criança pudesse ser encaminhada a um hospital de referência, no qual veio a óbito 3 dias após uma cirurgia de glossectomia.

Conclusão: Os autores relataram um caso no qual um hemangioma de língua evoluiu com uma complicação séria com risco de obstrução das vias aéreas, choque hipovolêmico, síndrome compartimental, infecção e necrose. Ressaltam a importância do diagnóstico e tratamento precoce de hemangiomas cavernosos em áreas susceptíveis a trauma a fim de prevenir complicações sérias e potencialmente fatais.

Palavras-chave: Hemangioma; patologia oral; estomatologia; lesão oral

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Introduction

Hemangiomas are considered benign vascular tumors characterized by rapid proliferation of endothelial cells, comprising a space containing blood that is coated with a layer of endothelium. Hemangiomas are considered potentially serious lesions due to the risk of difficult-to-control bleeding [1]; however, reports of these complications are poorly described in the literature.

The hemangioma is the most common benign tumor of vascular origin with a prevalence that ranges between 10-12% of children under 1-year old and may be present in up to 30% of newborns weighing up to 1kg at birth [2]. These lesions are three times more common in female babies and white children, and the head and neck is the most prevalent area. In this area, the most common sites are the skin of the face, oral mucosa, lips and tongue, and the size can vary from a few millimeters to several centimeters (0.25 cm^3 to 200 cm^3) [2].

Hemangiomas can be classified in different ways: the most accepted classification divides and organizes the hemangiomas into capillary, cavernous and mixed types. The capillary type is the most common and is characterized by the presence of small vessels surrounded by a connective tissue deficient in elastin, causing small, localized lesions. Cavernous hemangiomas are composed of large thin-walled vessels causing extensive infiltrated injuries. Mixed hemangiomas are described as a combination of both types, presenting vessel proliferation with defined lesions and diffuse areas, and may be found infiltrating the adjacent bone tissue [3]. Despite the high prevalence of hemangiomas in children [2], most cases are of the capillary type, a less aggressive lesion and often self-limiting. The occurrence of aggressive cavernous hemangiomas is quite rare, especially in the tongue [4].

Most cavernous hemangiomas are asymptomatic, being found incidentally by imaging studies such as ultrasound, magnetic resonance imaging (MRI), computerized tomography, and angiographic methods. Contrast examinations are quite effective in the diagnosis of cavernous hemangioma, and among these, MRI is more accepted because it is a method of high sensitivity and specificity [1].

The treatment of cavernous hemangiomas depends on the clinical changes. The risk of functional impairment, development of large lesions, and evolutionary complications are the main factors considered when determining the therapeutic option. Small lesions can be removed by conventional surgery, electrosurgery or cryosurgery [5]. In large lesions, treatment may include the use of sclerosing agents to lessen the lesion. In emergency conditions such as airway obstruction and bleeding, indication of an interventionist management-clinical or surgical-such as airway maintenance, hemostasis and surgical resection are necessary and are well established in the literature [6].

Although hemangiomas are essentially benign and often self-limiting, they may under specific conditions put the

patient in a life-threatening condition [7]. This paper reports a case of tongue cavernous hemangioma in a 2-year-old child after a local trauma that evolved with serious complications, including the risk of death.

Case Description

A 2-year-old female child was admitted to an emergency hospital with a huge swelling on the tongue (Fig. 1). The patient was referred for evaluation to the oral and maxillofacial surgery department. During the anamnesis, the child's parents reported that she was playing when she had a fall from height and bit her own tongue. From this moment came a light bleeding associated with a fast and continuous growth of the tongue. The parents had a previous MRI with contrast before trauma, on which was observed two large areas of uniform uptake of contrast medium at the base of the tongue and sublingual area measuring $3 \times 4 \text{ cm}$ in its largest section, which gave support to the clinical diagnosis of cavernous hemangioma of the tongue (Fig. 2).

