Case Report

Recurrent angioleiomyoma in the neck: A rare case report

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ABSTRACT

Angioleiomyomas are infrequent benign soft tissue tumors. They arise from smooth muscle cells of the vessel wall. They are common in lower extremities. Neck is an uncommon site. Recurrence and malignant transformation are extremely rare. We report a case of 55-year-old male with recurrent angioleiomyoma in neck. Rapidly growing tumors can mimic malignant clinically. Careful histological examination and immunohistochemistry are indispensible for definitive diagnosis. Treatment is surgical resection and extended follow-up.

Key words: Angioleiomyoma, benign, neck, recurrent

INTRODUCTION

Angioleiomyomas are subtype of leiomyoma. They are infrequent benign soft tissue tumors arising from smooth muscle cells of the vessel wall. They are preferentially located on the lower extremities. Neck is an uncommon site. Recurrence and malignant transformation are extremely rare.[1,2] We are tempted to report one such case of angioleiomyoma in the neck which recurred after surgical excision.

CASE REPORT

This is a case report of 55-year-old male patient, who presented with a firm subcutaneous swelling on the left side of the neck below the angle of the mandible for 1 year. He gave history of pain and rapid increase in size for 15 days. He also had history of removal of tumor from the same site 4 years back at another hospital. Previous available reports with him suggested it was leiomyoma. No other significant medical history or illness was present. On local examination, scar measuring 1 cm in length was present at the site of the swelling. Firm subcutaneous freely movable swelling measuring 4 cm × 3.8 cm was palpable on the left side of neck below the angle of mandible. There was no cervical lymphadenopathy or any other significant findings. A provisional clinical diagnosis of lymphadenopathy was made. Repeated fine needle aspiration yielded frank blood. No lymphoid cells or epithelial cells were seen. A Vascular lesion was suspected.

Excision biopsy was done for definitive diagnosis. Surgical specimen received for histopathological examination was well-delineated, homogenous yellow white tumor mass measuring 3 cm × 3 cm × 2.8 cm. Cut section showed few areas of hemorrhage [Figure 1a]. Histopathological examination showed well-encapsulated tumor, which was composed of numerous blood vessels of different caliber, interspersed with bundles of mature smooth muscles. Smooth muscle cells lacked nuclear pleomorphism and mitotic activity. Blood vessels were mixture of small slit like, dilated and thick wall [Figure 1b and c Arrows]. Foci of mature adipose tissue and lymphocytic inflammatory cells were seen [Figure 1d]. Angiomyolipoma and angioleiomyoma were suspected. Thick vessel walls lacked internal and external elastic lamina on Verhoff's elastic stain [Figure 1e]. Smooth muscle cells showed immunopositivity with smooth muscle actin and desmin but lacked immunopositivity with HMB-45 [Figure 1f]. Final histological diagnosis of angioleiomyoma was made.

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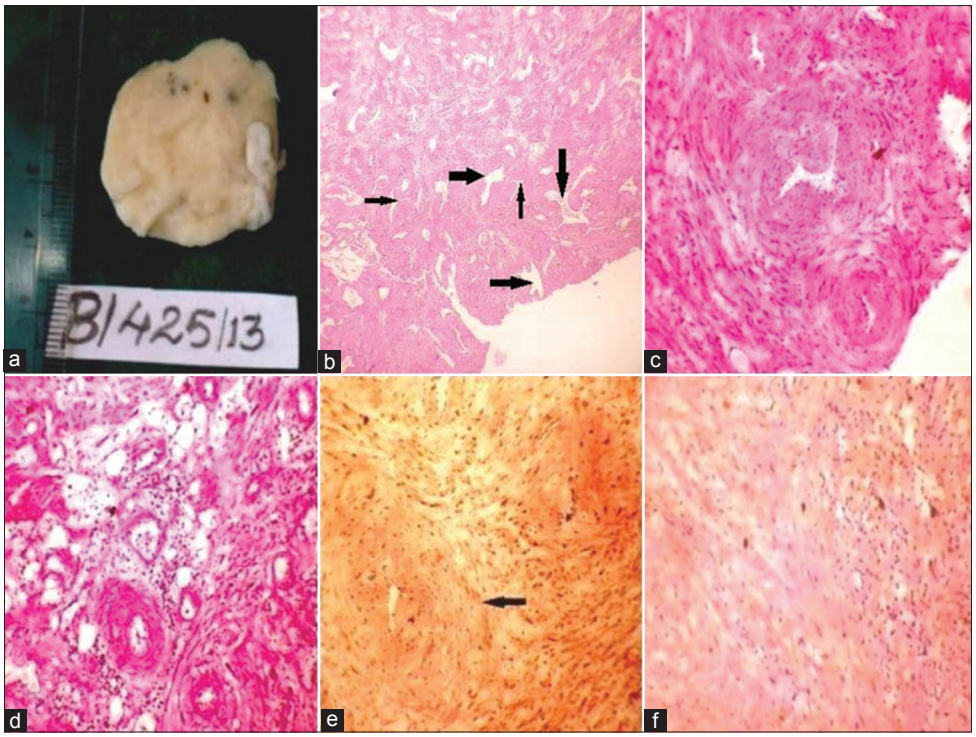

