Cavernous Haemangioma of the Submandibular Triangle Eluding the Gland – A Case Report

Sujeeth Kumar Shetty, Revati Kale

Department of Oral and Maxillofacial Surgery, JSS Dental College and Hospital, JSS Academy of Higher Education and Research, Mysore, Karnataka, India

Abstract

Rationale: Haemangiomas are benign vascular malformations. They are common neoplasms of infancy but seldom manifest at birth. Although common in the head & neck region, they are rare in the submandibular region. Despite being benign, surgical treatment is necessary if it causes significant functional or aesthetic compromise. **Patient Concerns:** The patient's primary complaint was swelling in the submandibular region. **Diagnosis:** Initial examination and imaging studies were suggestive of sialolithiasis due to the presence of multiple calcifications. Ultrasonographic and magnetic resonance imaging were suggestive of haemangioma with multiple phleboliths. **Treatment:** Excisional biopsy was done without compromising the submandibular gland. A histopathological examination was done post-operatively to confirm the diagnosis. **Outcomes:** Post-operative recovery was uneventful and 6-month follow-up showed no recurrence. **Take-away Lesson:** This case highlights the importance of considering haemangioma in the differential diagnosis of submandibular region masses and emphasises the preservation of the submandibular gland when the extent of the lesion permits.

Keywords: Haemangioma, magnetic resonance imaging, phlebolith, submandibular, vascular malformation

Introduction

Benign proliferation of endothelial cells causes vascular malformations, with haemangioma being one such lesion.[1] Haemangioma is a common soft-tissue tumour, predominantly observed in the head & neck region. Although most common in infants, haemangioma does not present itself at birth; instead, it develops in phases, namely proliferative, involution and involuted. The clinical presentation may be in the form of a red macule, papule or nodule, depending on the degree of congestion and depth of the lesion.[1] Haemangiomas with changes in blood flow dynamics cause thrombus formation which may calcify, forming phleboliths.[2] Such lesions in areas of salivary glands, radiologically mimic sialolithiasis.[3]

Haemangiomas can be cavernous, venous, capillary or racemose. Dilated vessels are seen in both venous and cavernous types; however, the vessels in the venous variant have thick and fibrous walls with smooth muscles, whereas the cavernous variant has thin-walled spaces lined with endothelium.[4] According to the International Society for the Study of Vascular Anomalies (ISSVA), vascular anomalies are grouped into two types–Endothelial proliferative vascular tumours and vascular malformations due to structural abnormalities. As per this classification, a lesion previously deemed as 'cavernous haemangioma' is considered 'low flow vascular malformation'.[5] Haemangioma of the craniofacial region is relatively common; however, it is rare in the submandibular triangle. Here, a case of cavernous haemangioma with phleboliths in the submandibular triangle is presented.

Case Report

A 28-year-old female presented to the Outpatient Department of Oral and Maxillofacial Surgery at JSS Dental College & Hospital, with swelling over the right lower third of her

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Address for correspondence: Dr. Revati Kale, Department of Oral and Maxillofacial Surgery, JSS Dental College and Hospital, JSS Academy of Higher Education and Research, Mysore - 570 015, Karnataka, India.
E-mail: revati1507@gmail.com

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face for one year. The swelling was insidious in onset with a gradual increase in size, with no change in size during meals. On examination, a 3 cm × 3 cm diffuse swelling was seen over the right submandibular region. The swelling was tender and firm in consistency with palpable and tender right submandibular lymph nodes. A panoramic radiograph revealed multiple calcific foci in the right submandibular region, mimicking sialolithiasis [Figure 1]. Ultrasonography showed a heteroechoic mass with multiple calcifications, lateral to the submandibular salivary gland, with no obvious duct dilation of the gland [Figure 2]. Magnetic resonance imaging showed a hyperintense lesion with heterogeneous post-contrast enhancement and multiple calcifications within the lesion [Figure 3], suggestive of vascular malformation or haemangioma along with enlarged submandibular lymph nodes. The patient was planned for excision of the lesion under general anaesthesia after a routine pre-anaesthetic workup.