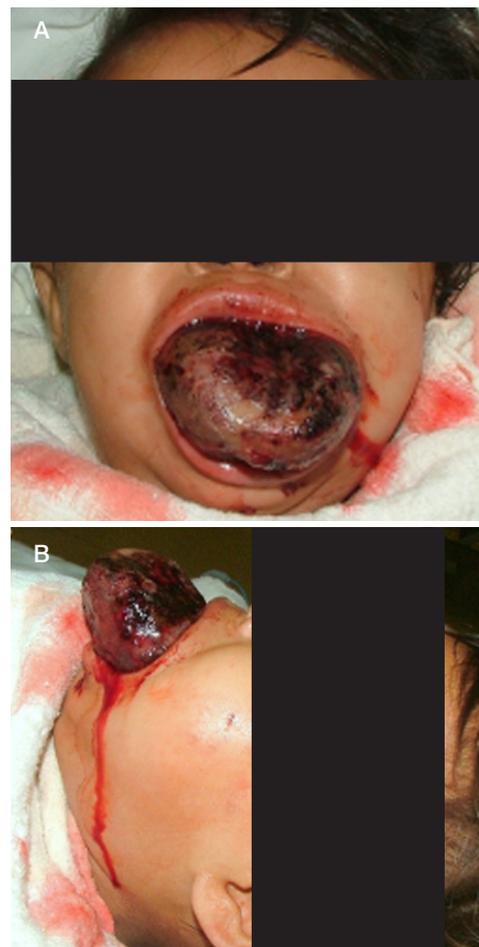


Fig. 1. Front view (A) and lateral (B) of patient in supine position. A large swelling of the tongue with protrusion and almost complete obstruction of the opening lip may be observed. When the child was placed in the supine position, the tongue was repositioned, thus blocking the airway and causing severe discomfort to the patient (A and B).

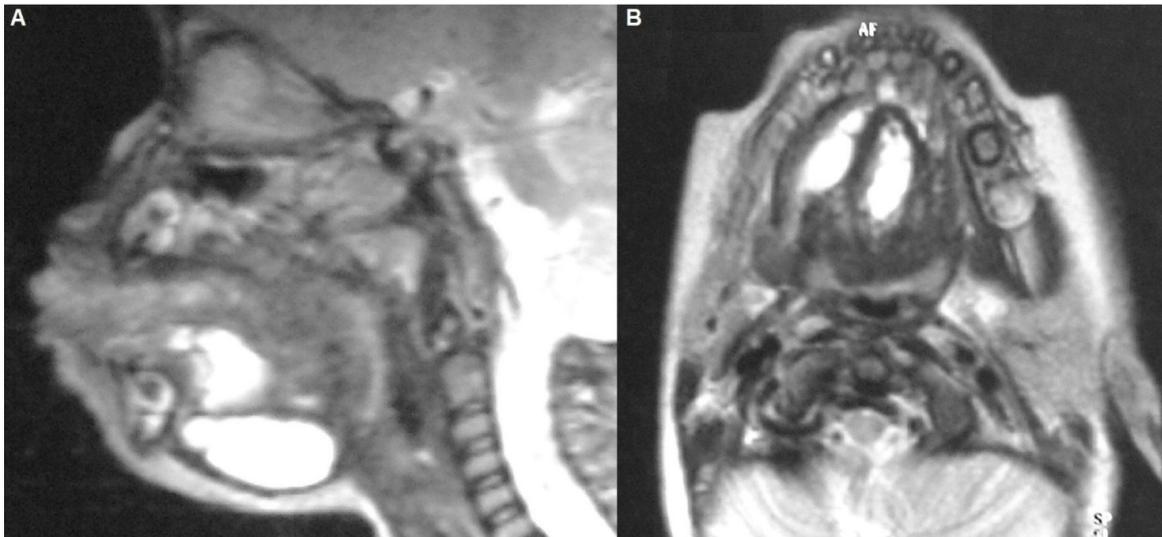


Fig. 2. MRI images with contrast in sagittal (A) and axial cuts (B) prior to the traumatic event. Two large uniform areas of contrast uptake are observed at the mouth floor and tongue base (A). In the axial section, two areas at the tongue base can be seen, located one on each side of the midline suggesting involvement of the left and right lingual arteries (B). The association of clinical and imaging features was sufficient for clinical diagnosis of tongue cavernous hemangioma.

On physical examination the following were seen: a large swelling with protruding tongue; inability to close the mouth; and airway obstruction when the patient was placed in the supine position. The tongue was hard and had a purplish/blackened color, including local pale areas suggestive of ischemia (Fig. 3).

Laboratory tests showed a significant anemia (hematocrit of 21.3% and hemoglobin concentration of 7.0 g/dL) and a platelet count within the normal range (395.000 mm³).

The child received emergency care and was transferred to the intensive care unit, and received support from oxygen facial mask, antimicrobial agents to prevent infection of the tongue, and transfusion of one unit of packed red cells.