Figure 1: (a) Yellow white well delineated tumor. (b) Small slit like (thin arrows) and dilated (thick arrows) blood vessels and smooth muscle cells (H and E, ×100). (c) Thick wall blood vessels. (d) Foci of mature adipose tissue and lymphocytes (H and E, ×400). (e) Thick vessel walls lacked elastic lamina (Verhoff's elastic stain, ×400). (f) Smooth muscle cells lacked immunoreactivity with HMB-45 (Immunohistochemistry, ×100)

DISCUSSION

Earlier little attempt was made to differentiate angioleiomyoma from leiomyoma. In 1937, Stout AP first designated them as vascular leiomyoma to contrast them with leiomyoma.[3] Angioleiomyomas usually occur as a slowly enlarging mass of long duration. Most cases are usually less than 2 cm. Peak incidence is in 4th to 6th decade. Females are commonly involved than males. Half of the cases have pain. In this case size of the lesion was more than 2 cm. Patient had history of pain and rapid increase in size of tumor mass of shorter duration.

Though angioleiomyomas can occur at any site, they are preferentially located in the soft-tissues of lower extremity in females. Tumors in males are common in the upper extremity, head and neck.[4] In the lower extremities they are often below the knee. Common sites in the head are ear, lips nose, nasal cavity and face.[1] Rarely they have been reported in intracranium.[5,6] Usually they are solitary, rarely they can be multicentric.[6]

Three histological types are described. (1) Solid: In which smooth muscles cell bundles are interspersed with small, slit like vascular channels. (2) Cavernous: In which there are dilated vascular channels and vessel wall could be barely distinguished from intravascular smooth muscles bundle. (3) Venous: In which there are thick wall blood vessels with less compact smooth muscle bundles. Histological findings in our case were a mixture of all the three types.

A differential diagnosis of angiomyolipoma was considered for the reason that they are known to occur at extra renal sites and presence of mature adipose tissue. However, smooth muscle cells lacked immunoreactivity with HMB-45 which ruled out angiomyolipoma. Foci of lymphocytic inflammatory cells and mature adipose tissue can be seen in angioleiomyoma. Recurrence after surgical excision is extremely rare and reported by very few authors.[1,7] Recurrence in the neck is an uncommon site. Rarely malignant changes have been reported in recurrent tumors.[2] We conclude that angioleiomyomas are benign soft-tissue neoplasms arising from smooth muscle cells of the vessel wall. Neck is an uncommon site. Recurrence in neck is extremely rare. Rapidly growing tumors can mimic malignant lesion clinically. Careful histological examination and immunohistochemistry is indispensible for definitive diagnosis. Treatment is surgical resection and extended follow-up.

REFERENCES

1. Hachisuga T, Hashimoto H, Enjoji M. Angioleiomyoma. A clinicopathologic reappraisal of 562 cases. Cancer 1984;54:126-30.
2. Herren DB, Zimmermann A, Büchler U. Vascular leiomyoma in an index finger undergoing malignant transformation. J Hand Surg Br 1995;20:484-7.
3. Weiss SW, Goldblum JR. Benign tumors of smooth muscle.

In: Enzinger and Weiss's Soft Tissue Tumor. 5th ed. USA: Mosby Elsevier; 2008. p. 520.
4. Wang CP, Chang YL, Sheen TS. Vascular leiomyoma of the head and neck. Laryngoscope 2004;114:661-5.
5. Anderson TD, Weinstein GS. Recurrent angiomyoma (vascular leiomyoma) of the larynx after laser excision. Otolaryngol Head Neck Surg 2000;123:646-7.
6. Shinde SV, Shah AB, Baviskar RB, Deshpande JR. Primary intracranial multicentric angioleiomyomas. Neurol India 2012;60:115-7.
7. Mahima VG, Patil K, Srikanth HS. Recurrent oral angioleiomyoma. Contemp Clin Dent 2011;2:102-5.

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