RESEARCH PAPER

Angiosarcoma of the Gluteal Region Masquerading as a Chronic Gluteal Abscess: A Case Report and Review of Literature

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ABSTRACT

Background: Angiosarcoma is a rare, highly aggressive malignant endothelial tumour which makes up about 2-4% of all soft tissue sarcomas. Primary angiosarcoma of the gluteal region is extremely uncommon and may present with a clinical picture of a benign inflammatory process such as a gluteal abscess, making the diagnosis difficult..

Case Presentation: We report a case of a 49-year-old female who presented with a one-month history of rapidly progressive, soft, fluctuant swelling in the lower quadrant of the right gluteal region associated with pricking-type pain of four days' duration. The swelling had no punctum, was not hot or red and was located about 3 cm from the anal verge. A well-defined soft tissue lesion was identified within the subcutaneous planes of the right paramidline inferior gluteal region on magnetic resonance imaging (MRI) which suggested a possible neoplastic aetiology due to intralesional haemorrhage. All the wide local excisions (WLE) were carried out under general anaesthetic with a margin of 1 cm. Histopathological examination (HPE) confirmed the diagnosis of angiosarcoma of soft tissue (Grade II, FNCLCC grading; ICD-O Code 9120/3). Immunohistochemistry (IHC) revealed diffuse strong immunoreactivity for CD31 and CD34, and a Ki-67 proliferative index of 25–30%. Final pTNM stage was pT2NxMx. The patient was then referred to the medical oncology department and was to be treated for the next few months with external beam radiotherapy.

Conclusion: This case highlights the critical importance of gluteal soft tissue swelling, even when clinical features resemble those of a chronic abscess, warrants a high index of suspicion for malignancy. MRI is the modality of choice, and tissue diagnosis with extensive IHC is essential for definitive diagnosis. Wide local excision with reasonable margins and adjuvant radiotherapy are the mainstays of treatment.

Keywords: Angiosarcoma; gluteal region; soft tissue sarcoma; masquerading abscess; wide local excision; immunohistochemistry; CD31; CD34; FNCLCC grading

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1. INTRODUCTION

Angiosarcoma is a malignant vascular tumor, derived from endothelial cells of blood or lymphatics and exhibits, in varying degrees, the morphological and immunohistochemical features of its vascular antecedents. It is one of the rarest and most aggressive soft tissue malignancies seen in practice and makes up about 2-4% of all soft tissue sarcomas and less than 1% of all malignant tumors. Angiosarcomas most frequently develop on the skin surface (more than 50%), such as in the face, scalp, and breast, especially after radiation therapy, and after chronic lymphoedema, as classically described in Stewart–Treves syndrome. The other primary sites are deep soft tissues, viscera (particularly liver), and bone. A primary soft tissue angiosarcoma of the gluteal area is very rare and has only been reported a few times in the English-language literature. Gluteal angiosarcoma is a rare and difficult diagnosis to make. Pyogenic infections and soft tissue masses occurring in the gluteal region are usually considered inflammatory or infectious, e.g. gluteal abscesses, pilonidal disease, perianal sepsis; the gluteal region is the most frequent site of intramuscular injection. While superficial soft tissue angiosarcomas have a typical reddish-blue integumentary appearance, deep soft tissue sarcomas are usually soft, fluctuant and slowly growing and sometimes mimic a chronic abscess. This mimicry,

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combined with the unusual location of the growth, frequently leads to a delay in diagnosis and poor initial treatment, which have a negative impact on the oncological outcomes. A rare case of primary soft tissue angiosarcoma of the right gluteal region in a 49 year old female patient with clinical presentation of gluteal abscess is presented and confirmed on histopathological and immunohistochemical analysis after wide local excision, as Grade II angiosarcoma. The objective of this report is to identify the clinical, radiological, pathological, and therapeutic differences and similarities of this unusual presentation of soft tissue gluteal mass and to stress the importance of tissue diagnosis in resolving the differential diagnosis of all gluteal soft tissue masses and to establish that it is not a benign inflammatory process

