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(CASE REPORT)

Rare Metastasis of Cutaneous Leiomyosarcoma to the Inguinal Region: Case report and Literature review

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Abstract

Introduction: Cutaneous leiomyosarcoma (CLM) is a rare malignancy arising from smooth muscle cells in the skin. It constitutes 2%-3% of all cutaneous soft tissue sarcomas and is known for its propensity for local recurrence and distant metastasis. Common metastatic sites include the lungs, liver, and brain. Diagnosis involves clinical examination and histopathological analysis, while management typically involves surgical excision, with adjunctive therapies like radiation and chemotherapy often considered due to its aggressive nature. Metastasis rates are reported as 12% for dermal and 51% for subcutaneous CLM.

Case Report: A 40-year-old female with a history of excised leiomyosarcoma on the right buttock presented with a large, painful mass in the right inguinal region. Imaging revealed a substantial subcutaneous mass with significant local invasion and inguinal lymphadenopathy. Histopathological examination confirmed metastatic leiomyosarcoma. The patient showed anemia and elevated white blood cell count.

Literature Review: Various cases reveal that CLM typically presents as nodular masses, primarily managed by surgical excision. Metastasis is rare but can occur, particularly in subcutaneous tumors. Inguinal region metastasis is highly unusual, with most cases showing spread to distant organs or regional lymph nodes. Imaging and histopathological findings are crucial for diagnosis.

Discussion: CLM, while less metastatic compared to other sarcomas, requires vigilant monitoring due to its potential for local recurrence and distant spread. Metastasis to the inguinal region is exceptionally rare, highlighting the need for ongoing research to better understand such anomalies.

Conclusion: CLM generally has a better prognosis than other sarcomas, but early detection of recurrence and metastasis is crucial. The rare occurrence of inguinal region metastasis emphasizes the importance of comprehensive surveillance and further research to understand this phenomenon better.

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Keywords: Cutaneous leiomyosarcoma; Metastatic leiomyosarcoma; Inguinal region metastasis; Soft tissue sarcoma; Skin cancer; Leiomyosarcoma treatment; Surgical excision; Radiation therapy; Chemotherapy; Metastasis;

1. Introduction

Cutaneous leiomyosarcoma arises from the smooth muscle cells in the skin and can metastasize to other organs, including the lungs, liver, and brain. Metastatic cutaneous leiomyosarcoma (CLM) is a rare and aggressive malignant tumor that arises from the smooth muscle cells in the skin. Representing only 2%-3% of all cutaneous soft tissue sarcomas, this neoplasm typically originates in the dermis or subcutaneous tissue and has a high propensity for local recurrence and distant metastasis.[1]

Leiomyosarcomas are known for their ability to spread, often metastasizing to organs such as the lungs, liver, and brain, making management and prognosis challenging [2]. Diagnosis typically involves a combination of clinical examination and histopathological analysis to confirm the presence of smooth muscle cells and malignancy.

The treatment for CLM primarily involves surgical excision with wide margins to minimize recurrence. However, due to its aggressive nature, additional therapies such as radiation and chemotherapy are often considered. The five-year metastasis rates for dermal and subcutaneous LMS are reported to be 12% and 51%, respectively, highlighting the critical need for effective management strategies and ongoing research to improve patient outcomes.[3]

2. Case Report

A 40-year-old female presented with complaints of pain, occasional fever, and a lump in the right inguinal and hypogastric region persisting for three months. (Figure-1) Clinical examination revealed a hard, mildly tender mass measuring 12x12 cm in the right inguinal region, warm to the touch with no discharge, and accompanied by right inguinal lymphadenopathy. A digital rectal examination (DRE) showed no positive findings. The patient's medical history indicated a leiomyosarcoma on the right buttock that was surgically removed one year ago.(Figure-2)

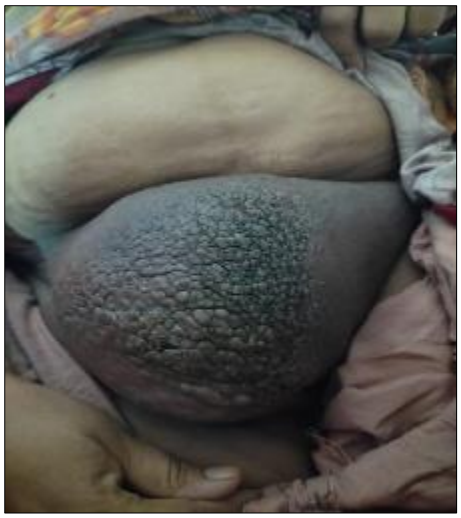

Figure 1 A large, well-defined lobulated mass in Right Inguinal region

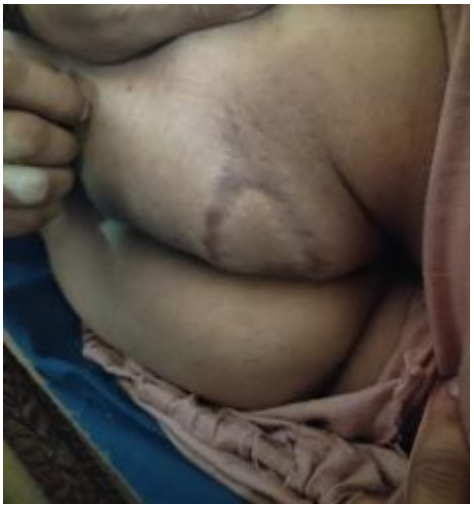

Figure 2 Surgical resection mark of Leiomyosarcoma excision on Buttocks

MRI Pelvis with contrast imaging showed a large, well-defined, rounded to lobulated mass measuring 12 x 10.6 x 12.2 cm in the subcutaneous tissues of the right lower abdominal wall and inguinal region, extending towards the midline. The mass displayed isointense signals to surrounding muscles on T1-weighted imaging (T1WI), heterogeneous signals on T2 fat-suppressed (T2FS) and T1-weighted imaging (T1WI) with areas of high and intermediate signal intensities, and heterogeneous high signals on short tau inversion recovery (STIR) images. Patchy diffusion restriction was noted on diffusion-weighted imaging (DWI) and apparent diffusion coefficient (ADC) images, with post-contrast images showing heterogeneous enhancement.

Additionally, there was increased overlying skin thickness (6.5 mm), and the mass was inseparable from the skin and subcutaneous fat in its lower part, affecting the right pectineus muscle and causing slight lateral displacement of the ipsilateral femoral artery. Enlarged peri-lesional and multiple left inguinal lymph nodes were also noted.

A contrast-enhanced CT (CECT) of the chest, abdomen, and pelvis showed no soft tissue density nodules in the bilateral lung fields, no axillary lymphadenopathy, and subcentimetric mediastinal lymph nodes. The heart and pericardium appeared normal, as did the thyroid gland, trachea, bronchial tree, pulmonary arteries, and hilar regions. Abdominal and pelvic findings included post-surgical changes in the right buttock with no recurrent or residual lesions, a normal-sized liver with no focal lesions or biliary ductal dilatation, a partially distended gall bladder, normal-sized spleen and pancreas contour, and no evidence of adrenal masses. Both kidneys were normal in size without renal or ureteral calculi, hydroureter, or hydronephrosis. There was no evidence of appendicitis, bowel obstruction, abdominal ascites, or intrinsic or extrinsic bladder masses. The bony structures were free of lytic or blastic lesions, and no free fluid was noted.

Ultrasound (USG) of the bilateral inguinal region showed a well-defined, round to oval heterogeneous lesion with a hyperechoic center, solid-looking with few anechoic areas. The lesion was located 3-8 mm beneath the skin, measuring 65.5 x 60.0 mm. There was no internal vascularity on Doppler. The overlying skin showed subcutaneous edema, with multiple adjacent inguinal lymph nodes.

Histopathology of the excised right buttock tumor revealed a fungating growth consistent with cutaneous leiomyosarcoma, measuring 16.0 x 10.0 x 6.0 cm. Lymphovascular invasion was not observed, and the resection margin was uninvolved by the tumor (1.2 cm away). The pathological stage was pT4, pN. A biopsy confirmed metastatic leiomyosarcoma in the right inguinal region.

Laboratory results showed normal bilirubin levels (0.8 mg/dL), SGPT at 26 U/L, urea at 16 mg/dL, and creatinine at 0.6 mg/dL. The complete blood count (CBC) revealed hemoglobin (HGB) at 7.9 g/dL, platelet count at 648,000/μL and white blood cell (WBC) count at 14,840/μL.

3. Laboratory Results

Table 1 Liver and Renal Function test result of the Patient

Test	Results	Unit	Reference Range
Bilirubin	0.8	mg/dL	<1.1
SGPT	26	U/L	Female: 09-36
Urea	16	mg/dL	15-45
Creatinine	0.6	mg/dL	Female: 0.6-1.1

Table 2 Complete Blood Count (CBC)

Test	Results	Unit	Reference Range
RBC	4.18	x 10^6/µL	4.0-6.0
Hematocrit (HCT)	25.4	%	35.0-55.0
Mean Corpuscular Volume (MCV)	60.8	fl	75.0-100.0
Red Cell Distribution Width (RDW)	21.5	%	11.0-16.0
Hemoglobin (HGB)	7.9	g/dL	11.0-16.0
Mean Corpuscular Hemoglobin (MCH)	18.9	pg	25.0-35.0
Mean Corpuscular Hemoglobin Concentration (MCHC)	31.2	g/dL	31.0-38.0
Platelets	648,000	/µL	150,000-450,000
Plateletcrit	0.472	%	0.108-0.282
Mean Platelet Volume (MPV)	7.3	fl	8.0-11.0
Platelets Distribution Width (PDW)	14.9	fl	9.0-17.0
Lymphocyte Percentage (LYM%)	19.2	%	20.0-45.0
White Blood Cells (WBC)	14,840	/µL	4,000-11,000

The patient presents with a large metastatic leiomyosarcoma in the right inguinal region, a recurrence from the initial leiomyosarcoma on the right buttock, which was previously excised. The current lesion shows significant local invasion and lymphadenopathy, confirmed by biopsy and imaging. Hematologic findings indicate anemia and elevated white blood cell count.

4. Literature Review

Table 3 Different cases about Cutaneous Leiomyosarcoma

Case	Demographics	Location of Lesion	Presentation	Treatment	Follow-up	Metastasis	Reference
Case1	21 cases, predominantly older adults (mean age 65)	Varied, often extremities	Varies, often presented as nodules or masses	Surgical excision	Generally favorable; focus on local recurrence monitoring	Not detailed, but generally low risk for metastasis	Soares Queirós C, Filipe P, Soares de Almeida L. Cutaneous leiomyosarcoma: a 20-year retrospective study and review of the literature. An Bras Dermatol. 2021 May-Jun;96(3):278-283. doi: 10.1016/j.abd.2020.10.003. Epub 2021 Mar 21. PMID: 33775481; PMCID: PMC8178579.
Case2	Single patient, details not extensively documented	Not specified	Described as a painful mass	Surgical excision	No recurrence or metastasis reported	No metastasis	Ciurea ME, Georgescu CV, Radu CC, Georgescu CC, Stoica LE. Cutaneous leiomyosarcoma - Case report. J Med Life. 2014 Jun 15;7(2):270-3. Epub 2014 Jun 25. PMID: 25408738; PMCID: PMC4197510.
Case3	33 patients, mostly Caucasian males, median age 63.5 years	Commonly extremities	Varies; commonly erythematous or pigmented nodules	Surgical excision, some with additional treatments	Low recurrence rates; no metastasis in most cases	25% had metastasis, typically to regional lymph nodes	Deneve JL, Messina JL, Bui MM, Marzban SS, Letson GD, Cheong D, Gonzalez RJ, Sondak VK, Zager JS. Cutaneous leiomyosarcoma: treatment and outcomes with a standardized margin of resection. Cancer Control. 2013 Oct;20(4):307-12. doi: 10.1177/107327481302000408. PMID: 24077407; PMCID: PMC4504421.
Case4	41 cases, varied age and gender	Primarily trunk and extremities	Described as firm nodules	Mohs micrographic surgery	2 cases of recurrence; no metastasis reported	50% of subcutaneous cases had distant metastases	E. Rodríguez-Lomba, I. Molina-López, V. Parra-Blanco, R. Suárez-Fernández, A. Pulido-Pérez. Clinical and Histopathologic Findings of Cutaneous Leiomyosarcoma: Correlation With Prognosis in 12 Patients. *Actas dermo-sifiliográficas/Actas dermo-sifiliográficas*. 2018;109(2):140-147. doi:https://doi.org/10.1016/j.adengl.2017.12.008
Case5	Case report; specific demographics not detailed	Lower extremity	Presented as a mass	Wide local excision with 1 cm margins	Emphasis on disease-free survival; specific follow-up details not provided	No metastasis reported	Chalfant V, Schriber T, Sabri A, et al. (April 03, 2021) Primary Cutaneous Leiomyosarcoma of the Lower Extremity: A Case Report and Literature Review. Cureus 13(4): e14282. doi:10.7759/cureus.14282

