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A Recalcitrant Huge Pyogenic Granuloma in a Young Child Mimicking as Lingual Hemangioma

Küçük Yaştaki Bir Çocukta Lingual Hemanjiyomu Taklit Eden Tedaviye Dirençli Dev Piyojenik Granülom

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ABSTRACT

Pyogenic granuloma of the oral cavity is rare, more so over the tongue in a young child. It is a soft tissue lesion thought to be due to reactive exuberant tissue reaction to local irritation or trauma. We report a case of a recalcitrant huge pyogenic granuloma in a 3- year-old girl who had the lesion ever since she was 1 year old in which there was a recurrence after surgical excision. Radiological imaging suggested the lesion as a lingual hemangioma arising from the lingual arteries. Trial of propranolol was in vain and subsequent re-excision and avoidance of irritative factors yielded successful outcomes in the treatment of the patient.

Keywords: *Pyogenic granuloma, lingual hemangioma, child*

ÖZ

Oral kavitenin piyojenik granülomları nadir olup, küçük çocuklarda daha sıklıkla dil üzerinde görülmektedir. Lokal irritasyon veya travmaya karşı aşırı gelişen doku reaksiyonuna bağlı bir yumuşak doku lezyonudur. Bu makalede, 1 yaşından beri lezyonu olan ve cerrahi eksizyon sonrası rekürrens görülen 3 yaşındaki bir kız çocuğunda tedaviye dirençli bir dev piyojenik granülom olgusu sunmaktayız. Radyolojik görüntüleme bu lezyonu lingual arterlerden kaynaklanan bir lingual hemanjiom olarak yorumlamıştır. Tedavide propranolol denenmiş ancak etkisiz olmuştur, sonrasında re-eksizyon ve irritasyona neden olan faktörlerden uzak kalınması hastanın tedavisinde başarılı sonuçlar vermiştir.

Anahtar kelimeler: *Piyojenik granuloma, lingual hemanjiyom, çocuk hasta*

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INTRODUCTION

Pyogenic granuloma (PG) of oral cavity is a soft tissue lesion thought to be a reactive exuberant tissue reaction to local irritation or trauma. PG is commonly subdivided into lobular capillary hemangioma (LCH type) and non-LCH type. It is a common lesion occurring on the gingiva. It can occur on the palate, tongue, lips, buccal mucosa as well as other sites but they are rare. These lesions are usually seen between patients aged 11 and 40 years[1].

In this case report, a recalcitrant huge pyogenic granuloma mimicking as lingual hemangioma in a 3-year-old girl was presented.

CASE REPORT

A 3-year-old girl presented to us with tongue swelling ever since she was 1 year old. Examination revealed a 3x2cm multilobulated swelling over the middle portion of tongue with induration of its underlying muscle (Figure 1). Excision was done and histopathological examination revealed it as LCH. Unfortunately, the swelling recurred two months after excision. MRI/MRA showed a mass hypointense on T1 weighted sequence, hyperin-