2. CASE PRESENTATION

A 49-year-old female (P3L3) presented to the out-patient department with a swelling in the lower quadrant of the right gluteal area which had been growing for one month. The swell was insidious and was fast in onset. She had developed pricking type pain over the swelling 4 days before presentation which worsened on sitting. There was no history of fever, discharge from the swelling, any lump elsewhere, loss of weight or appetite, or altered bowel habits. She had a history of Type 2 Diabetes Mellitus (2 years) and Systemic Hypertension (10 years) that were both treated regularly and was on regular medication for Dyslipidaemia and Hypothyroidism. The patient was conscious, oriented, afebrile, and had a blood pressure of 150/90 mmHg on general exam. On local examination, a 4 x 5 cm irregular swelling was seen in the lower quadrant with a normal overlying skin surface, no skin changes or punctum, and no erythema. The swelling was soft, fluctuant and mildly tender on palpation and located about 3cm away from the anal verge. No lymphadenopathy was found in the inguinal region. Digital rectal examination showed normal sphincter and no sinuses, fissures or external openings. MRI of the pelvis showed a well-demarcated lobulated soft tissue mass in the subcutaneous compartment of the right paramidline inferior buttock, containing foci of haemorrhage, which were consistent with neoplastic aetiology. Incidental findings were partial sacral agenesis with spina bifida, intramural seedling fibroid and mild left hydrosalpinx. Metastatic work up was performed. The chest computed tomography (CT) scan showed no pulmonary metastasis..

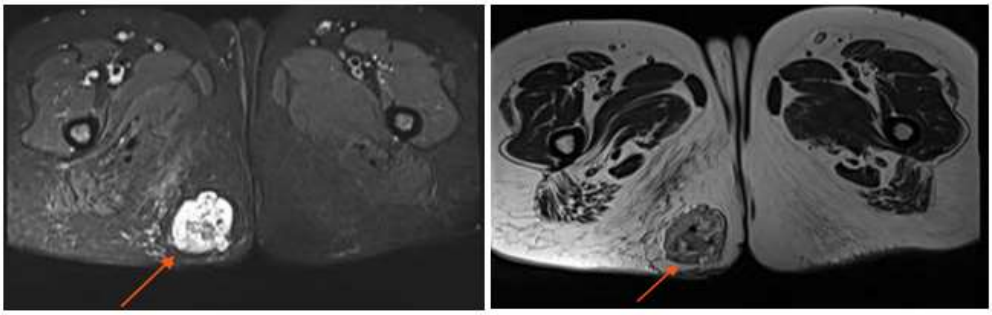

Figure 1: MRI pelvis (axial sequences) demonstrating a well-defined lobulated soft tissue lesion in the subcutaneous plane of the right paramidline inferior gluteal region (orange arrows). The lesion exhibits heterogeneous signal intensity with areas of T1 hyperintensity, consistent with intralesional haemorrhage, and is confined to the subcutaneous plane without deep muscular invasion. These features are suggestive of a neoplastic aetiology rather than an infective or inflammatory process.

Pre-operative optimisation of co-morbidities and the patient was then taken up under general anaesthetic in the prone position for Wide Local Excision (WLE). Intraoperatively, a soft tissue mass was found, located about 3 cm from the anal verge, measuring 5 × 6 cm in the right gluteal area. A circumferential skin incision was made with a 1 cm margin and the tumour was excised en bloc, haemostasis was achieved, a drain was inserted and the wound was closed layer-by-layer. Excised specimen was submitted for histopathological examination..

Macroscopic examination of the excised specimen showed a soft tissue mass with an elliptical skin fragment, 7 × 5.8 × 4.3 cm in overall dimensions. A lesion was found on serial sectioning, 4 × 3cm, grey-brown to tan-brown, friable with papillary excrescences and areas of haemorrhage. The lesion was located within 0.4 cm of the superior resected margin, 0.7 cm of the medial resected margin, 0.5 cm of the skin margin, 1 cm of the deep resected margin, and 2 cm or more of the inferior and lateral resected margins (Figure 2).

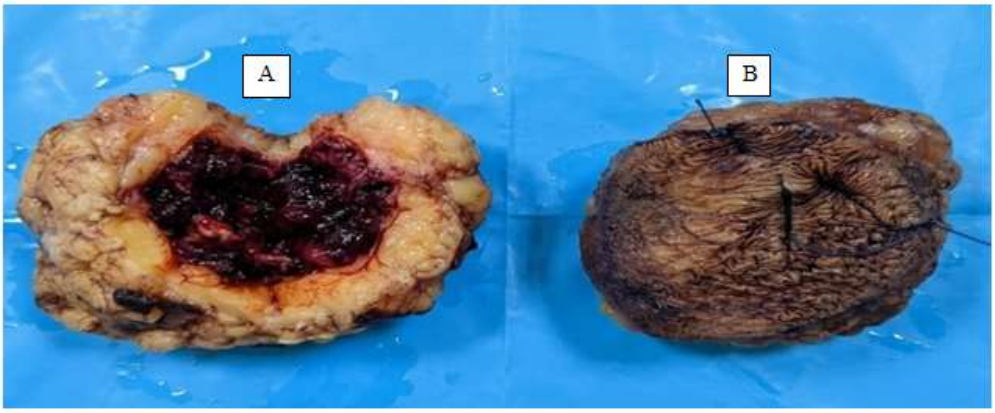

Figure 2: Gross specimen following wide local excision of the right gluteal soft tissue mass. (A) Cut section revealing an ill-circumscribed, grey-brown to tan-brown friable lesion measuring 4 × 3 cm with papillary excrescences and central haemorrhage, set within surrounding fibrofatty tissue. (B) External surface of the intact specimen (7 × 5.8 × 4.3 cm) with attached elliptical skin (5.7 × 4.5 × 0.3 cm) showing grey-brown skin surface with a linear incision defect

Histology of the excised lesion showed the presence of a well-formed papillary structure, a well-developed anastomosing network of vessels, ramifying channels of vascular tissue (vasoformative areas) and infiltrating nests of tumour in the subcutaneous tissue. Epithelioid to spindle shaped individual tumour cells with moderate cytoplasm and conspicuous nucleoli, exhibiting significant pleomorphism. Mitotic activity was vigorous at 10-12 mitoses/10 high power fields (HPF). Necretic and haemorrhagic areas were detected. All resection margins (R0) were negative for tumour, with the closest resection margin being the superior resected margin (0.4 cm); no involvement was seen in the epidermal and dermal margins, with the tumour confined to the subcutis. Vascular invasion was found. [Figure 3].

The FNCLCC histological grading gave a total score of 5 (Tumour Differentiation 2, Mitotic Activity 2 and Necrosis 1) which was considered as Grade II. Pathological stage was pT2 NxMx (final).

Immunohistochemical staining showed diffuse strong positivity for CD31 and CD34 and a Ki-67 proliferative index of 25–30% [Figures 4 and 5; Table 3]. It was diagnosed as Angiosarcoma of Soft Tissue (ICD-O Code 9120/3) Grade II (FNCLCC).

