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A Rare Case of Glomus Tumor on the Mucosal Surface of Lower Lip

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Abstract

Glomus tumors are mesenchymal neoplasms derived from glomus bodies with rare presentations in the oral cavity. Glomus tumors present as a purple or pink vascular nodule or papule, sized <1 cm, and imitate vascular neoplasms such as hemangiopericytoma or hemangioma. Glomus tumors represent less than 2% of all benign soft tissue tumors. Only 27 cases of benign glomus tumors with oral cavity involvement have been reported to date. The most-reported oral tumors involved the lips (54.2%), followed by hard palate, gingiva, tongue, and buccal mucosa. The mean age of presentation of the labial glomus tumors is 48.7 years, with no gender predilection, in contrast to the subungual site, which occurs more in females. The etiology of the glomus tumors remains unknown. Subungual glomus tumors present as stabbing pain, cold intolerance, and tenderness of the fingertips, whereas labial glomus tumors mostly present as a painless, small, and slow-growing lesion. Treatment is surgical resection of the tumor. The recurrence rate of labial glomus tumors is unclear. In this article, we present the case of a 62-year-old man with a 2-month history of painless, soft lump on the mucosal surface of the lower left lip. Excisional resection of the tumor was performed in the clinic, and the histopathologic finding was consistent with solid glomus tumor. At 1 year follow-up there was no recurrence.

Keywords

glomus tumor, oral cavity, labial glomus tumor

Introduction

Glomus tumors are mesenchymal neoplasms derived from glomus bodies. The glomus bodies are located between the venous and arterial systems in the subungual dermis of the digits.[1] The role of glomus bodies is thermoregulation in response to temperature changes.[2] Glomus tumors present as a purple or pink vascular nodule or papule that can imitate a vascular neoplasm such as hemangiopericytoma or hemangioma.[3] Initially, glomus tumors were considered as a variant of angiosarcoma. In 1924, Masson published his findings that revealed that glomus tumors are histologically similar to smooth muscle cells of the normal glomus bodies. Since then glomus tumors are considered under the category of smooth muscle tumors.[4] The occurrence of glomus tumors in the oral cavity is rare. In this article, we present a case with a 2-month history of a 1 cm lesion on the lower-left labial mucosa of the lip.

Case Presentation

A 62-year-old man presented to the dental clinic with a 2-month history of painless, round, and nonerosive lump on the inner surface of the lower left lip. The patient reported that he first noticed a small lump on the lip, which gradually increased in size up to 1 cm. He denied any history of trauma, ulceration, drainage, and bleeding. On examination, a 1-cm round, nontender, and mobile lump on the lower left labial mucosa was observed. The remainder of the physical examination and review of systems were unremarkable. His past medical history included diabetes mellitus type 2. Family history was unremarkable for any type of malignancy in his first-degree relatives. Medication history included metformin and aspirin. He was a former smoker, half a pack per day for 12 years. He quit smoking 6 years ago when he was diagnosed with diabetes. He denied alcohol and drug use. The patient consented for an excisional biopsy. The lesion was excised under local anesthesia in the clinic. Histopathologic examination revealed submucosal proliferation of monotonous, bland compact epithelioid cells arranged in sheets, and punctuated by blood vessels suggesting glomus tumors.

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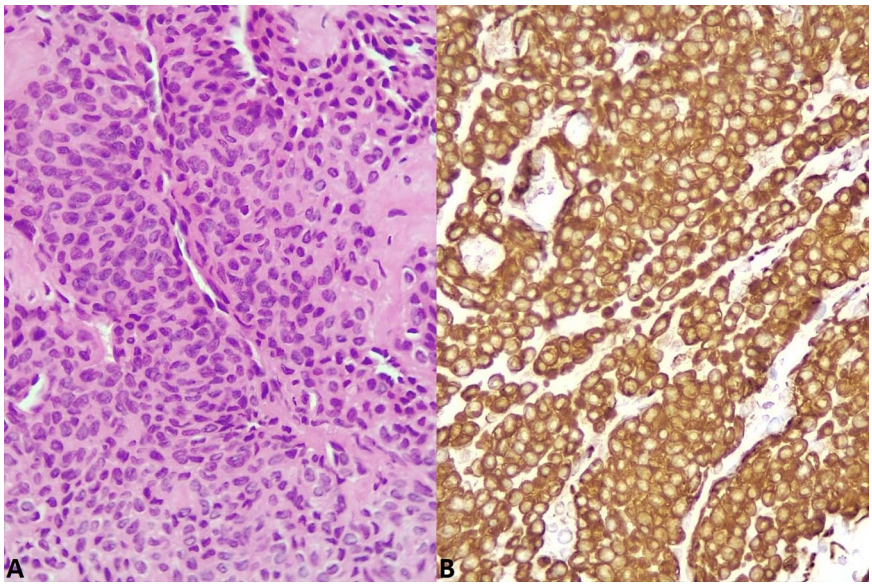