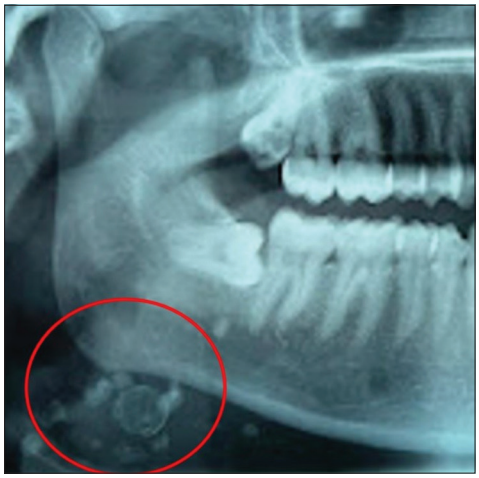

Figure 1: Panoramic radiograph with circle showing multiple radiopaque foci in the right submandibular region

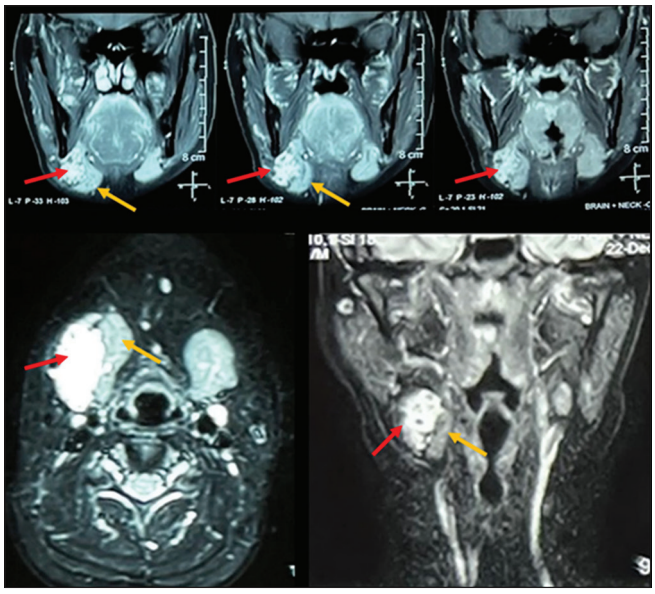

Figure 3: Magnetic resonance imaging showing lesion posterolateral to the gland. Red arrow represents lesion and orange arrow represents submandibular gland

A submandibular incision was used, and blunt dissection was carried out through the subcutaneous fat, fascia and platysma muscle was dissected to expose the lesion [Figure 4]. Careful identification and ligation of feeder vessels were done to prevent excessive bleeding. On exposure, the lesion was found to be wellcircumscribed, which facilitated the identification of the boundaries of the lesion to avoid damage to the adjacent submandibular gland [Figure 5]. The submandibular lymph node was also excised along with the lesion. The excised sample was sent for histopathological evaluation.

On gross examination, the specimen measured approximately 2.7 cm × 2.5 cm × 1.5 cm [Figure 6]. On microscopic examination, fibrocollagenous tissue was noted with dilated and congested blood vessels [Figure 7]. Some of the blood vessels showed calcific thrombi (phleboliths). Lymph nodes excised showed reactive changes. A confirmatory diagnosis of cavernous haemangioma with phleboliths was given.

Discussion

Haemangiomas are benign neoplasms arising due to abnormal endothelial proliferation. They are congenital neoplasms that do not present themselves until sudden growth causes pain or

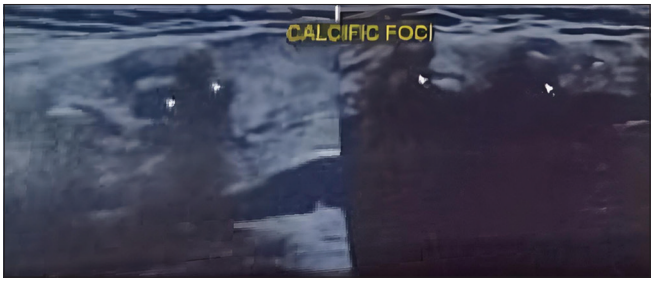

Figure 2: Ultrasonography image showing calcifications in the lesion

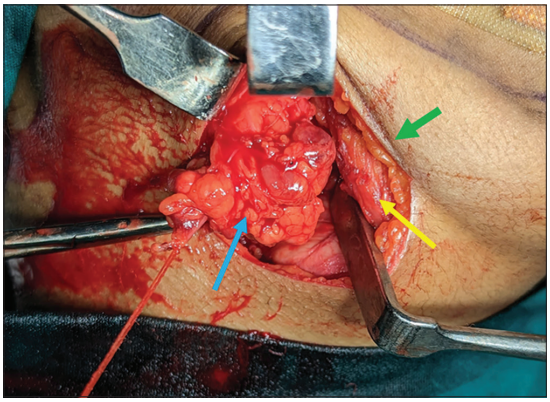

Figure 4: Excision of the lesion through right submandibular incision. Solid green arrow shows approach through submandibular incision, solid blue arrow depicts the lesion, solid yellow arrow shows submandibular gland adjacent to the lesion