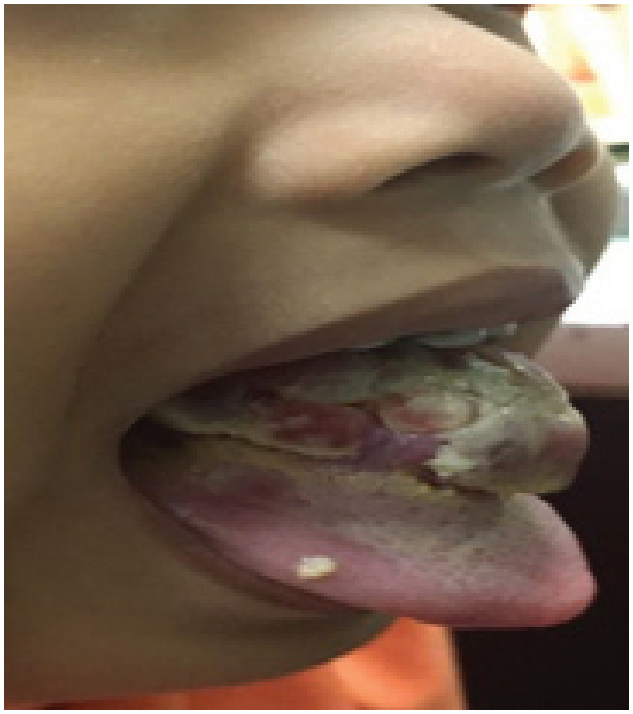

Figure 1. Multilobulated mass at the middle portion of the tongue.

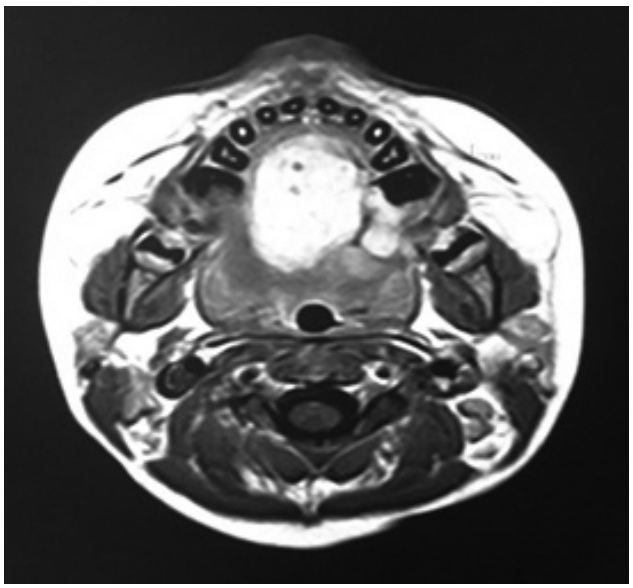

Figure 2. Hyperintense lesion with flow voids seen on MRI T2 sequence.

tense on T2 weighted sequence involving the superior longitudinal muscle of the tongue with enhancement post gadolinium. Flow voids were seen within the lesion both on T2-weighted and post gadolinium sequences (Figure 2). It was supplied by superficial branch of bilateral lingual arteries with the suspicion of feeder

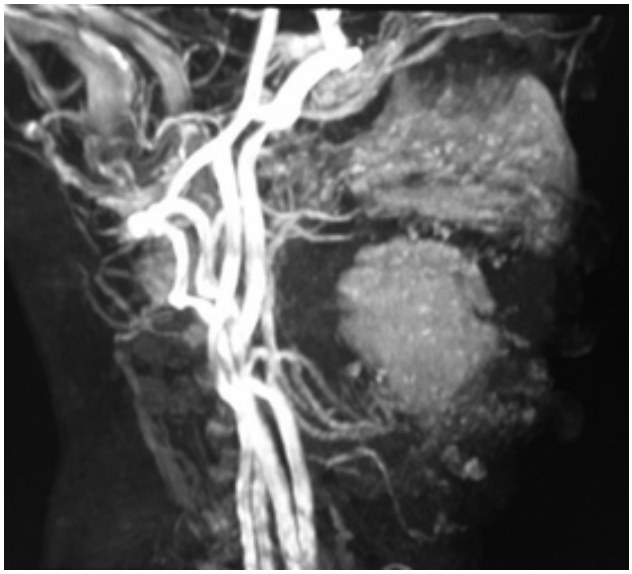

Figure 3. Feeder arteries are seen arising from the lingual arteries.

