CASE REPORT

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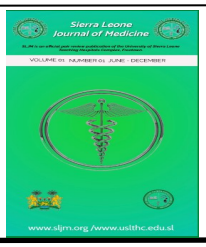

Cutaneous Leiomyoma of the Leg: A Case Report and Literature Review

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ABSTRACT

Cutaneous leiomyoma is a rare benign smooth muscle tumor that can be mistaken for other nodular cutaneous lesions. We present the case of a 27-year-old female graduate who developed a swelling on her left leg over a period of 2 years. Initially painless, the lesion became painful a year later, with pain exacerbated by heat. The lump measured 1 x 1 x 1 cm, was tender, and not warm. Initially diagnosed clinically as an epidermal inclusion cyst, she underwent excision biopsy revealing leiomyoma upon histological examination. Subsequent to excision, all symptoms resolved completely, with no recurrence noted. Hence, it is crucial to consider cutaneous leiomyoma as a potential differential diagnosis for such cutaneous lesions.

Key words: Cutaneous, Leiomyoma, Leg, Africa, Nigeria

1. INTRODUCTION

Leiomyoma is a benign tumour of smooth muscle origin. Published report indicates that it accounts for 4.4% of all benign soft tissue tumours.[1,2] Cutaneous leiomyoma (CL) either hereditary or sporadic, originates from the smooth muscles in the dermis and subcutaneous tissue; it is rare and represents 3.5- 4% of all leiomyomas[3]. CL usually present as a painful and tender cutaneous or subcutaneous lesion.[3,4,5,6] CL was first described by Rudolf Virchow in 1854 but it was not well known until after Besnier published a comprehensive report on it in 1880[6]; from then many case reports and series [6,7,8,4,5,9,10] as well as an epidemiological study[3] have emerged in literature. The numerous case reports and series from different parts of the globe indicate that CL is well-known and rare tumour with varying pattern of clinical presentation that can be confused with other cutaneous /subcutaneous lesions[6]. Besides, the rate of 76-94.6% missed preliminary clinical diagnosis of CL in published reports [4,5] also shows that it is an uncommon tumour prone to delayed or missed diagnosis. The diagnostic dilemma associated with the diagnosis of CL, especially in Africa where it is a very rare lesion with scanty published reports, necessitated this case report presenting a young female patient who had a painful solitary CL of the leg in a Nigerian setting

2. CASE PRESENTATION

A 27-year-old female graduate presented to the Family Medicine Clinic on the account of swelling in the left leg of 2 years duration. The lump, an incidental finding while she was washing her legs in the bathroom, was felt but initially not visible. It gradually increased in size and became visible and painful a year later. The pain was localised and sharp, aggravated by tactile stimuli and a long distance walk. Exposure to hot temperature on a sunny day or from burning firewood in the kitchen triggered and aggravated the pain. The pain was not related to her menstrual cycle. There was no associated low back pain and relieving factor. Prior to presentation, she was treated with antibiotics and analgesics when the lump transited to a painful form but there was no relief. There was no associated fever and trauma. She was referred to the Orthopaedic Surgery clinic based on clinical and ultrasonography diagnosis of epidermal inclusion cyst in the left leg.

Patient was clinically stable with normal vital signs, anicteric and not pale. In the lateral aspect of the middle third of her left leg about 15cm from the lateral malleolus was a very tender firm lump of

about 1x1x1cm in size with no differential warmth. There was neither induration of the surrounding soft tissues nor skin discoloration over and around the lump. There was no similar lump elsewhere. However there was a pre manubrium sternal keloid mass of about 6 x 2 x 1cm in size. The inguinal lymph nodes were not palpable. The neurovascular status of ipsilateral lower limb was intact.

Her haematocrit was 33.3% and the white blood cell count was within normal limit. The Erythrocyte Sedimentation Rate Westergren was 9mm/hr. Her HIV and HBsAg screening were negative. She also underwent Ultrasonography examination that shows a well-defined mass of 7.1x 6.8mm with heterogeneous content at lateral aspect of the left leg. It shows posterior enhancement and no vascularity on Doppler interrogation. The plane radiograph of her left leg showed no involvement of fibula and the surrounding soft tissue was normal. A preoperative diagnosis of epidermal inclusion cyst with neuroma and glomus tumour as possible differentials was made based on the findings from clinical and ultrasonography examination. A retrospective abdominopelvic ultrasonography revealed normal uterus and ovaries.

She underwent excision biopsy. Under local anaesthesia using 2% xylocaine with adrenaline, through a 2cm longitudinal incision, access was made to a well encapsulated nodular lump of about 1 x1 x 1cm in size in the subcutaneous plane which was not attached to the skin. The recovery after the excision was uneventful, wound healed in two weeks and there was immediate total resolution of symptoms and no recurrent mass seen on a follow up visit three months post excision.

Histopathological sections shows a benign mesenchymal proliferation consisting of long interlacing fascicles of smooth muscle cells. The cells had abundant eosinophilic fusiform cytoplasm and did not present nuclear atypia or mitotic activity. There were hyalinised areas and supporting blood vessels within the stroma. These Histopathological features are consistent with Leiomyoma as shown in Figure 1.

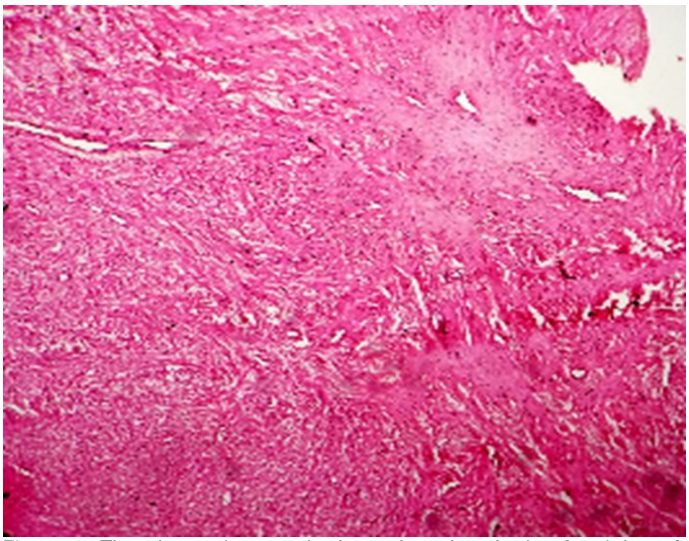

Figure 1: *The photomicrograph shows long Interlacing fascicles of smooth muscle cells. The Cells have abundant eosinophilic fusiform cytoplasm and do not present nuclear atypia or mitotic activity. There are hyalinised and supporting blood vessels seen within the stroma*

3. DISCUSSION

Cutaneous leiomyomas are rare benign smooth muscle tumours. The smooth muscles in the piloerector, vascular and external genitalia respectively gives rise to piloleiomyoma (PLM), angioleiomyoma (ALM) and genital leiomyoma (GLM), which are distinct variants of CL[3,4]. These variants can present either as a solitary or multiple cutaneous/ subcutaneous lesion[3,4,5]. The mean age at presentation varies in diverse studies; it is in a range of 38.2 to 50.8 years [9,10]. CL affects both male and female but the prevalence varies in both sexes in diverse published reports.[3, 4, 6, 9-10] The anatomical site distribution of CL also varies; the involvement of the lower extremities ranges from 9.3-43.4% of the cases in previous studies [3, 6,10]. In this patient, CL was observed as a solitary nodular lesion in the subcutaneous tissue plane in the extensor aspect of the left leg.

CL is usually painless at the onset but later becomes characteristically painful and tender.[6] However, a case of CL presenting prior to its transition to a painful form could be reported as painless. The extent this category of patient accounts for the varying prevalence of pain(54.1-89%) among the patients in diverse published reports is yet to be elucidated [5, 9, 11, 12 13]. In this patient, the initially painless lesion that became painful a year later is quite similar to the usual pattern reported by Stout in a systematic review of case series[6]. Pressure, cold, prolonged walking, trauma and light touch have all been identified as aggravating factors of the pain [6,12,13]. In this case, heat observed as a trigger factor of pain is quite different from cold that was identified as one of the common aggravating factors of pain in most previous reports[6]. However, it is similar to heat that Alam et.al reported as the least of the aggravating factors of pain among patient with multiple CL and uterine leiomyoma[13]. The reason for the trigger and worsening of pain by heat in this case of solitary CL without uterine leiomyoma is not evident.

Missed preliminary clinical diagnosis of CL is more often the norm as was the case in this patient. A cutaneous lump that was initially painless and later painful and tender can mimic an infection complicating a soft tissue tumour such as sebaceous or epidermal cyst. In this case, the attending physician mistook it for a soft tissue tumour complicated by infection then commenced antibiotic therapy that was not beneficial to the patient. Besides, CL can be easily confused with other nodular cutaneous lesions such as fibromatosis, gaint cell tumour of tendon sheet, ganglion, and glomus tumour. The diagnosis is made by subjecting the tissue to histology. Ghanadan et al reported distinct histology features for each subtype of CL: hyaline/myxoid changes are purely features of ALM that are not seen in PLM whereas pigmented rete ridges, entrapped eccrine glands and hair follicles are purely features of PLM that are absent in ALM.[5] The distinct histopathology findings in this case is consistent with ALM subtype of CL. However, the treatment of CL is the same regardless of the histologic subtype. Surgical excision of CL is curative with total resolution of symptoms. CL recurrence after excision is also very rare. The patient underwent surgical excision of the lesion and there was no recurrence as at three months post excision.

3.1 Conclusion

CL is a rare lesion and maintaining a high index of suspicion is a requisite for reduction in the high rate of missed diagnosis associated with it. Considering the paucity of published report, this case report contributes to literature and may increase awareness of this lesion among clinicians and pathologists alike especially in African setting.

Patient Consent Statement

A written informed consent was obtained from the patient

Conflicts of Interest

The authors declare no conflicts of interest

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