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TITLE: Glomus tumour: An unusual cause of a lump in the upper lip

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ABSTRACT

Introduction

Glomus tumours are rare, benign vascular neoplasms seen most often in the nailbeds of the hands. We report a rare case of a glomus tumour presenting as a painless lump in the upper lip.

Case Report

A 73 year old gentleman was referred with a 3 month history of a soft, round, painless lump of around 1cm, within the labial mucosa adjacent to his upper right central incisor. The patient had a history of adenocarcinoma of the lung, managed successfully with upper lobe resection and chemotherapy. He had peripheral vascular disease and COPD. A differential diagnoses included a mucocele, haemangioma or salivary gland neoplasia. Histology of the excisional biopsy showed benign spindle cells and thin walled blood vessels favouring a glomus tumour. On review the biopsy site was well healed with no sign of recurrence. Unfortunately, further follow up was declined by the patient.

Conclusion

Glomus tumours arise from glomus bodies, which are arteriovenous anastomosis involved in thermoregulation. They are not to be confused with carotid body tumours, which historically shared the same name. Glomus tumours are classically found in the digits in the hands where greatest numbers of glomus bodies exist. They are rare in the oral cavity, with few reported cases. Age distribution is similar between tumours of the hand and oral tissues, however the strong female predilection seen in subungual tumours is not mirrored in oral lesions. Recurrence is seen in up to a third of digital tumours, although there is no available data on recurrence rates in oral tumours.

Keywords: Glomus Tumour, Lip, Oral Surgery, Vascular Neoplasms, Oral Mucosa, Biopsy
**INTRODUCTION**

Glomus tumours are rare, benign neoplasms usually arising from glomus bodies found in the nailbeds of the digits. They should not be confused with paragangliomas, or carotid body tumours, which are neuroendocrine neoplasms which have historically shared the name “glomus tumour” [1]. Glomus bodies are arteriovenous anastomoses, which play a role in thermoregulation through the shunting of blood in response to changes in temperature [2]. Whilst these tumours do occur throughout the body, they remain rare in the oral region. We report a case of a glomus tumour presenting as a painless lump in the upper lip.

**CASE REPORT**

A 73 year old gentleman was referred by his general medical practitioner with a 3 month history of a painless lump on the upper left labial mucosa. On presentation the lesion was around 1cm in diameter, soft, round and freely mobile. There was no evidence of bleeding or ulceration and the patient reported a gradual increase in size over the preceding weeks.

The patient had a history of T2bN1 adenocarcinoma of the lung 3 years previously. This was managed successfully with a right upper lobe resection and four cycles of vinorelbine/cisplatin. He was in remission and under six monthly review with an oncologist. He also had a history of peripheral vascular disease and chronic obstructive pulmonary disease. Medications included Atorvastatin, Clopidogrel, Tiotropium bromide, Fluticasone and Salbutamol.

The patient was an ex-smoker, having smoked heavily up until 5 years previously. He drank alcohol only occasionally.

Differential diagnosis included; mucocele, haemangioma or a salivary gland neoplasm. An excisional biopsy was undertaken under local anaesthesia where the lesion was excised intact.

Histological examination of the specimen revealed oral mucous membrane, with underlying stroma showing a well-defined lesion (Figure 1). CD31 was positive, highlighting thin walled blood vessels. The lesion was positive for smooth muscle actin (Figure 2) and negative for S100. There was no atypia or malignancy, with features and immunostaining favouring a benign glomus tumour.
On review at eight weeks post-surgery, the patient had no complaints and the biopsy site was well healed, with only a small amount of scarring remaining. The patient was advised of the benign histology but uncertain prognosis in terms of recurrence. Unfortunately, the patient refused any further follow up.

**DISCUSSION**

Glomus tumours can be subcategorised into; solid glomus tumours, glomangiomas and glomangiomyomas, depending on the relative proportions of glomus cells, vascular structures or smooth muscle within histological samples. Solid glomus tumors are most common, making up 75% of cases. Malignant glomus tumours, known as glomangiosarcomas, are exceedingly rare, with fewer than 22 reported prior to the year 2000 [3].

These tumours are usually found in the hands, where there are the highest numbers of glomus bodies. Symptoms classically involve a triad of well localised stabbing pain, tenderness of the fingertips, and an intolerance to cold stimuli [4]. Glomus tumours can affect a wide range of sites, occurring throughout the body, usually in skin or superficial soft tissue [3]. They are more rarely found in deep visceral locations and along the gastrointestinal and genitourinary tracts [5]. Aetiology is uncertain, however familial variants of multiple glomus tumours have been identified [6]. There is also a link between subungual tumours and neurofibromatosis type 1 [7, 8].

Whilst uncommon, glomus tumours of the hands are seen relatively frequently when compared to those found in the oral region. A recent review found only 22 cases of glomus tumours affecting oral tissues, the first case being reported in 1943 [9]. A PubMed/Medline search confirmed this case to be one of 8 labial glomus tumours reported in the English literature [9-16].

Glomus tumours are reported to present most commonly in the fourth to sixth decades of life [5], a finding mirrored in labial tumours, which had an average age of 58 on presentation [9]. In the subungual region there is a strong female predilection, however there is an even distribution between sexes at other sites [3], and 55% of labial glomus tumours have occurred in males [9]. In contrast to glomus tumours presenting at other sites, where pain is a frequent presenting symptom [4], none of
the reported cases of labial tumours involved a painful history. This makes tumours
difficult to distinguish from other, more common painless swellings of the lips.
Management of these tumours is surgical, with recurrence rates in tumours of the
digits reported to be in the range of 4-15% [17]. There is insufficient data available
from previously reported cases to comment on recurrence rates in labial glomus
tumours. However, the easily accessible nature of the lips makes routine follow up
by general dentists or the patient themselves simple.

CONCLUSION
In conclusion, glomus tumours affecting the labial mucosa are rare, but should be
considered in a differential diagnosis of small, painless swellings of the oral cavity.
Solitary glomus tumours of the oral cavity are rarely reported, and we hope this case
increases awareness of such lesions to clinicians.

CONFLICT OF INTEREST
The authors declare that they have no conflicts of interest.

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Liam Monaghan
Group 1- Conception and design, Acquisition of data, Analysis and interpretation of
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Group 2- Drafting the article, Critical revision of the article
Group 3- Final approval of the version to be published

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Group 2- Drafting the article, Critical revision of the article
Group 3- Final approval of the version to be published
REFERENCES


FIGURE LEGENDS

Figure 1: X10 image of haematoxylin-eosin stained tumour

Figure 2: Positive smooth muscle actin stain of tumour cells

FIGURES

Figure 1: x10 image of haematoxylin-eosin stained tumour
Figure 2: Positive smooth muscle actin stain of tumour cells