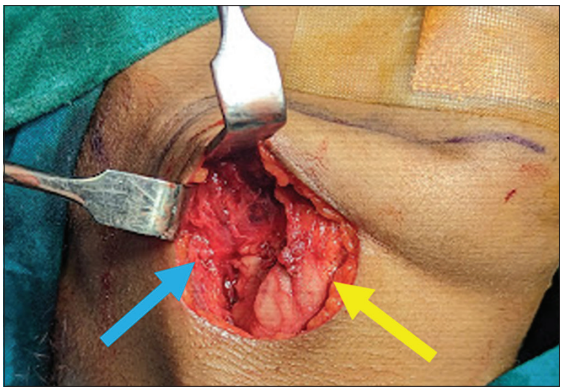

Figure 5: Preservation of submandibular gland - Blue arrow showing site of excised lesion lateral to the submandibular gland. Yellow arrow showing preserved submandibular gland

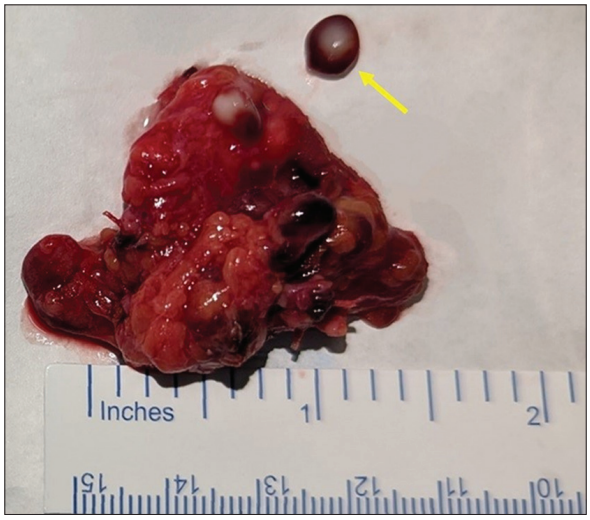

Figure 6: Excised specimen-Gross appearance of excised neoplasm with solid yellow arrow showing phlebolith

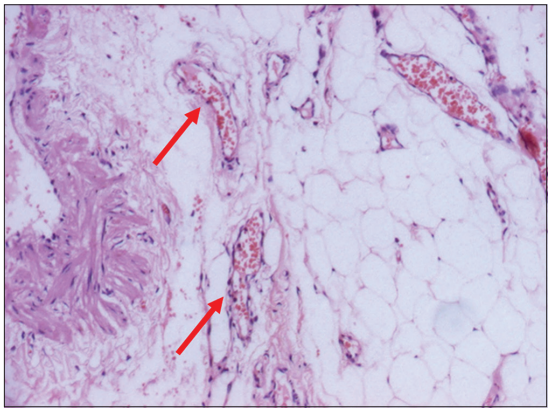

Figure 7: Microscopic examination under 10x magnification using hematoxylin and eosin staining showing enlarged and engorged blood vessels marked with red arrows

deformity.[1] Haemangiomas can involve the skin, muscle[2] and gland parenchyma directly or by secondary invasion from subcutaneous vessels.[4] Cavernous variants are usually seen in adults, whereas infantile haemangiomas are capillary.[6] A cavernous variant is usually less circumscribed. Phleboliths are idiopathic calcium deposits seen in haemangiomas, even in the absence of calcium-phosphate derangements.[7] Although diagnosis is based on history, examination and imaging, some lesions require excision due to aesthetic compromise, airway obstruction or excessive bleeding tendency.[8]

Haemangiomas of the head & neck region are relatively common; however, they are a rare entity in the submandibular region.[3] A lesion in the submandibular area with radio-opacities primarily suggests sialolithiasis, therefore justifying further confirmatory investigations.[3] A literature review conducted in 2019, showed only 18 documented cases of submandibular haemangioma, of which 17 were cavernous and only one was found to be of venous type.[9] A literature search done in 2022 reported 18 cases of submandibular haemangioma, where the treatment involved resection of tumour along with the submandibular gland.[10] To the best of our knowledge, all documented cases of submandibular haemangioma included the submandibular gland, which makes this case an exception as the lesion did not invade the gland parenchyma.

In conclusion, there are three noteworthy points in this case. First, cavernous haemangiomas rarely occur in the submandibular triangle. Second, the clinical and radiographic findings are suggestive of sialolithiasis, thus misguiding the physician and warranting further investigations. Third, haemangiomas in the submandibular triangle usually involve the gland; however, since this lesion was lateral to the gland, we were able to preserve the gland itself, thus preserving the function.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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