The tumors generally manifest as nodules or masses (usually painful/tender to a varying extent). The mainstay of treatment is surgical excision with additional treatments such as Mohs micrographic surgery in certain cases.

Yet, metastasis is a troubling issue as documented in Case 3 with one out of four having distant disease and by example for subcutaneous cases representing half that Episode (Case 4 had unknown but likely high malignant potential). Metastasis to the inguinal region was seen in our case, underscoring the need for surveillance against metastatic disease as well

The characteristic radiological findings on imaging such as MRI and CT are also discussed in our case demonstrating a large, well-circumscribed lesion with heterogeneously high signal intensity or dense soft tissue mass within it. An understanding of the other distinguishing features of these lesions by histopathology is essential to arrive at their diagnosis showing fungating growth consistent with cutaneous leiomyosarcoma.

In total, all these cases serve to emphasize the need for a high index of suspicion when considering cutaneous leiomyosarcoma in entity based on nodules or masses suspected among other differential diagnoses especially from any age who presented with such lesions as well as appropriate imaging and histopathological work up.

5. Discussion

Cutaneous leiomyosarcoma (CLM) is a rare skin-etiology that involves the smooth muscle cells in origin. The clinical behavior and management of the tumor can differ substantially depending on its clinicopathologic features, including site. Although CLM has a lesser tendency towards metastatic behaviour compared to other sarcomas, it is not completely devoid of the potential for distant spread especially in cases occurring at deeper locations within subcutaneous tissues.

Metastasis: Metastatic to CLM is rare, however if occur it usually present as deeper subcutaneous tumors rather than in purely cutaneous lesions. Subcutaneous Leiomyosarcomas Approximately 30-60% Subcutaneous leiomyosarcoma may metastatic, in contrast to its cutaneous counterpart which is more likely localized with rare distant metastasis [4] Most metastases were to regional lymph nodes or distantly systemically (lung), rather than the inguinal region specifically [6].

Management: The mainstay of treatment for CLM is surgical resection with negative margins. A complete surgical excision is usually curative for localized tumors. In the setting of metastatic disease, consideration may be given to chemotherapy or radiotherapy; however these treatments are less frequently used as cutaneous lesions tend not to have a high propensity for metastasis [7].

Local recurrence is a concern with CLM>: Particularly subcutaneous, high grade tumors. Therefore the rate of recurrence is reported to be approximately 50% with incomplete resection tumors [8]. Close monitoring is essential for early diagnosis of recurrences, which frequently can be controlled with further surgical resections.

Inguinal Region Metastasis: In particular, metastasis to the inguinal region is very uncommon. Unlike MCL, which can metastasize to local and distant sites in some patients, the inguinal region is not a typical site for spread of CLM. Predominant metastasis of the lesions are tend to be distant organs or regional lymph nodes rather than localized areas as such inguinal region [9].

6. Conclusion

To conclude, although Cutaneous Leiomyosarcoma in general has a better prognosis compared to other sarcomas early careful checking about any recurrence and metastasis even worst subcutaneous tumors is vital. Real rarity of inguinal region metastasis demands more research and case recording to get better understanding about the rare phenomenon.

Compliance with ethical standards

Disclosure of conflict of interest

No conflict of interest to be disclosed.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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