



Spindle Cell Haemangioma of Cheek

Amay Vikrama Rathore*, Deepika Garg, Prakash Nagpure, Sanjeev Kumar V S, Swini Kaushal and Rohit Dabas

Mahatma Gandhi Institute of Medical Sciences, Sewagram, Wardha, Maharashtra, India

***Corresponding Author:** Amay Vikrama Rathore, Mahatma Gandhi Institute of Medical Sciences, Sewagram, Wardha, Maharashtra, India.

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Abstract

Spindle cell hemangioma (SCH) is a distinct benign vascular lesion that typically occurs in the dermis or subcutaneous tissue of distal extremities. Although SCH can occur in a variety of anatomic sites, very few cases have been reported in the head and neck region [1]. Spindle cell haemangioma presents as a solitary tumour or as multiple nodules clustered within the same region. In this article we present a rare case report of spindle cell hemangioma of left side of cheek in a 45 years old adult with a long-standing swelling present over face for 8 years. We excised the tumor and sent for histopathological examination which was reported as spindle cell hemangioma. Microscopic finding revealed cavernous vascular space with intervening thick septations consisting of spindle cell.

Keywords: Myxoid Degeneration; Cavernous Vascular Space; Phleboliths; Maffucci's Syndrome; Klippel-Trenaunay Syndrome; Congenital Lymphedema

Introduction

Spindle cell hemangioma is a benign tumour that is comparatively rare. It affects almost exclusively the dermis and subcutaneous tissue of the distal extremities and rarely the proximal extremities, axilla, trunk, vulva, penis, spleen, pancreas, bones, and head and neck. Lesions range in size from a few millimetres to a few centimetres, with the majority being less than 2.0 cm. The colour of the skin or mucosa on top is normal or bluish, and its substance is firm. As a benign lesion, spindle cell hemangioma is currently best treated with conservative excision without adjuvant chemotherapy or radiation [1]. Following surgical excision, local

recurrence rate of up to 58% has been reported. Recurrences occur more commonly in patients with multiple lesions at presentation, occurring near surgical sites rather than within them [1]. Only 15 SCH instances have been documented in the head and neck area as of 2020 [2]. SCH is equally prevalent in both sexes and spans a large age range. Although isolated lesions are more common in men and multifocal lesions in women, there is no discernible gender bias. Due to the tumour's asymptomatic nature, pain is only seldom reported it typically exists for years before being diagnosed [3]. Clinically, the majority of cases are identified as intraoral vascular neoplasms, which include fibroma, pyogenic granuloma, peripheral

giant cell granuloma, peripheral ossifying fibroma, necrotizing ulcerative gingivitis, and inflammatory fibrous hyperplasia [4]. First described in 1986 by Weiss and Enzinger, the spindle cell hemangioma (SCH) is a unique vascular lesion characterized by cavernous blood vessels separated by spindled cells reminiscent [5]. Spindle cell haemangiomas are composed of two parts: cavernous vascular spaces with bland, flattened endothelial cells lining them, and more substantial spindle cell portions. There may be organized, hyalinised thrombi called phleboliths in the cavernous vessels. The solid spindled areas are composed of bland spindled cells with tapered ends that frequently contain slit like vascular spaces. In contrast to the septa of cavernous haemangiomas, the cavernous vascular septa also exhibit spindled cells, giving them a more cellular appearance. Additionally, the spindle cell component includes blister cells, which are rounded epithelioid endothelial cells with cytoplasmic vacuoles [5]. SCH lacks the ability to metastasize, and there is only single instance of malignant transformation with metastasis documented in the literature [5].

Case Report

A 45 years old male patient having complaint of a swelling over left side of cheek since past 8 years. He noticed the swelling over cheek have episodes of growth and remission in the last 12 months with no aggravating and relieving factor, with no other symptoms of pain, numbness, visual complaints and nasal complaints.

On inspection: 0.8 x 0.5 cm size sub-epithelial swelling present over left side of cheek, with normal skin no sinus, no discharge.

On palpation: inspection finding confirmed, solitary soft circumscribed sub-epithelial swelling measuring 0.5x 0.5 x 0.5 cm present over left side of cheek, mobile, non-tender with intact skin. No sinus, no discharge, not attached to skin and underlying muscle or bone, non-pulsatile and no local rise of temperature. This swelling was too small and non-pulsatile and not attached to the underlying bone and skin it appears to be like a benign swelling like lipoma so we planned excision for this swelling directly without fine needle aspiration not adequate material extracted from such swelling dur-

ing aspiration. Excision of swelling take place after a linear incision of 2 cm over left side of cheek after palpation of the swelling. We excised this sub-epithelial swelling and specimen was sent for histopathology. Wound closed in primary after excision (Figure 3).

Gross specimen of excised swelling (Figure 1) showing single grey brown strawberry like soft swelling of size 0.8x0.5x0.3 cm removed from left side of cheek.

