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A case report angiomyxoma presenting as Bartholin's cyst

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Abstract
Aggressive angiomyxoma (AA) is a rare, slow-growing mesenchymal tumor that predominantly affects women of reproductive age and often mimics common vulvar conditions like Bartholin's cysts. We report the case of a 41-year-old woman who presented with a 6-month history of painless, progressively enlarging left vulval swelling. She sought medical attention due to embarrassment and complaints from her husband. A working diagnosis of Bartholin's cyst was made, and the patient was counselled for surgery. Intraoperatively, findings deviated from the presumed diagnosis, revealing a well-encapsulated gelatinous mass. Marsupialization with wide local excision was performed under face mask anaesthesia, and the excised specimen was sent for histopathological analysis. Histology confirmed AA. This case highlights the importance of a high index of suspicion for atypical vulvar masses, the role of histopathology in confirming the diagnosis, and the need for diligent follow-up to manage the recurrence risks associated with AA.

Keywords: Aggressive angiomyxoma, Bartholin's cyst mimicry, vulvar mass diagnosis, histopathological analysis

Introduction
Aggressive angiomyxoma (AA) is a rare, slow-growing mesenchymal tumor predominantly affecting the pelvic and perineal regions of women of reproductive age [1]. First described by Steeper and Rosai in 1983, AA is characterized by its locally infiltrative behavior and tendency for recurrence despite being histologically benign [1]. Due to its nonspecific clinical presentation and similarity to other vulvovaginal lesions, AA is often misdiagnosed preoperatively, with many cases initially mistaken for Bartholin's gland cysts, lipomas, or other benign masses [2].

The Bartholin's gland, located bilaterally at the posterior vaginal introitus, commonly gives rise to cysts or abscesses due to obstruction or infection. While such cysts are relatively common in women of reproductive age, the masquerading of a rare tumor-like AA as a Bartholin's cyst is exceedingly uncommon. It poses a diagnostic challenge; a high index of suspicion is essential for early diagnosis, especially in atypical presentations, as AA requires wide local excision to reduce the risk of recurrence.

This case report highlights a 41-year-old female presenting with what was initially suspected to be a Bartholin's cyst, later diagnosed as AA upon histopathological examination. This report underscores the importance of thorough clinical evaluation and histological confirmation in managing vulvovaginal masses. We discuss the diagnostic challenges, treatment approach, and follow-up recommendations to raise awareness among clinicians regarding this rare entity. The case adds to the limited existing literature and aims to improve AA recognition to ensure timely and appropriate management.

Patient and Management
A 41-year-old multiparous woman presented to the gynaecology clinic of a private hospital in southwest Nigeria with a 6-month history of painless left vulval swelling. The swelling had gradually enlarged over time, leading the patient to seek medical attention after her husband expressed concerns and she began to feel increasingly self-conscious. There was no associated pain, ulceration, pruritus, or discharge from the swelling. There was no history suggestive of sexually transmitted infections or trauma to the area.

The patient reported no menstrual irregularities or systemic symptoms. Her obstetric history was uneventful, with all deliveries occurring at term via spontaneous vaginal delivery. There was no personal or family history suggestive of malignancy, gynecological tumors, or any other significant illnesses. On further inquiry, she had not sought medical attention earlier due to the absence of pain and a preference for ineffective home remedies.

Her general and systemic examinations were unremarkable. Significant perineal/vaginal examination revealed a well-defined, non-tender mass in the left labium majus, measuring approximately 8 cm in its largest dimension. The mass was soft, mobile, and not attached to underlying structures, with normal overlying skin. There were no signs of inguinal lymphadenopathy.

Based on the clinical presentation and examination findings, a working diagnosis of Bartholin's cyst was made. The condition was explained to the patient, and she was counselled on the need for surgical intervention. After obtaining informed consent, she was worked up for surgery. At surgery, under face mask anaesthesia, the findings were inconsistent with the initial diagnosis. Instead of the expected cystic structure, a well-encapsulated mass with a soft, gelatinous consistency was observed. Despite this unexpected finding, marsupialization with wide excision was performed to ensure adequate drainage and prevent recurrence. The procedure was well tolerated, and the patient recovered uneventfully. She was discharged on the second postoperative day in good condition after receiving counselling on vulval hygiene and the importance of strict follow-up.

