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Uncommon presentation of retroperitoneal angiomyxoma: A case study of gluteal swelling

Kumaravelu Muthiah Kumar[1], Rosita Louis[2], Kye Mon Min Swe[3], Kavin Raj Purushottaman[4]

[1]Consultant Surgeon, Kumaravelu Surgical Clinic, Regency Specialist Hospital, Masai, [2]Senior Lecturer in Primary Care, [3]Associate Professor in Education Research, School of Medicine, Newcastle University Medicine Malaysia, Iskandar Puteri, Johor, Malaysia, [4]Resident Doctor, Department of Neurosurgery, University Hospital of Wales, Cardiff, United Kingdom

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ABSTRACT

Angiomyxoma is a rare mesenchymal tumor characterized by a myxoid stroma containing varying degrees of vascular components. While it commonly occurs in the pelvic region, it can also be found in the retroperitoneum, which poses diagnostic challenges. In this case, a general practitioner initially suspected an abscess, as retroperitoneal angiomyxomas often present as asymptomatic masses, sometimes causing discomfort or abdominal fullness due to compression of surrounding structures. This report describes a case of retroperitoneal angiomyxoma presenting as a gluteal abscess in a 46-year-old single female. She noticed the swelling 2 weeks before presenting to her general practitioner. While she experienced some discomfort, there were no significant systemic symptoms. An ultrasound and subsequent imaging, including a computed tomography scan of the abdomen and pelvis, revealed an unusual mass in the retroperitoneum. The tumor appeared as well-defined, lobulated masses with a heterogeneous signal, reflecting its myxoid and vascular components.

Key words: Abscess; Retroperitoneal neoplasms; Angiomyxoma; Tumor burden; Gluteal swelling

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INTRODUCTION

Patients commonly seek treatment from their family physicians for gluteal swellings. They present with pain, fever, and redness in the area if the swelling is inflammatory, like an abscess.[1] Rarely do swellings arise from the retroperitoneum, and this poses a diagnostic dilemma as doctors do not anticipate swellings from this area to be retroperitoneal growth. The retroperitoneal space is a potential space bordered medially by the psoas muscle and the vertebral column, anteriorly by the peritoneum and abdominal organs, posteriorly by the quadratus lumborum and the iliacus muscles, superiorly by the diaphragm, and inferiorly by the pelvis. Sarcomas comprise a third of these retroperitoneal growths, whereas benign pathologies such as neurogenic tumors, paragangliomas, and angiomyxomas are also reported. A computed tomography (CT) scan with contrast is the preferred mode of investigation. A complete surgical resection is recommended.[2]

This case report aims to create awareness among general practitioners to be cautious of gluteal swellings, as some of them can be retroperitoneal tumors in disguise.

CASE PRESENTATION

SK, a 46-year-old female, was referred by her General Practitioner on August 25, 2021, to the surgeon in Regency Specialist Centre, Johor Bahru, for a lump in the right gluteal area with concerns of an abscess. The lump was noticed by the patient 2 weeks before seeking treatment. She had no significant medical history and was not on any

Address for Correspondence:
Dr. Kye Mon Min Swe, Associate Professor, Faculty of Medical Sciences, Newcastle University, Newcastle upon Tyne, NE1 7RU, United Kingdom/Newcastle University Medicine Malaysia (NUMed), 1, Jalan Sarjana 1, Kota Ilmu, Educity@Iskandar, 79200 Iskandar Puteri, Johor, Malaysia. **Mobile:** 601-115133799. **Email:** kye-mon.min-swe@newcastle.edu.my

long-term medication. She was single and worked as an environmental officer.

Physical examination on admission showed a swelling in the right gluteal region, approximately 5 cm × 5 cm, which extended to the perineum. There was no regional lymphadenopathy, and all other systems were normal. A wide local excision was done on August 30, 2021, during which a colonoscopy was also performed. A uterine curettage showed a shedding endometrium in the proliferative phase with no other pathology (Figure 1).

Differential diagnoses include

- A right intramuscular gluteal abscess: This would be characterized by pain, swelling, redness, warmth, and tenderness to palpation
- A liposarcoma: This would be more commonly seen in an older age group and is more commonly seen in the lower extremities. The swelling would be non-tender and typically not erythematous
- A metastatic non-small cell lung cancer to the right gluteus: It is a rare site of metastases and is seen in smokers. Physical examination would likely reveal regional lymphadenopathy and pain.[3]

Laboratory investigations revealed iron-deficiency anemia with elevated neutrophils, C-reactive protein, and erythrocyte sedimentation rate. A renal profile showed mild hyponatremia and hypokalemia, hypocalcemia, and hypochloremia. A fasting glucose was found to be normal. The colonoscopy was found to be normal with extraluminal compression, but it did not hinder the passage of the scope. An ultrasound scan was done, which suggested a soft-tissue sarcoma as there were small vessels within the mass but no collections, calcification, or hemorrhage.

A CT scan with contrast showed an enormous right perineal collection measuring 15 cm × 11.20 cm × 20 cm extending into the pelvic cavity, which caused vaginal compression. Possibilities included abscesses or soft-tissue neoplasms (Figure 2a and b).

The biopsy reported that the lump, which extended to the ischiorectal fossa, measured about 190 × 120 mm and weighed 830 g. Cut sections showed a myxoid to gelatinous, brownish-gray surface with areas of congestion. It was reported as a retroperitoneal pelvic tumor with features suggestive of deep angiomyxoma of the retroperitoneum (Figure 3).

The histopathological report has shown that a partly circumscribed tumor is composed of monotonous small spindled and stellate fibroblastic cells in the background of the abundant myxoid stroma.

Figure 1: Uterine ultrasound

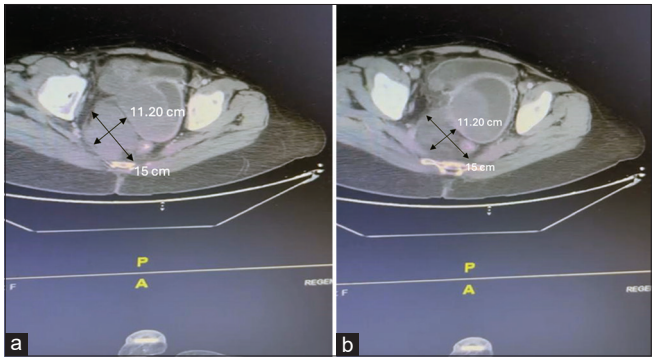

Figure 2: (a) computed tomography (CT) scan with contrast. (b) CT scan with contrast

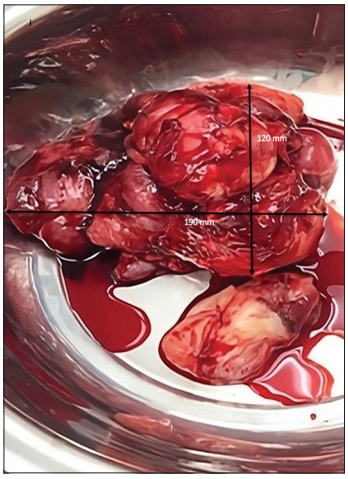

Figure 3: Biopsy specimen of the fully resected tumor

Immunohistochemistry

Reports have revealed that the tumor cells show focal positivity for CD34 and smooth muscle actin (SMA). The tumor cells are strongly positive for desmin, estrogen receptor, and progesterone receptor. The tumor cells are negative for epithelial membrane antigen, CD117, S-100, and CD99.