Fig. 3. The front view of tongue in detail. A blackened/purplish tongue aspect can be observed, as well as a pale area near the apex of the right side suggesting local ischemia. The tongue general aspect suggests great distension, and the points and blackened areas indicate sites of internal bleeding.

The child was kept supine in an inclined position, a condition that allowed a more comfortable breathing and had an oxygen saturation of 98% with the oxygen mask (Fig. 4). The child showed no episodes of major bleeding in the period when she was hospitalized, and after blood transfusion there was an improvement in the anemia (hematocrit of 30.5%) and hemoglobin concentration (9.1 g/dL).

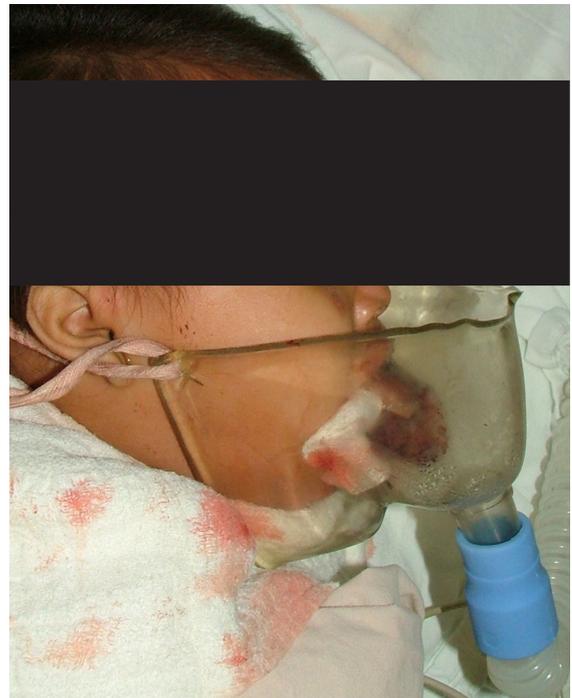


Fig. 4. Patient lateral view. The patient was placed in a supine lateral position with a facial oxygen mask. In this position, she was relatively comfortable and kept a stable oxygen saturation of 98%.

With the implemented measures, the clinical condition was stabilized and remained so until her transfer after two days of hospitalization to a cancer hospital for specialized treatment. The child underwent a risky surgery on an urgent basis, nine days after her initial hospital admission. A total glossectomy was performed, but the child died 3 days after surgery, due to an association between acute respiratory failure and pneumonia.

Discussion

Hemangiomas are common lesions, but in most cases are self-limiting [7]. However, when it manifests as cavernous type, care should be taken for potential complications such as infection, thrombocytopenia, airway obstruction, and severe bleeding [8].

Cases of giant hemangioma of the tongue are quite rare, and only a small number of cases have been reported in the literature. In these cases, the main complications encountered were related to the risk of airway obstruction, bleeding, deformity of jaw growth, and speech and swallowing difficulties [4,7]. These presented complications were responsible for conditions such as low oxygen saturation, hypovolemia and malnutrition, requiring surgical treatment. The cases were treated surgically—either by resection of the lesion [4] or by the external carotid artery ligation—in order to sharply reduce blood perfusion to the lesion [7]. Unlike the present paper, in all situations described above, lesions were found in adult patients; only one case with some similarities was observed in a very young child [4].

This case was observed in a 2-month-old baby with fast growth in the first year of life. In the course of child development, the lesion continued to grow, and due to the excessive growth of the tongue produced speech and swallowing problems, as well as a jaw growth deformity. Associated with this, the patient suffered from recurring infections caused by *Candida* and sleep problems. The patient underwent surgical resection of the lesion at 18-months old, reestablishing their speech and masticatory functions after one year postoperatively [4]. Nevertheless, all the complications presented in this case were appearing slowly and continuously, and progressed without episodes of acute emergency, as presented in this paper. This paper presented a case of a 2-year-old child, who had been diagnosed with a tongue cavernous hemangioma prior to trauma, but until then, had not been subjected to specific treatment. The child was involved in an accident with a fall from their own height, resulting in the tongue being bitten, which led to major internal bleeding and formation of a large hematoma. It was necessary to perform an emergency surgical treatment, but the child did not survive and died 3 days after the procedure. An MRI contrast examination performed prior to trauma, suggested a lesion with large blood supply, since the size of the vessels involved (3x4 cm), which increases the potential for a serious complication. The diagnosis through imaging tests is extremely important for the diagnosis of cavernous hemangioma as biopsy is not

indicated for this kind of vascular lesions due to the high bleeding risk [1].

