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CASE REPORT OPEN ACCESS

Superficial Myofibroblastoma of the Vulva in an Adolescent; A Case Report

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Abstract
Background: Superficial myofibroblastoma (SMF) of the lower genital tract is a rare benign mesenchymal tumour first reported in English Literature in 2001 as "superficial cervicovaginal myofibroblastoma" It was formerly believed to occur exclusively in the cervix and the vagina but the name "superficial myofibroblastoma of the lower genital tract" was proposed after it was also described in the vulva. It is more common among the perimenopausal and postmenopausal women.
Case Presentation: The index report was a case of a 15-year-old secondary school student who presented in our facility with a painless vulva swelling of about 11 months duration. Examination of the vulva showed a firm non-tender mass at the mucocutaneous junction of the left labia at 2-5 O'clock position, involving the left labial majora. The hymen was intact. She had an excisional biopsy of the mass and histological findings were conclusive of Myofibroblastoma.
Discussion: It was the first case to be reported in an adolescent and the first to be reported in Nigerian or African literature.
Conclusion: Differential diagnosis abound and there is a need for closer attention to tumours of the lower genital tract to enhance its prompt diagnosis.

Keywords: Adolescent, Benign tumour, Lower genital tract, Superficial myofibroblastoma, Vulva

Background:

Superficial myofibroblastoma (SMF) of the lower genital tract is a rare benign mesenchymal tumour. It was first reported in English literature in 2001 as "superficial cervico-vaginal myofibroblastoma" (1). Since then, only a total of 52 cases have been reported in English literature to date (1, 2, 3, 4, 5, 6, 7. 8. 9. 10, 11). For many years, SMF has been known as "superficial cervico-vaginal myofibroblastoma" as it was believed to occur exclusively in the cervix and vagina but the name "superficial myofibroblastoma of the lower genital tract" was proposed after it was also described in the vulva (2). So far, only six vulvar SMFs and 5 cervical SMFs have been described while the other 41 were vaginal (1, 2, 3, 4, 5, 6, 7. 8. 9. 10, 11).
Most of the reported cases of SMF were in peri-menopausal and postmenopausal women with an

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age range of 40 to 80 years, only three were reported in the younger age group of less than 40 years with the youngest being 23 years (1, 2, 3, 4, 5, 6, 7. 8. 9. 10, 11). A case has been reported in pregnancy (7). No case has been reported in an adolescent; hence this is the first reported case in an adolescent.

Case Presentation

Patient Information

Miss O.O., a 15-year-old secondary school student presented in our facility on 7th May 2021 with a painless vulva swelling of about 11 months duration. She had no other symptoms. There was no history of trauma.

She was not yet sexually active. Her last menstrual period was two weeks before the presentation. She attained menarche at the age of 12 years and her menses had been regular.

She had never had any surgery before. She had no known medical condition. Family and psychosocial history was not contributory. A review of the system was not significant.

Clinical Findings

Her general and systemic examinations revealed normal findings.

Examination of the vulva showed a firm non-tender mass at the mucocutaneous junction of the left vulva at 2 to 5 O'clock position, involving the left labial majora as shown in Figure 1. The hymen was intact.

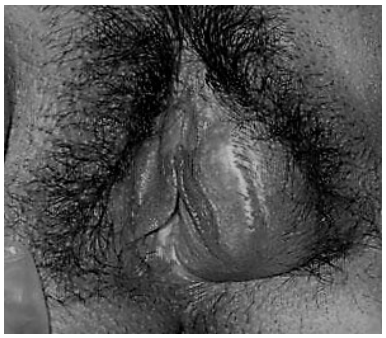

Figure 1: Clinical examination of the vulval mass involving the right labium before excision

Timeline

She had not sought treatment anywhere before the presentation.

Diagnostic Assessment

Based on physical examination, a Batholin's cyst was suspected. In preparation for surgery, she had her urine and blood collected for urinalysis and full blood count which were normal.

Therapeutic Intervention

She and her mother were counselled on the diagnosis. Consent for surgery was obtained from the mother. At surgery, the mass was discovered to be a solid mass, hence an excisional biopsy was done and the specimen was sent for histology.

Histology showed a single piece of un-encapsulated greyish-white tissue, measuring 4.5 x 3 x 1cm. Serial cut sections show homogenous greyish-white surfaces. Histologic sections as shown in Figure 2 revealed a well-circumscribed lesion composed of cellular proliferation spindle-shaped cells with oval nuclei and inconspicuous nucleoli. These cells are arranged in fascicles in a myxoid to collagenous stroma. There are hypocellular areas as well. Atypical cells are not seen. The diagnosis was Myofibroblastoma.

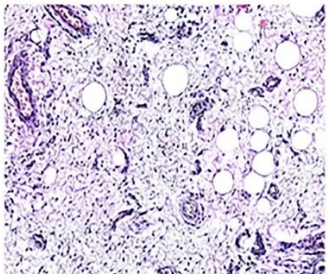

Figure 2: Histology of the excised Vulval Mass with features consistent with myofibroblastoma.

Follow-up and Outcome of Intervention
At the first follow-up, she presented the histology report of the mass. A definitive diagnosis of the lesion was then made based on the histology as "Superficial Myofibroblastoma of the Vulva in an Adolescent". She had been followed up for two years so far and there had been no recurrence.

Discussion
To the best of our knowledge, this was the first case of myofibroblastoma of the female lower genital tract to be reported in an adolescent. It was also the first to be reported in Nigerian or African literature. It was the seventh case of vulval myofibroblastoma to be reported amongst all age groups, further reiterating its rarity. Apart from the vulval swelling, there were no other symptoms. This was similar to the majority of the previously reported cases which were also asymptomatic (1, 9, 10, 11).
In this case, the tumour was solitary and measured 4.5cm x 3cm x 1cm and this was in keeping with findings in other reported cases (1, 2, 3, 5, 6, 7. 8. 9. 10, 11). Although a substantial number of reported cases had a history of use of hormone replacement therapy tamoxifen or birth control pills (1, 2, 5), we reported a case of a 15-year-old who never used any hormone-related medication or history of hormonal disorder. