



Primary Cervical Leiomyoma Presenting as a Lateral Neck Swelling in a Middle-Aged Female – A Rare Case Report

Ankita Sehgal¹, Anand Bhandary Panambur^{2*}, Thoppil Reba Philipose³

¹Resident, Department of General Surgery, Mangalore, India

²Assistant Professor, Department of General Surgery, Mangalore, India

³Professor, Department of Pathology, A.J. Institute of Medical Sciences & Research Center, Mangalore, India

Received: July 01, 2024

Published: July 23, 2024

© All rights are reserved by

Anand Bhandary Panambur, *et al.*

***Corresponding Author:** Anand Bhandary Panambur, Assistant Professor, Department of General Surgery, Mangalore, India.

DOI: 10.31080/ASCR.2024.05.0568

Abstract

Leiomyomas are benign tumors originating from smooth muscle, typically observed in middle-aged women's lower extremities, gastrointestinal system, skin, and uterine myometrium. Because there isn't much smooth muscle in the head and neck area, less than 1% of all leiomyomas arise there. Although the exact origin of leiomyomas is still unknown, the most widely accepted theories include multipotent mesenchymal cells and vascular smooth muscle, which make up the majority of the walls of small blood arteries. After surgery, histopathological and immunohistochemical tests are required for a conclusive leiomyoma diagnosis. We are describing a case of leiomyoma in a middle-aged female patient who initially presented with a lateral neck swelling.

Keywords: Leiomyoma; Lateral Neck Swelling; Primary Cervical Leiomyoma; Excision Biopsy

Introduction

Leiomyoma is a benign soft-tissue neoplasm arising from smooth muscle. Virchow initially described it in 1854, and Blanc first reported it in 1884 [1]. Leiomyoma is rare in the head and neck region due to the lack of smooth muscle. Leiomyomas are most commonly found in the uterine myometrium (95%), followed by the skin (3%), the gastrointestinal system (1.5%), and the head and neck region (less than 1%) [2,3]. The tumor typically arises in the fourth and fifth decades, with a little female predisposition, and manifests as a slow-growing, asymptomatic lesion. The histogenesis of leiomyomas is still debated, although the most common hypotheses are vascular smooth muscle, which is the primary component of the wall of tiny blood arteries, and multipotent mesenchymal cells [4,5]. Histopathological and immuno histochemical analyses after surgery are necessary for the definitive diagnosis of

leiomyoma. We are reporting a case of leiomyoma presenting as a lateral neck swelling in a middle aged female.

Case Report

A 46 year old female presented to our hospital with a left posterolateral neck swelling, which she reported had been present since last 2 years. Physical examination demonstrated a firm nontender swelling measuring 5 cm x 3 cm deep to sternocleidomastoid muscle not fixed to surrounding structures in the left posterolateral aspect of the neck. FNAC was performed which showed neurofibroma. CECT neck showed heterogeneously enhancing oblong lesion in the posterior cervical space on the left side deep to the left sternocleidomastoid muscle (Figure 1a). It is seen abutting the carotid vessels and indenting the internal jugular vein (Figure 1b). Patient underwent excision biopsy and specimen (Figure 2)

sent for histopathological examination. Histopathology revealed leiomyoma of the left cervical swelling (Figure 3a). Immunohistochemistry confirmed the diagnosis with Desmin +ve and S-100 -ve (Figure 3b). Patient is on follow-up.

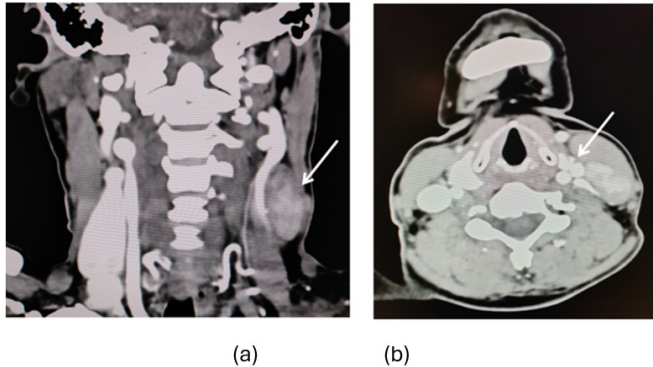


Figure 1: CECT neck showed heterogeneously enhancing oblong lesion in the posterior cervical space on the left side deep to the left sternocleidomastoid muscle (a). It is seen abutting the carotid vessels and indenting the internal jugular vein (b).

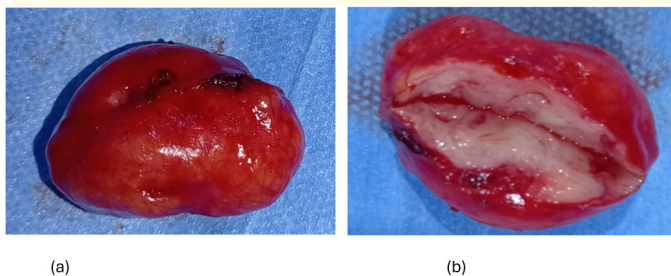


Figure 2: Excision biopsy gross specimen (a) shows well encapsulated, well circumscribed lesion. The cut surface of the excised mass (b).