The excised specimen was sent for histopathological examination, which revealed an encapsulated lesion composed of spindle cells with oval to elongated nuclei within a myxoid stroma. Numerous thin- and thick-walled small-to-medium-sized blood vessels were distributed within the lesion, with perivascular lymphocytic aggregates observed. Notably, there was no evidence of mitosis, necrosis, or cellular atypia. These histological features were diagnostic of angiomyxoma.

Postoperatively, the patient was seen weekly for four weeks, then monthly for six months. During this period, she reported no complaints, and there were no clinical signs of recurrence or metastasis. She was counselled extensively on recognizing symptoms and signs of recurrence or metastasis, such as rapid mass growth or systemic symptoms, and advised to report promptly if any were observed.

Discussion

Aggressive angiomyxoma (AA) is a rare, benign but locally infiltrative mesenchymal tumor primarily affecting women of reproductive age [1]. Its presentation often mimics common vulvar conditions, such as Bartholin's cysts, leading to frequent misdiagnosis and delayed definitive management [2]. This case of a 41-year-old woman highlights the diagnostic challenges associated with AA and underscores the importance of a high index of suspicion for rare vulval masses.

The strength of this case lies in its meticulous documentation of clinical presentation, surgical findings, histopathological confirmation, and postoperative follow-up. Despite the initial misdiagnosis, the surgical team demonstrated adaptability in managing the unexpected intraoperative findings. The histopathological examination, revealing the myxoid stroma, spindle cells, perivascular lymphocytic aggregates, and a lack of mitosis or necrosis, confirmed the diagnosis of angiomyxoma [3, 4]. The absence of recurrence during a 10-month follow-up reinforces the importance of complete excision and diligent monitoring, as incomplete removal is associated with recurrence rates as high as 36-72% [5, 6].

This case emphasizes several lessons. Firstly, it highlights the limitations of relying solely on clinical evaluation for diagnosing vulval masses, particularly in settings with limited access to advanced imaging modalities. Secondly, it underscores the critical role of histopathology in establishing a definitive diagnosis. Thirdly, the psychological impact of vulvar masses, as seen in this patient's embarrassment and delayed presentation, necessitates a sensitive and holistic approach to patient counselling and management.

The need for a high index of suspicion cannot be overstated. While AA is rare, it should be considered in patients presenting with atypical vulval masses, especially those that are slow-growing and painless. Advanced imaging techniques, such as MRI, can aid in preoperative planning by delineating the tumor's extent and its relationship with adjacent structures [7].

Further studies are needed to explore the pathogenesis, optimal management strategies, and recurrence patterns of AA. Given its predilection for young women and potential for recurrence, research should focus on identifying molecular markers for early diagnosis and targeted therapies. Additionally, long-term, multicenter studies can provide valuable insights into the natural history of AA and refine follow-up protocols to improve patient outcomes.

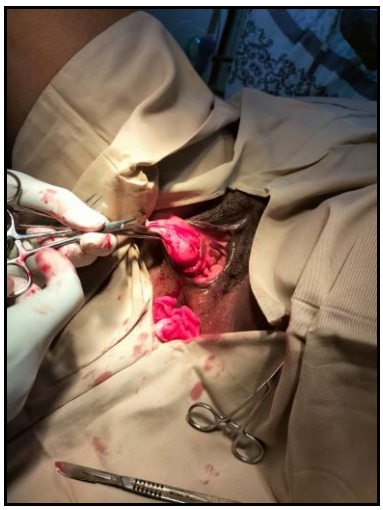

Fig 1: Angiomyxoma presenting as Bartholin's cyst

Conclusion

This case underscores the diagnostic challenges of aggressive angiomyxoma (AA), often mistaken for Bartholin's cysts. Accurate diagnosis relies on histopathology, as clinical findings may be misleading. Complete excision and careful follow-up are critical to reducing recurrence risks. The case highlights the need for a high index of suspicion, patient-centered care, and further research to improve the understanding and management of this rare tumor.

Conflict of Interest
Not available

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