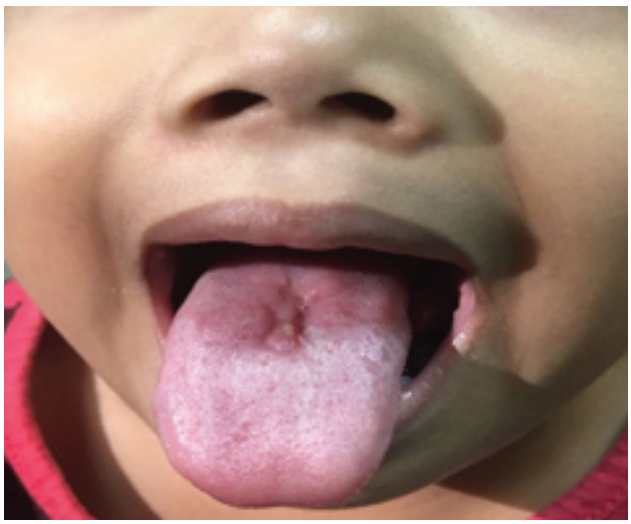

Figure 4. Healing lesion after surgical excision.

vessels also arising from deep lingual branches (Figure 3). Hence, radiological report gave an impression of lingual hemangioma arising from lingual arteries.

Her family did not prefer repeat surgery initially and a trial of oral propranolol for 10 months was given. However, as the swelling continued to increase in size causing difficulty in oral intake and the patient had episodes of bleeding, a second excision was agreed upon when she was 6 years of age. A multilobulated swelling measuring 4x3 cm was excised with monopolar diathermy with adequate resection margin including a

cuff of normal tongue musculature. As the lesion was supplied by branches of the lingual arteries, the excision was done with extreme care avoiding the location of the deep lingual branches estimated based on MRA thus avoiding the need of embolization therapy prior to surgery. Intra-operatively, there were minimal bleeding which was easily controlled and primary closure was done using sutures. Histopathological report again described the lesion as lobular capillary hemangioma showing proliferation of small blood vessels arranged in a lobular growth pattern with plump endothelial cells. The stroma was edematous with acute and chronic inflammatory infiltrates. Follow- up was performed for one and a half year after excision which did not reveal any signs of recurrence (Figure 4).

DISCUSSION

Pyogenic granuloma (PG) was first coined by Hartzell in 1904 and initially postulated to be associated with pyogenic organisms[2]. However, this term is now regarded a misnomer as PG does not contain pus and is not a granulomatous disease. PG of the oral cavity is not uncommon but only four percent of cases occur on the tongue[3,4]. Most regard PG as a benign neoplasm arising due to various stimuli including chronic local irritation, hormonal factors, trauma and certain drugs. Up to a third of these lesions occur following trauma or irritation with poor oral hygiene. PG disrupts balance between angiogenesis enhancers and inhibitors whereby there is increased levels of basic fibroblast growth factor (bFGF) and vascular endothelial growth factor (VEGF) and decreased levels of thrombospondin-1, angiostatin and estrogen receptors are believed to contribute to this condition[5].
Sternberg et al.[6] suggested three distinct phases to describe the course of pyogenic granuloma. The "early phase" reveals a dense cellular stroma and minimal lumen formation. The next phase described as "capillary phase" demonstrates lobules which are vascular and full of intraluminal red blood cells whereas non-LCH type does not show such organization and resembles granulation tissue. The final phase referred to as "involutionary phase" exhibits perilobular and intralobular fibrosis suggestive of healing phase of pyogenic granuloma. Colour of PG varies according to its vascularity during its clinical course. Early PG is usually pinkish resembling the color of normal mucosa. Mature PG is reddish to purplish because of increased vascularity. Lastly, late healing lesions are pinkish to whitish. Clinical correlation of their appearance during its various phases and their histopathological presentation is important to arrive at a diagnosis.