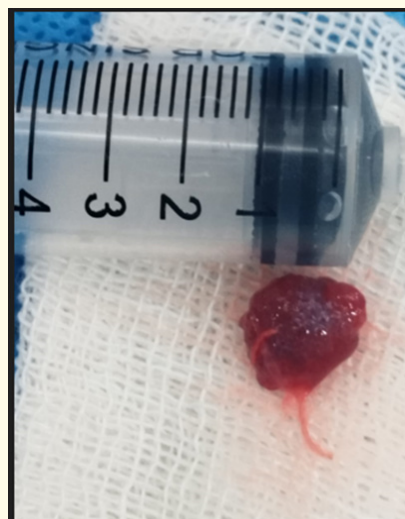


Figure 1: Showing gross picture.

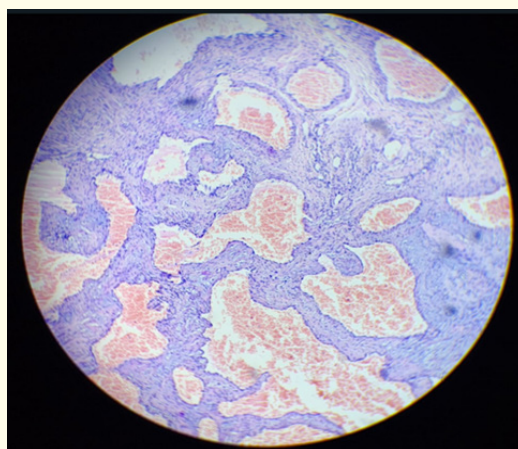


Figure 2: Microscopic picture.



Figure 3: Defect after removal of specimen and after 7 days.

Microscopic finding shows poorly circumscribed lesion which is showing thin wall cavernous vascular spaces with intervening solid areas mainly composed of spindle cells with few areas showing myxoid degeneration. These features are suggestive of a spindle cell hemangioma. CD34 immunohistochemistry showed positive staining in endothelial cells (Figure 2).

Data availability statement

The data used to support the findings of this study are included within the case report.

Discussion

In order to prevent misdiagnosis, spindle cell haemangioma (SCH), an uncommon vascular tumour of the head and neck, must be identified [3]. It is often present for several years prior to diagnosis due to its slow clinical progression. These instances range in age from one and a half to seventy years [2]. These cells have immunoreactivity to endothelial markers, including factor VIII-related antigen, vimentin, CD34, and CD31 [1]. The various vascular tumours that are included in the differential diagnosis of SCH are pyogenic granuloma, Kaposi sarcoma, cavernous haemangioma, epithelioid haemangioma, intravascular papillary endothelial hy-

perplasia, and Kaposi form haemangioendothelioma [7]. This tumour resembles Kaposi sarcoma due to the spindle cells and sporadic numerous lesions. In addition to having a higher mitotic rate and nuclear atypia, spindle cell hemangioma does not have the infiltrative growth pattern seen in Kaposi sarcoma [7]. SCH does not display the distinctive glomeruli-like nests that are seen in Kaposi form haemangioendothelioma [7]. Features of Kaposi's sarcoma, like male preponderance and sporadic multifocal development, are similar to those of SCHs. Although Kaposi's sarcoma lacks epithelioid cells and hardly ever has cavernous arteries with thrombi and phleboliths, its spindle cells respond to the endothelial marker CD34. However, SCH does not express human herpes virus 8 latent nuclear antigen-1 or exhibit the hyaline globules seen with Kaposi's sarcoma [4]. The polypoid, limited, exophytic lobular proliferation of capillaries in a fibro-myxoid stroma with surface ulceration and cavernous sections lacking spindle cell development is what sets apart pyrogenic granuloma from SCH. The latter is more acellular, lacks spindle and epithelioid endothelial cells, and has many papillae, whereas SCH may show areas that resemble intravascular papillary endothelial hyperplasia [7]. IDH1 (isocitrate dehydrogenase) and IDH2 somatic mosaic mutations have recently been identified in SCH, but not in other vascular diseases. According to one study, 71% of SCH patients had an IDH1/IDH2 mutation. According to a different study, IDH1/IDH2 mutations were present in 16/17 SCH cases, whereas these mutations were absent from other vascular abnormalities involving lymphatic malformation. Thus, it was determined that this mutation can be utilized to diagnose SCH because it is quite specific for it [8]. In our situation, however, IDH1 IHC was not performed. Multifocal tumours that start in the same anatomical area account for the majority of SCH recurrences, according to certain views. A vascular abnormality may be the cause of these tumours, or they may spread intravascularly [5]. Multiple SCH lesions may be linked to varicose veins, congenital lymphedema, Klippel-Trenaunay syndrome, and Maffucci's syndrome [8]. But in our case, there is no findings of these syndromes.

Conclusion

Spindle cell haemangioma is a rare, benign vascular tumour that can occasionally present in unusual sites such as the cheek. Due to its rarity in the head and neck region and its histopathological similarity to other vascular neoplasms, accurate diagnosis is crucial to avoid unnecessary aggressive management. Complete local excision remains the treatment of choice, with a generally excellent prognosis and minimal risk of recurrence in solitary lesions. Awareness of this entity among clinicians and pathologists is important for early recognition and appropriate management.

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