Glomus Tumour of the Lip Mimicking Squamous Cell Carcinoma - A Rare Case Report

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INTRODUCTION

Glomus tumour is a rare soft tissue neoplasm arising from glomus body, which is in an arteriovenous anastomosis located particularly in the dermis. This tumour occurs most commonly in hands and feet, and is seldom found in other sites. The purpose of this report is to describe an unusual case of glomus tumour in the lip. A 17-year-old woman with a firm, painless and ulcerated lump in her lower lip was admitted to our clinic. Excisional biopsy was performed, and histopathological analysis revealed the lesion to be a subtype of glomus tumour called as a glomangioma.

Most glomus tumours are benign and may be treated by simple surgical excision. A typical glomus tumour of the hand is readily diagnosed, but it may occur anywhere such as oral cavity or internal organs, and its small size and atypical anatomical site presents a diagnostic dilemma. Therefore, a glomus tumour should be considered in the differential diagnosis of mass in the lips.

Glomus tumour was first mentioned by Wood as a painful subcutaneous tubercle.[1,2] It is classified as a pericystic (perivascular) tumour by the World Health Organization. Perivascular tumours are most frequently noticed in the superficial soft tissues at any age and are not seen commonly in the oral cavity. Synonyms for glomus tumour include glomangioma, glomangiomyoma, glomangiomatosis, glomangiopericytoma, and Popoff tumour.[3]

Glomus tumour is presumed to arise from glomus body, which may be defined as a special arteriovenous anastomosis located in the stratum reticular of the dermis. It is lined by smooth muscle and glomus cells.[4] The glomus body has been implicated in playing a role in thermal regulation.[5] It is ubiquitous but digits are the most common sites.

Clinically, the lesion is usually seen as a painful nodule located in the nail bed of the distal phalanges. Occurrence in the oral cavity is particularly rare. In this article, we present a rare case of glomus tumour located in the lower lip mimicking a malignant tumour.

PRESENTATION OF CASE

A 17-year-old woman presented with a three-month history of reddish and asymptomatic swelling on the right side of the lower lip, close to the midline (Figure 1). Her medical history was unremarkable. The lesion was initially started as a small acne that was noted first by the patient. It was approximately 5 cm in diameter at the time of examination. The lesion was sessile and covered with a rough, crusted, and ulcerated surface. On palpation, the lesion was painless and hemorrhagic. Its consistency was slightly firm without induration. The lower lip was swollen, and everted, but no pathologic induration was palpable. The oral mucosa of the lower lip adjacent to the lesion was hyperaemic.

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The extra oral examination revealed an enlarged, tender, and non-mobile lymph node in the right submandibular region. The clinical appearance of the lesion suggested a malignant lesion. An excisional biopsy was performed under local anaesthesia. The surgical specimen was fixed with 10 % formalin and sent for histopathological analysis. The histopathologic examination revealed ectatic vascular channels and capillary sized vessels surrounded by small clusters of glomus cells (Figure 2).

A diagnosis of glomangioma was rendered on the basis of microscopic characteristics. After surgery, the patient had an uneventful recovery with primary healing and had no evidence of recurrence over 6 months of follow-up.