Surgical technique used

The surgical approach was an extraperitoneal one proceeding from the groin to the perineum. The incision was approximately 12 cm long. A wide local excision was done, during which a colonoscopy was also performed. The patient underwent general anesthesia. Careful, blunt, and sharp dissection was performed to mobilize the tumor, which was well-encapsulated but adherent to surrounding soft tissue. Special attention was given to preserving nearby neurovascular structures, particularly the sciatic nerve and gluteal vessels. Hemostasis was achieved using bipolar cautery and ligation where necessary. The tumor was completely excised en bloc without rupture. A closed suction drain was placed in the surgical bed before the layered closure of the abdominal wall.

The main challenges included:

1. The tumor's proximity to major vessels and nerves requires meticulous dissection to prevent injury to these structures
2. Dense adhesions to adjacent musculature and fascia prolonged the operative time to more than 2 h
3. The large size of the mass made manipulation and complete exposure difficult in the confined retroperitoneal space.

Post-operative outcome

The patient had an uneventful immediate post-operative recovery. The drain was removed on post-operative day 3, and the patient was discharged on day 5. At 3-month follow-up, the patient remained asymptomatic, with no evidence of recurrence on imaging. The patient initially reported mild numbness in her gluteal region, but improved over subsequent visits. The patient was counseled about the importance of regular follow-up due to the high risk of recurrence associated with angiomyxoma. Possible causes of electrolyte imbalance are passage of excessive mucus per rectum, an excess of anti-diuretic hormone, excess free water or pseudo hyponatremia, renal solute conservation, renal solute loss, or tumor lysis syndrome.[4]

DISCUSSION

Angiomyxoma is a rare locally aggressive neoplasm of mesenchymal origin that occurs mainly in adult women. In women, the vulva is commonly affected, and in men, the spermatic cord and inguinal regions are the most commonly affected organs. This condition has a female predominance (6:1) but occurs in all age groups, most commonly in the third to fifth decade of life and commonly in the perineal area. Immunohistochemically, the tumor cells are positive for CD34 and SMA, desmin, estrogen, progesterone, and SMA focally. Most tumors are idiopathic. Estrogen and progesterone positivity indicate a hormonal influence on the growth of the tumor.[5]

This case stands out from existing reports due to its unusual presentation as a gluteal swelling initially suspected to be an abscess rather than the more typical pelvic or perineal mass commonly associated with aggressive angiomyxoma (AAM). While most reported cases present with pelvic pressure, abdominal discomfort, or urinary symptoms, this patient had no systemic complaints and was managed initially under the assumption of a soft-tissue infection. The extent of tumor spread from the retroperitoneum into the gluteal region is rarely described in the literature, making this presentation noteworthy. By documenting this case, we highlight the need for clinicians to consider deep-seated neoplasms in the differential diagnosis of atypical gluteal swellings and emphasize the role of advanced imaging and histopathology in distinguishing AAM from more common soft tissue conditions. This case contributes to the growing understanding of the variable clinical spectrum of AAM and underscores the importance of early recognition to guide appropriate surgical management.

CONCLUSION

General practitioners are in a pivotal position to attend to patients presenting with gluteal swellings. Although a majority of these lesions are inflammatory, a reasonable degree of suspicion would be desirable in some cases, especially when there is a lack of systemic symptoms so as not to miss a retroperitoneal tumor. A thorough history taking, clinical examination, and radiological investigations are essential.

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Authors' Contributions:
KMK- Literature search, prepared the first draft of the manuscript, implementation of the study protocol, data collection, manuscript preparation; **RL-** Literature search, data collection, manuscript preparation, editing, and manuscript revision; **KMMS-** Review manuscript, manuscript revision and submission of manuscript; **KRP-** Manuscript preparation and preparation of figures, manuscript revision.

Work attributed to:
Newcastle University Medicine Malaysia, Regency Hospital Johor.

Orcid ID:
Dr. Kumaravelu Muthiah Kumar - https://orcid.org/0009-0004-4546-6282
Dr. Rosita Louis - https://orcid.org/0009-0002-4111-6475
Dr. Kye Mon Min Swe - https://orcid.org/0000-0001-5986-6958
Dr. Kavin Raj Purushottaman - https://orcid.org/0009-0000-2811-0920

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