Extravasation of blood into the region of the tongue and floor of the mouth, and a consequently local hematoma formation, were responsible for a large blood loss that was detected after laboratory tests. Regular values of hemoglobin and hematocrit in children aged 6 months to 18 years are respectively: 12-16 g/dL hb and 35-45% ht [9]. The child showed very low levels of hemoglobin (7.0 g/dL) and hematocrit (21.3%) confirmed the diagnosis of hypovolemic anemia. There was a great risk of hemodynamic compromise, which was stabilized with transfusion of packed red blood cells, which re-established new parameters of hemoglobin (9.1 g/dL) and hematocrit (30.5). Among the related complications, there is the risk of irreversible hypovolemic shock, which is often associated with losses of more than 25% of total blood volume, progressing to cellular ischemia, acidosis, cellular necrosis, and subsequent multiple organ failure [10].

Another situation that put the patient in a life-threatening condition was the possibility of airway obstruction due to tongue swelling, thus obstructing the passage of oxygen. This risk was evident when the child was placed in the supine position. There was a retroposition of the tongue, which was blocking her breathing, forcing the patient to be rested in a supine inclined position, and constant observation. At this age, the child did not have cognitive development to take a position that favors airway maintenance, and therefore remained a constant risk of being subjected to tracheal intubation or emergency tracheostomy [11]. The child initially attended a hospital trauma department, which did not have the necessary resources to perform the required treatment. Thus she waited to be transferred to a specialized center. If the child were intubated, the chances of extubation would be greatly reduced, since the prospect of clinical improvement without specific treatment of the lesion would be remote. Because of the possible complications reported, it was decided to keep the patient under constant observation. Despite the large increase of the tongue, she remained calm, with a good breathing pattern and maintained an adequate oxygen saturation (98%).

After the establishment of internal bleeding and clot packing, the tongue was quite distended and purplish. This could lead to two complicated situations: the first would be a well-known lesion in the upper and lower limbs, which is compartment syndrome. This syndrome is characterized by increased pressure in a muscle compartment that compromises the blood supply to tissues, evolving into a circulatory collapse and extensive necrosis of the area [12].

The huge tongue distension and its consequently poor perfusion could evolve with necrosis and clot depacking, consequently causing a risk of difficult-to-control bleeding. The second situation would be the potential risk of clot infection, mainly due to the etiology being a fall from own height with tongue bite. The infection risk associated with bite wounds is rather high [13], mainly in the area in which the local circulation is greatly compromised. Infectious



processes in the mouth floor are serious, as they can diffuse through facial spaces, and cause a situation similar to angina, particularly if we consider that the airways were already quite affected by the lesion [14].

The cavernous hemangioma in the tongue of the child had been previously diagnosed, but specific treatment had not yet been implemented. Selective embolization is an excellent alternative for this specific lesion [15], since neither the tongue nor the hemangioma itself would need surgery, if the patient had not developed this complication. This case emphasizes the potential complications that can be associated with cavernous hemangiomas and suggests that treatments should be performed as early as possible to avoid potentially fatal complications, as occurred in this case. The child underwent a series of complications that could put their life at risk including the risk of hypovolemic shock due to anemia, airway obstruction caused by a large swelling of the tongue, the establishment of an infectious process directly associated to the bite site, and lastly, the development of compartmental syndrome, which might evolve to extensive tissue necrosis, and still initiate severe bleeding. Eventually, the patient died due to the association of pulmonary infection and acute respiratory failure after a risky surgery. Tongue cavernous hemangiomas show a great risk for patients, whereas the tongue is an inquisitor mobile organ, which is more susceptible to trauma and subsequent complications [15].

The authors report a rare case of tongue cavernous hemangioma in a 2-year-old child, which after a local trauma developed complications including airway obstruction, hypovolemic shock, compartment syndrome, infection and necrosis, placing the patient in a framework of potential risk of death, which was confirmed three days after a surgical glossectomy. Although complications associated with hemangiomas are cited in the literature, few cases have been described of these complications. This paper demonstrates the importance of careful evaluation of cavernous hemangioma

lesions in areas susceptible to trauma, such as the tongue, and supports the idea that appropriate measures should be taken early before these lesions evolve into life-threatening conditions.

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