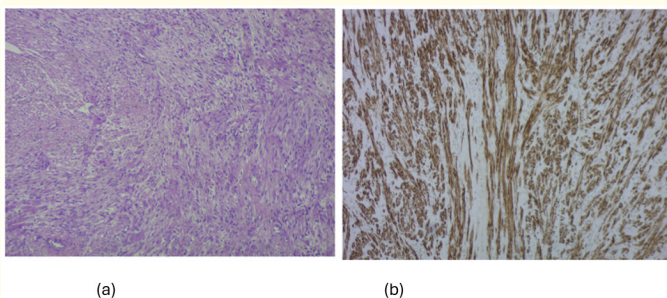


Figure 3: (a) Well-circumscribed lesion having spindle-shaped cells arranged in whorls and fascicles with eosinophilic cytoplasm. Tumor has rich vascular channels with thick muscular vessel walls and intervascular smooth muscle bundles (H&E stain). (b) High-power view showing immunohistochemical staining with smooth muscle desmin highlights the proliferated smooth muscle cells.

Discussion

Leiomyoma is a non-epithelial benign tumour that most usually affects the uterus, esophagus, and skin. Less than 1% of all leiomyomas occur in the head and neck, as this region contains relatively little smooth muscle. They are usually solitary, rounded, and well-demarcated masses. Congenital origin, blood flow disturbance, infection, and estrogen involvement [6] are all postulated mechanisms for the formation of leiomyoma, but no consensus has yet been achieved. Progesterone receptors are expressed in the nucleus of tumour cells, and progesterone is implicated in tumour genesis and proliferation [7]. This may be related to the higher incidence in females (ratio is 1: 3.75) [8].

Smooth muscle neoplasms are further divided into three categories by the WHO: leiomyoma (solid leiomyoma), angiomyoma (vascular leiomyoma), and epithelioid leiomyoma (leiomyoblastoma). The most prevalent is leiomyoma, which differs from angiomyoma in terms of the degree of angiogenesis in the tissue [9]. There is evidence to support the theory that leiomyoma in the head and neck is caused by the vascular smooth muscle, which is the major component of the small blood vessel wall [2,3]. Another theory is that multipotent mesenchymal cells are also responsible, however the histogenesis [10] is still debated.

The clinical differential diagnosis relevant to extra-oral leiomyoma includes fibroma, lipoma, neurofibroma, dermatofibroma, lymphangioma, hemangioma, and soft-tissue cysts such as dermoid cyst [12]. Proliferating spindle cells in a tangled arrangement are a characteristic histopathological feature of leiomyoma. In addition to histopathology, immunohistochemistry using alpha-SMA and h-caldesmon is also precise and reliable for definitive diagnosis [8]. Alpha-SMA is most commonly used as a myogenic marker, and h-caldesmon is expressed exclusively in smooth muscle and is a highly specific marker for it [11]. In our case, we have done Desmin and S-100 to differentiate leiomyoma from neurofibroma. Local resection with an adequate safety margin of normal-appearing tissue is the treatment of choice with recurrence being extremely rare. Despite the fact that these tumors have a vascular origin, significant bleeding following excision is rare.

Conclusion

Leiomyoma is a rare benign tumor in the head and neck with a favorable prognosis. The diagnosis is primarily based on histological findings. Surgical excision is the only means to diagnose and provides good results, with rarely seen recurrences. In our patient, it manifested as a painless swelling in her neck. Preoperative imaging and cytology are not particularly helpful for establishing a

preoperative diagnosis. Clinicians should be aware of the presentation and behavior of this tumor and consider it as a differential diagnosis for painless neck swelling.

Bibliography

1. Kloefer HW, *et al.* "Hereditary multiple leiomyoma of the skin". *American Journal of Human Genetics* 10 (1958): 48-52.
2. Gianluca S., *et al.* "Leiomyoma of oral cavity: Case report and literature review". *Annals of Stomatology (Roma)* 2 (2011): 9-12.
3. Baden E., *et al.* "Leiomyoma of the oral cavity: A light microscopic and immunohistochemical study with review of the literature from 1884 to 1992". *European Journal of Cancer Part B: Oral Oncology* 30B (1994): 1-7.
4. S Erkilic., *et al.* "Primary leiomyoma of the thyroid gland". *Journal of Laryngology and Otology* 117.10 (2003) 832-834.
5. G Vincenzi., *et al.* "Atypical (bizarre) leiomyoma of the nasal cavity with prominent myxoid change". *Journal of Clinical Pathology* 55.11 (2002) 872-875.
6. T Okada., *et al.* "Leiomyoma of the parotid gland; a case report". *Practica Oto-Rhino-Laryngologica* 96.8 (2003): 711-715.
7. G Marioni., *et al.* "Progesterone receptor expression in angioleiomyoma of the nasal cavity". *Acta Oto-Laryngologica* 122.4 (2002): 408-412.
8. R Meher and S Varshney. "Leiomyoma of the nose". *Singapore Medical Journal* 48.10 (2007): e275-e276.
9. E Baden., *et al.* "Leiomyoma of the oral cavity: a light microscopic and immunohistochemical study with review of the literature from 1884 to 1992". *European Journal of Cancer Part B* 30.1(1994): 1-7.
10. A Vincenzi., *et al.* "Atypical (bizarre) leiomyoma of the nasal cavity with prominent myxoid change". *Journal of Clinical Pathology* 55.11 (2002): 872-875.
11. MM Miettinen., *et al.* "Calponin and h-caldesmon in soft tissue tumors: consistent h-caldesmon immunoreactivity in gastrointestinal stromal tumors indicates traits of smooth muscle differentiation". *Modern Pathology* 12.8(1999): 756-762.
12. Brooks JK., *et al.* "Clinicopathologic characterization of oral angioleiomyomas". *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology* 94(2002): 221-227.