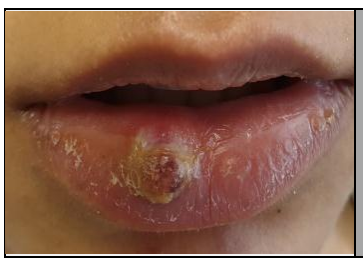

Figure 1.
The Sessile Lesion with a Rough, Crusted, and Ulcerated Surface

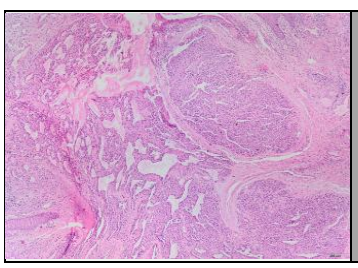

Figure 2.
Dilated and Small Caliber Vessels Surrounded by Glomus Cells H.E. x100

DISCUSSION

Glomus tumour accounts for 1.6 % of all soft tissue tumours of the extremities and is usually less than 1 cm in diameter.[6] It is a result of hyperplasia of one or more normal parts of the glomus body.[4] Patients typically complain of pain which occurs as a result of contraction of the glomus cells.[7] There are significant variations in the location of the tumour. It is most commonly situated in hands and feet, but is also found in a variety of organs, which do not have glomus body, such as trachea, liver, lungs and stomach.[8-11] Furthermore, intraosseous lesions have been found in the distal extremities.[12,13] In oral cavity, rare cases have been reported in hard palate and mandible.[14,15] Women are affected more commonly than men.[7]

In most cases, the lesion is solitary, but occasionally multiple lesions may arise.[16] Solitary lesions are encapsulated and usually subungual, whereas multiple lesions are unencapsulated, and rarely subungual, asymptomatic, and present in earlier ages with slight predominance in men.[16] In addition, familial cases demonstrating autosomal dominant inheritance have been reported.[17,18]

The cause of the glomus tumour is unknown, but trauma has been suggested as a cause. One theory holds that glomocytic differentiation may occur from perivascular cells in the tumour area.[5] This theory explains the fact that tumour may occur in the exceptional areas.

Histopathologically, glomus tumour is composed of cells resembling the modified smooth muscle or glomus cells of the normal glomus body.[5] WHO in 2018 classification of skin tumours divide these myopericytic tumour as: glomus tumour, glomuvenous malformation, glomus tumour of uncertain malignant potential and malignant glomus tumour. Solid glomus tumours account for 75 % of cases and are comprised of small cuffs of glomus cells around capillary sized vessels. Glomuvenous malformations account 20 % of cases and are characterized by dilated vascular channels with intervening small cluster of glomus cells. This type of glomus tumours are also known as glomangiomas. They most frequently occur in atypical anatomic sites and may also contain spindle cells resembling smooth muscle (glomangiomyoma). The number of glomus cells outside the endothelial lining varies from one to several layers, while the stroma is collagenous. In the present case, histopathological features of glomangioma was observed accordingly. A patient can also show multiple histologic tumours at the same time.[16]

Glomus tumour is a rare neoplasm and involvement of the oral cavity is even rarer. Including this case, there are only 14 recorded labial glomus tumours, nine of which were in the upper lip and five in the lower.[19] The tumour generally presents as a slowly growing, painful swelling in the distal extremities. The case reported here is unusual for the absence of these symptoms, central ulceration and site of the lesion.

A combination of magnetic resonance imaging (MRI) and clinical examination is used to support the diagnosis. MRI of glomus tumours show the lesions to have an intermediate-low signal on T1 weighted spin-echo images, high signal intensity on T2 weighted spin-echo images, and homogeneously avid enhancement after administration of the contrast agent (Gadolinium).[20] However, MRI is not always reliable, especially if the lesion is small and situated in the soft tissue.[7,21] Depending on the small size of the tumour, problems can arise in clinical diagnosis, and it may be easily misdiagnosed. Small sized lesions on the lips are generally seen as an asymptomatic nodule and clinical differential diagnosis might include benign or malign minor salivary gland tumour, reactive salivary lesions and benign mesenchymal neoplasms of different origin. As the lesion progressively increase in size, the clinician is confronted with sorting through a variety of malign pathologic entities. The clinical characteristics of the present case such as non-healing and central ulceration are more consistent with a malign condition.

Glomus tumour is generally benign and easily treated with excision, recurrence after removal is uncommon; however, it may be seen in deeply-located lesions and re-excision is enough.[3,21] Therefore, it may be wise to follow patients who have been treated for the condition with periodic evaluations. Malignant transformation is reported in exceptionally rare cases and wide local excision has been recommended as adequate treatment based on few case reports because of its rarity.[9,22]

This case demonstrates that the glomus tumour can develop as an aggressive and ulcerated lesion mimicking a malignant tumour. Unusual cases of glomus tumour leading to a diagnostic enigma may pose a treatment challenge.

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