Figure 1. (A) Tumor cells in a sheet-like pattern, showing round to oval nuclei with eosinophilic cytoplasm and no obvious atypia or mitosis (hematoxylin and eosin 400×). (B) Tumor cells are strongly positive with smooth muscle actin immunostaining (400×).

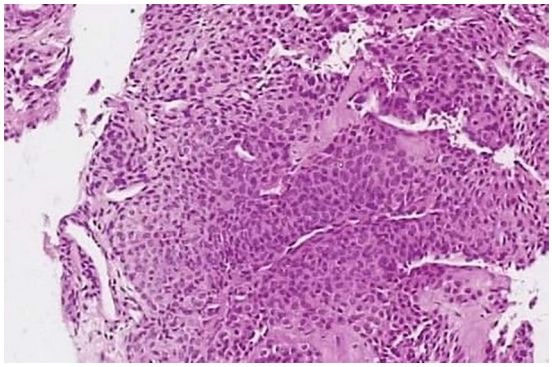

Figure 2. Lower magnification demonstrates a sheet-like pattern of monotonous round-avoid tumor cells (hematoxylin and eosin, 100×).

There was no atypia or malignant cells (Figures 1A and 2). Immunostaining with smooth muscle actin was diffusely positive, which supported the diagnosis of solid glomus tumors (Figure 1B). The patient followed up with us after 1 year and there was no recurrence of the tumor.

Discussion

Glomus tumors are rare mesenchymal neoplasm, which usually present in subungual areas of the toes and fingers. Glomus tumors represent <2% of all benign soft tissue tumors.[5] Extradigital presentations of glomus tumors are very rare; less than 1% of glomus tumors have been found on the head region.[6] The most-reported sites of oral glomus tumors are lips (54.2%), followed by hard palate, gingiva, tongue, and buccal mucosa.[7] Few cases have been reported with the involvement of the lung, trachea, heart, uterus, and stomach.[8]

Glomus tumors are subcategorized as solid (75% of cases), glomangiomas, with vascular predominance (20%) and glomangiomyomas, with smooth muscle cell predominance (5%), depending on the proportion of glomus cells, blood vessels, and smooth muscle.[9]

The size of glomus tumors in the dermis or subcutaneous tissues of extremities is usually <1 cm.[9] However, the size of glomus tumors in the head and neck area are larger and averaged 1 to 1.5 cm.[6] There are no data suggesting that tumor size influences the patients' prognosis.[3]

Few cases of malignant glomus tumors have been reported. A malignant variant of glomus tumors is glomangiosarcoma. Malignancies in the head and neck area are very rare.[10] Spector et al[11] reported a case with glomangiosarcoma in the head and neck with intracranial metastasis. Kreutz et al[12] published a case with glomangiosarcoma metastasis to the jaw from a distant location. The diagnosis of malignant glomus tumors is based on histopathological examination. Tumors with a size of more than 2 cm, moderate-to-high nuclear grade, and atypical mitotic figure more than 5 per 50 HPF (high-power field) will be considered highly suspicious for malignancy.[7]

Folpe et al in 2001 after analysis of 52 cases of glomus tumors and combining the histological features proposed

Table 1. Characteristics of cases with benign oral glomus tumors.