Histopathologically, the absence of atypical cells and bizarre vascular channels helps to differentiate PG from Kaposi's sarcoma whereas absence of any granular bacterial material differentiates it from bacillary angiomatosis[6]. During the different stages of PG, certain lesions come in as differential diagnosis. Younger lesions may be mistaken for granulation tissue, hemangioma, Kaposi's sarcoma, and bacillary angiomatosis. Older lesions can be mistaken for oral fibroma, peripheral giant cell granuloma, or peripheral ossifying fibroma as they show certain resemblances due to the presence of extensive fibrosis in the stroma. However, the increased vascular component and inflammatory infiltrate are indicative of PG and helps in differentiating it from those lesions. Capillary hemangioma can mimic PG as they have abundant proliferation of endothelial cells in their early stages and a lobular mass of well-formed capillaries in the mature phase, minus typical inflammatory feature[7,8]. Suffice to say, a good clinical observation and histopathological analysis are needed to confirm PG.

In our case, dilemma aroused as the imaging report gave the impression of lingual hemangioma as there were flow voids seen within the lesion and was supplied by the superficial branch of bilateral lingual arteries with the suspicion of feeder vessels also arising from deep lingual branches. Hemangioma is one of the most common soft tissue tumors of the head and neck, especially in a child. It is localized in the craniofacial area in 60% of the cases, but oral involvement is rare. Hemangiomas can have a variety of presentations. They can be lobulated or smooth, pedunculated or sessile and its color ranges from red purple to pinkish, blanching with pressure. Bleeding can also occur after minor trauma or even spontaneously.

A careful imaging plan is necessary if hemangiomas are suspected. Ultrasonography and magnetic resonance imaging (MRI) techniques should be the imaging of choice and for follow-up studies. MRI is the best option to evaluate the extent, volume of the lesion, and the anatomical information of the surrounding vital structures. On MRI examination, lesions are well-defined, lobulated, homogeneous, hypo-isointense at T1, and gross hyperintensity at T2-weighted sequences The lesion becomes heterogeneous with small focal areas of fat replacement during the involutional phase. Variable vascularity may be demonstrated. Extent of the lesion is well demonstrated by contrast-enhanced examination. Subsequently, the assessments should differentiate between slow- and fast-moving circulation patterns. Doppler sonography distinguishes arteriovenous malformations (AVMs) from lesions such as capillary hemangioma and venous-malformations. Size and volume of the lesion is important in hemangioma, in order to establish the growth pattern and response to therapy. Vascular pattern, including feeding arteries, draining vein, and matrix, are also of important hallmarks for the interventional radiologist, if specific therapy is envisaged.

Management of capillary hemangioma and PG varies depending on factors such as the age of patient, site of involvement, size of lesion, and its stages of growth. A wait and watch approach for spontaneous resolution is normally used for capillary hemangioma if the lesion is small and patient is asymptomatic. As for PG, if lesion is not bleeding, small and painless, routine monitoring is advised. When both PG and capillary hemangioma are large and causing symptoms, treatment is indicated. Invasive treatments for both capillary hemangioma and PG are somewhat similar regarding the treatment of choice which is being surgical excision. Ligation of vessels or embolization may need to be employed in the case of capillary hemangioma as it has higher risk of bleeding. A few newer treatment modalities are available for both types of lesions which include laser surgery with Nd:YAG, CO_2 laser, flash lamp pulsed laser, sodium tetradecyl sulphate sclerotherapy, cryosurgery with liquid nitrogen spray or cryoprobe, intralesional corticosteroids or ethanol are all shown to be effective[9,10]. In our view, the reason of recurrence after the first operation was due to inadequate resection which was done too superficially. Excision including a cuff of normal tissue is necessary to obtain a good resection margin and to avoid recurrence.

CONCLUSION

LCH can mimic lingual hemangioma and recur as a considerable size in early childhood. Surgical excision is the primary mode of treatment with the avoidance of irritative causes. Since CH and PG can mimic each other, biopsy is important for diagnosis and to exclude other serious conditions, whenever possible. Recurrence can occur in up to 16% of PG in which some of these cases requiring re-excision. It is likely that incomplete excision, failure to resolve etiologic factors or re-injury contribute to its recurrence.

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