Case Report

Hemangiolymphangioma of the cervical region: A rare case report

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ABSTRACT

Hemangiolymphangioma is a rare developmental anomaly. Vascular anomalies are classified under two categories, including vascular tumors and vascular malformations, based on cellular turnover, histology, and clinical features. Hemangioma is an example of a vascular tumor, and lymphangioma is an example of a vascular anomaly. Malformations can be seen in mixed combinations of vascular elements, such as blood and lymph. Histologically, vessels filled with both blood and lymph are named as lymphangiohemangioma or hemangiolymphangioma, depending on the dominant tissue present. This paper reports a case of hemangiolymphangioma in a 20-year-old male patient.

Key words: Cystic hygroma, Hamartomatous, Hemangioma, Lymphangioma

Vascular anomalies fall into two main categories: vascular tumors and vascular malformations. They differ in cellular turnover, histology, and clinical presentation [1,2]. Endothelial cell growth is a key feature of vascular tumors, like hemangiomas. Meanwhile, vascular malformations arise from issues in the development of blood vessels during embryonic and fetal stages; these include capillary, venous, and lymphatic types [3]. Hemangiomas are frequently found in infants and young children, often appearing within the first 8 weeks of life and affecting females more than males. Clinically, hemangiomas usually show up as pale spots with thin, thread-like features and are mainly classified as cavernous or capillary subtypes. They can occur in various locations, including the skin, mucous membranes, muscles, or bones [4-6].

Lymphangiomas are benign growths of the lymphatic system, marked by an abnormal development of lymphatic vessels. These lesions come from isolated lymphatic tissue that does not connect properly with the rest of the lymphatic network. They usually appear at birth or within the first 2 years of life, primarily in areas like the head and neck, armpits, chest, mediastinum, retroperitoneum, buttocks, and perineum [3]. Superficial lymphangiomas often show as raised bumps with a pink or yellowish color or as clusters of clear fluid-filled sacs. Deeper lesions appear as soft, diffuse masses underneath normal skin. Lymphangiomas are classified into three main types: superficial (lymphangioma circumscriptum), deep (lymphangioma cavernosum), and cystic (cystic hygroma) [7,8]. Mixed vascular malformations, such as lymphangiohemangioma or hemangiolymphangiomas, contain a mix of vascular elements, such as lymphatic and venous tissue, which makes them tough to categorize [6]. A histological check of these mixed malformations typically shows vessels filled with both blood and lymphatic fluid.

This report describes a case of a hemangiolymphangioma in the neck, highlighting the clinical and histological challenges of mixed vascular anomalies.

CASE PRESENTATION

A 20-year-old man came to the Surgery Department at Guru Gobind Singh Medical College and Hospital in Faridkot, Punjab, India, with a swelling in his neck that had been there since birth. He noted that the swelling had gradually increased in size and he had mild, dull aching pain over the past 2 months without radiation or aggravating factors.

Physical examination findings were unremarkable for an average-built male with stable vital signs. The pulse rate was recorded at 82 beats/min, while blood pressure was 118/76 mm Hg. Respiratory rate stood at 16 breaths/min, while body temperature was normal. No pallor, icterus, cyanosis, or lymphadenopathy was present. On local examination, the swelling had the following characteristics: It was soft in texture, non-pulsatile, compressible, painless, and without any

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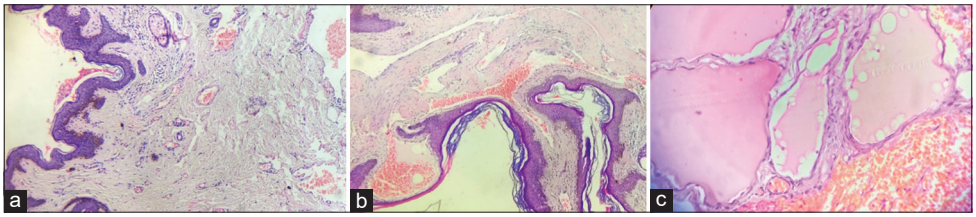

Figure 1: (a and b) Photomicrograph (H&E, stain ×100) shows stratified squamous epithelium showing many vessels spaces which are filled with either blood cells or lymphatic fluids; (c) Photomicrograph (H&E, stain ×400) showing dilated vessels filled with lymph

changes in the coloration of the skin or ulcers. There were no symptoms such as bleeding, discharge, trauma, fever, or difficulty breathing.