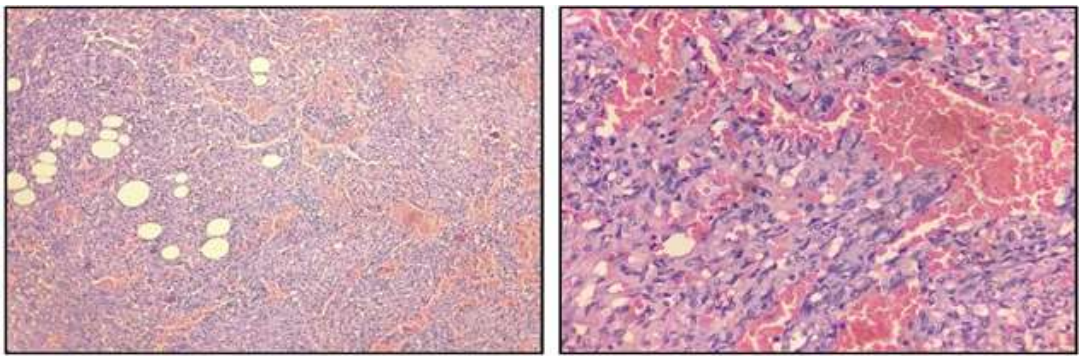

Figure 3: Haematoxylin and Eosin (H&E) stained sections. (A) Low-power view (×100) showing an infiltrating tumour within subcutaneous tissue arranged in solid sheets, anastomosing vascular channels, and vasoformative areas with surrounding fibrofatty tissue and areas of haemorrhage. (B) High-power view (×400) demonstrating tumour cells with epithelioid to spindle morphology, marked pleomorphism, conspicuous nucleoli, moderate cytoplasm, and brisk mitotic activity (10–12 mitoses/10 HPF), with extravasated erythrocytes in the background

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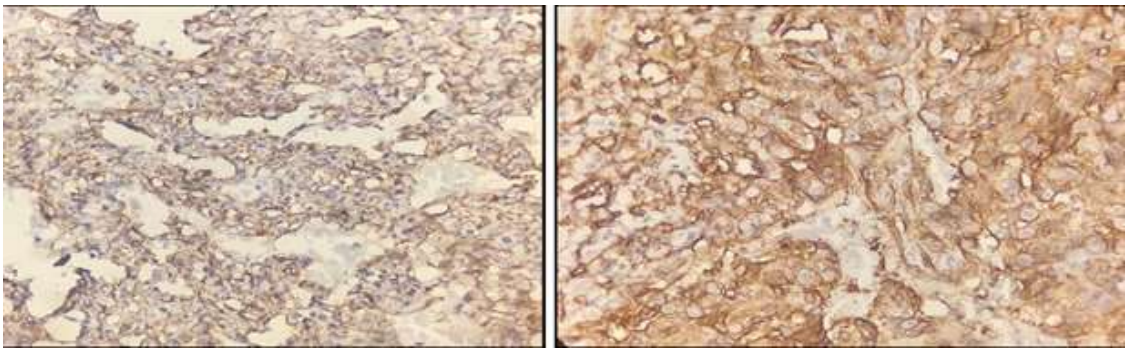

Figure 4: Immunohistochemical staining confirming endothelial differentiation. (A) CD31 (PM082) — diffuse strong cytoplasmic and membranous positivity in tumour cells, the most sensitive and specific marker for vascular endothelial lineage. (B) CD34 (PM083) — diffuse strong positivity in tumour cells, corroborating the endothelial origin of the neoplasm.

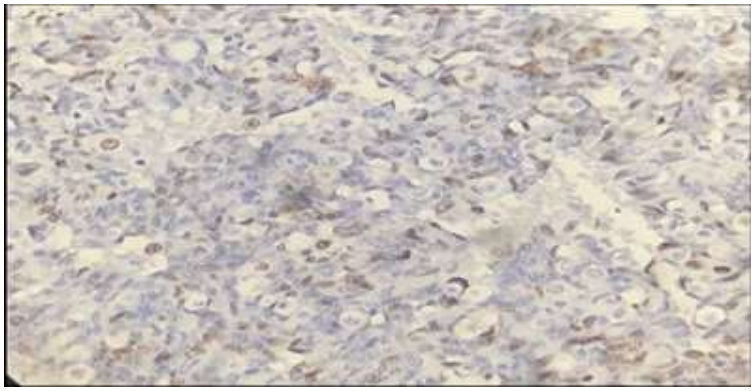

Figure 5: Ki-67 (PM210) immunohistochemical staining demonstrating nuclear positivity (brown) in 25–30% of tumour cells, indicative of high proliferative activity, consistent with FNCLCC Grade II angiosarcoma

The postoperative course was uneventful. The patient was referred to medical oncology and planned for adjuvant external beam radiotherapy (50–66 Gy to the primary site).

Table 1: Published cases of primary soft tissue angiosarcoma of the gluteal/buttock region

Author	Age/Sex	Site	Presentation	Diagnosis	Treatment	Outcome
Aust et al., 1997	58/M	Gluteal	Enlarging mass, pain	HPE + IHC	WLE + RT	–
Meis & Enzinger, 1991	52/M	Buttock	Deep soft tissue mass	HPE + IHC	WLE	12 mo (DOD)
Fury et al., 2005	44/F	Gluteal	Abscess-like swelling	Core biopsy	Surgery + CT	8 mo (DOD)
Ras et al., 2019	63/M	Gluteal	Fluctuant tender mass	FNAC + HPE	WLE + RT	18 mo (AWD)
Dhamne et al., 2021	47/F	Buttock	Subcutaneous lump	HPE + IHC	WLE + RT	14 mo (AWD)

WLE: *Wide Local Excision; RT: Radiotherapy; CT: Chemotherapy; DOD: Died of Disease; AWD: Alive with Disease;* ***HPE:*** *Histopathology; IHC: Immunohistochemistry; FNAC: Fine Needle Aspiration Cytology*

Table 2: Conditions mimicked by soft tissue angiosarcoma in the gluteal region (differential diagnosis)

Condition	Key Clinical Features	Distinguishing Feature
Gluteal/perianal abscess	Fluctuant, tender, erythematous; purulent aspirate	Fever, leukocytosis; purulent culture
Pilonidal disease	Natal cleft/upper gluteal; sinus/discharge	Post-natal cleft, pilonidal hair; MRI
Ischiorectal/perirectal abscess	Deep, tender, DRE palpable; close to rectum	DRE, TRUS, MRI pelvis
Epidermoid / sebaceous cyst	Soft, mobile, central punctum; superficial	Punctum; aspiration reveals cheesy material
Haematoma	Fluctuant; H/O trauma; haemorrhagic aspirate	Trauma history; USG shows echogenic fluid
Lipoma	Soft, non-tender, mobile; slow growing	MRI: homogeneous fat signal
Synovial / myxoid sarcoma	Deep, firm mass; no inflammatory signs	MRI heterogeneity; core biopsy + IHC

DRE: *Digital Rectal Examination; TRUS: Transrectal Ultrasonography; MRI: Magnetic Resonance Imaging; USG: Ultrasonography; GIST: Gastrointestinal Stromal Tumour; SFT: Solitary Fibrous Tumour*

3. DISCUSSION

Primary angiosarcoma of the gluteal region is very rare and only a few cases have been reported in the literature [Table 1]. The diagnostic dilemma in these situations is the clinical similarity with nonthreatening inflammatory diseases, such as chronic gluteal abscess, haematoma and pilonidal disease. Chronic lymphoedema (classic Stewart–Treves syndrome), prior irradiation, foreign bodies, vascular malformations, trauma and age per se are predisposing factors for angiosarcoma but none were found in the index case, which makes a diagnosis of a sporadic primary angiosarcoma. Cutaneous angiosarcomas have well defined, reddish-blue nodules or plaques, whereas deep soft tissue angiosarcomas in the gluteal region do not have the surface changes and appear as soft, fluctuant masses similar to purulent collections. In the present case, it was the positive fluctuation and softness that could be explained by the presence of intratumoral haemorrhage, which is characteristic of vascular tumours, and not by the presence of purulent material. Rapid onset of clinical signs, lack of antibiotic response, and absence of fever, erythema, leukocytosis, and punctum were important clinical features that led to a suspicion of neoplastic aetiology over infection. Various conditions may be mistaken for gluteal soft tissue angiosarcoma and recognition of these must be used to guide clinical assessment in all cases of unusual swellings of the gluteal region (Table 2).[2,7] Typical characteristics include a T1 isointense appearance to muscle, T2 hyperintensity and peripheral gadolinium enhancement. In our case, the presence of T1-hyperintense foci indicating intralesional haemorrhage is a unique feature of vascular tumours, and should not be treated with empirical incision and drainage as this poses a risk of tumour dissemination and wound complications. Angiosarcomas are histologically characterised by irregular vascular channels with atypical, pleomorphic endothelial cells having hyperchromatic nuclei and abundant mitoses, often with a solid sheet of poorly differentiated spindle or epithelial cells with little vasoformation, and therefore, difficult to recognise. Comprehensive IHC is therefore essential to the diagnosis [Table 3]. The CD31 (most sensitive and specific endothelial marker) was diffusely and strongly positive and the Ki-67 proliferative index was of high level (25–30%), consistent with Grade II (FNCLCC) biology. VEGF (angiogenic activity), Vimentin (mesenchymal activity) and Pan-Cytokeratin (negative in epithelial malignancy, thus completing the diagnostic panel), are recommended as supplementary markers. In our case, CT of the chest was performed to rule out pulmonary metastases and showed that the disease was localised, with a high risk of regional nodal involvement (20–30%) and haematogenous spread to the lungs (most common), liver, spleen and bone. Surgery involves R0 resection (complete resection with histologically negative margins), and the minimum recommended margin width is 1cm of grossly normal tissue (GNT). In the present case, complete resection with a confirmed R0 was achieved (margin width 0.4cm GNT). Grade II–III tumours should receive a strong recommendation for adjuvant external beam radiotherapy (50–66 Gy) to lower rates of local recurrence to 10–20% from the 40–50% otherwise anticipated. Our patient had a pT2 Grade II tumour, with no detectable metastases, which is a more favourable subset with an overall survival rate of approximately 35% for five years. However, careful follow-up by multi-disciplinary teams is important, because local recurrence and late metastases are the main modes of treatment failure.

Table 3: Immunohistochemical panel — results and diagnostic significance

Marker	Result	Significance
CD31 (PM082)	Diffuse strong positive	Most sensitive & specific endothelial marker; confirms vascular lineage
CD34 (PM083)	Diffuse strong positive	Panvascular endothelial marker; also expressed in GIST and SFT
Ki-67 (PM210)	25–30%	High proliferative activity; supports FNCLCC Grade II designation

4. CONCLUSION

Primary soft tissue angiosarcoma of the gluteal region is an exceptionally rare malignancy that has the potential to be misdiagnosed as a gluteal abscess, especially when it is soft, fluctuant and with minimal pain. If any of these is the case, a high index of clinical suspicion must be maintained for a gluteal soft tissue swelling that develops rapidly, does not have inflammatory features, or does not respond to antibiotic therapy. MRI is the imaging modality of choice for characterisation of soft tissue lesions and the presence of intralesional haemorrhage should be an immediate consideration of a vascular neoplasm. Definitive diagnosis and tumour grading requires tissue diagnosis with histopathology and the performance of a comprehensive IHC panel including CD31, CD34 and Ki-67. R0 margins with adjuvant radiotherapy is the mainstay of curative management and wide local excision. This case emphasises the need for careful oncological assessment of gluteal soft tissue masses and the dangers of misdiagnosis, and inadvertant incision and drainage of a malignant vascular tumour.

PATIENT CONSENT STATEMENT

Written informed consent was obtained from the patient for publication of this case report and accompanying images. Patient identifying information has been anonymised in accordance with the institutional policy and the Declaration of Helsinki.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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