Case	Year	Author	Age (years)	Gender	Location	Symptoms	Size (mm)
1	1954	King[16]	32	Male	Gingiva	Tenderness	6
2	1965	Harris and Griffin[17]	35	Female	Periodontium	Pain	5
3	1967	Sidhu[18]	10	Female	Hard palate	Unknown	Unknown
4	1976	Charles[19]	17	Female	Hard palate	No	Unknown
5	1979	Sato et al[20]	29	Male	Tongue	No	3
6	1981	Tajima et al[21]	63	Female	Tongue	No	Unknown
7	1985	Saku et al[22]	45	Male	Buccal mucosa	No	45
8	1986	Ficarra et al[23]	51	Female	Upper lip (mucosa)	No	20
9	1986	Moody et al[24]	65	Female	Upper lip	No	10
10	1987	Stajcic and Bojic[25]	55	Male	Tongue	Unknown	Unknown
11	1992	Geraghty et al[26]	71	Male	Hard palate	No	15
12	1995	Kusama et al[27]	57	M	Upper lip (mucosa)	Tenderness	Unknown
13	1996	Savaci et al[28]	55	Female	Buccal mucosa	Pain	10
14	1997	Sakashita et al[29]	54	Male	Upper lip (mucosa)	No	12
15	2000	Yu et al[30]	54	Female	Face, lower lip, buccal mucosa	No	Unknown
16	2001	Kessaris et al[3]	46	Female	Hard palate	No	18
17	2004	Rallis et al[31]	85	Female	Upper lip (mucosa)	Pain	13
18	2005	Lanza et al[32]	65	Male	Lower lip	Unknown	Unknown
19	2008	Ide et al[33]	57	Male	Upper lip	Unknown	8
20	2008	Ide et al[33]	54	Male	Upper lip	Unknown	12
21	2010	Boros et al[10]	34	Male	Lower lip (mucosa)	No	15
22	2010	Dérand et al[34]	11	Female	Lowe lip (vermilion)	No	3
23	2018	Vasconcelos et al[35]	67	Female	Upper lip (mucosa)	Pain	10
24	2018	Sánchez-Romero et al[36]	51	Female	Upper lip (mucosa)	Pain	10
25	2018	Smith et al[37]	26	Male	Lower lip	Pain	15
26	2018	Smith et al[37]	58	Female	Tongue	No	20
27	2018	Zou et al[38]	24	Male	The floor of the mouth	Pain	28
28	2019	Current case	62	Male	Lower Lip (mucosa)	No	10

classification of glomus tumors with atypical features: malignant glomus tumors (large size and deep location, marked atypia with mitotic activity, as glomangiosarcoma), symplastic glomus tumor (only nuclear atypia), glomus tumor of uncertain malignant potential (superficial location with high-mitotic activity or large size only or deep location only), and glomangiomatosis (histological features of benign glomus tumor with diffuse growth).[7]

The etiology of the glomus tumors remains unclear; however, familial glomus tumors found to have an autosomal dominant pattern with incomplete penetration.[13] Yoo et al[14] reported a case of double glomus tumors in the submandibular and parotid regions. Sixty percent of patients with multiple glomus tumors have had a positive family history, supporting the concept that this abnormality is an inherited disease.[15] The first case of the glomus tumor of the oral cavity was reported in 1943. To the best of our knowledge, only 27 cases of benign glomus tumor with oral involvement have been reported until now, in a review of the English-language literature (Table 1). Rajendran et al[39] reported a first intraoral case of glomangiosarcoma in English literature: a 51-year-old male with a glomangiosarcoma of tongue in size of 27 mm.

Subungual glomus tumors present as stabbing pain, cold intolerance, and tenderness of the fingertips, whereas labial glomus tumors mostly present as a painless, small, slow-growing lesion.[12,40] Glomus tumors usually appear in patients aged 40 to 70 years old.[41] Data show that the average age of presentation of the labial tumors is 48.7 years without sex predilection in contrast to the subungual site, which occurs more in females. However, other regions have shown even distribution between sexes.[10,42] Treatment is surgical resection of the tumor. The recurrence rate of subungual tumors is 4% to 15%, whereas there are insufficient documents to estimate the recurrence rate of labial glomus tumors.[43,44]

Conclusion

Labial glomus tumors have different clinical presentations compared to subungual glomus tumors. This difference in clinical presentation makes it difficult for clinicians to differentiate these tumors from other more common painless lesions of the lip. Glomus tumors should be in the differential diagnosis for well-circumscribed, nontender lesions in the oral cavity. We hope this article would help clinicians to be more aware of this rare disease.

Declaration of Conflicting Interests

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Ethics Approval

Our institution does not require ethical approval for reporting individual cases or case series.

Informed Consent

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