An ultrasound showed an uneven area measuring 1.1 × 0.5 cm in the superficial layer, with low blood flow, indicating a vascular malformation.

With aseptic precautions in place and proper anesthesia provided, surgical excision of the lesion was carried out successfully. The intra-operative course showed the presence of the lesion in the subcutaneous plane, which was subsequently removed. All hemostatic measures were taken, and the specimen obtained was sent for histopathological evaluation in the Pathology Department.

The gross inspection of the specimen revealed it was partially covered by the skin, had a soft texture, and measured about 1.8 × 1.5 × 0.5 cm. Microscopic assessment of Hematoxylin and Eosin (H&E) stained sections showed an epidermal lining, with the dermis and subcutaneous tissue containing large, dilated, interconnected blood vessels. These vessels had flattened endothelial cells and contained both blood and lymphatic fluid. The histopathological findings confirmed the diagnosis of hemangiolymphangioma (Fig. 1).

Follow-up was done for 6 months after surgery, and the patient demonstrated adequate healing of the wound without any signs of recurrence or functional impairment.

DISCUSSION

Vascular anomalies are classified into vascular neoplasms and vascular malformations [1,2]. Vascular malformations occur due to issues in vasculogenesis, while vascular tumors involve the abnormal growth of endothelial cells. The main difference between these two types lies in their histological features and rates of endothelial cell turnover [4]. Vascular neoplasms show increased endothelial cell turnover, while vascular malformations exhibit normal or reduced turnover [9].

Hemangiomas are the most common soft-tissue tumors in infants, often found in the head and neck areas. These lesions typically do not appear at birth but emerge within the first few weeks of life. They go through a rapid growth phase followed by natural regression. Histologically, hemangiomas are characterized by a high number of endothelial cells [10].

Vascular malformations are congenital lesions that last a lifetime, even if they are not obvious at birth. Histologically, these malformations consist of vessels with normal endothelium and can include veins, capillaries, arteries, lymphatic vessels, or a mix of these. In this case, histopathological analysis identified a combination of lymphatic and capillary vessels with red blood cells, confirming the diagnosis of hemangiolymphangioma [10].

The differentials in the case of cervical hemangiolymphangioma include cystic hygroma, cavernous hemangioma, branchial cleft cyst, venous malformation, lymphangioma circumscriptum, arteriovenous malformation, lipoma, and enlarged cervical lymph nodes. Radiographic imaging may be used to diagnose the condition, but biopsy is the most reliable method due to their similar appearances [8].

A few case studies on hemangiolymphangiomas of the mouth, buccal mucosa, tongue, and cervical area have been seen in the literature. Yarmand *et al*. discussed a unique case of lymphangiohemangioma of the buccal mucosa that was observed to be associated with painless swelling, showing histologically mixed lymphatics and vascular spaces [2]. In a similar study, Sobhana *et al*. demonstrated a hemangiolymphangioma of the buccal mucosa in a patient, and its complete surgical removal was recommended in order to avoid its recurrence [6]. In a case study done by Bezzola *et al*., a cervical cystic lymphangioma with vascular elements in an adult patient was discussed, which is indeed very rare [11]. These reports, combined with our case report, indicate that there can be variations in the clinical presentation of mixed vascular malformations and require histopathological confirmation for accurate diagnosis.

CONCLUSION

Hemangiolymphangioma is a rare condition, with few cases reported in medical literature. Accurate diagnosis of suspected cases is vital and requires thorough evaluation. Early identification and diagnosis are crucial to initiate effective treatment and reduce the risk of complications. Surgical removal is generally the preferred treatment for these lesions. In addition, regular follow-up is important to monitor for any signs of recurrence.

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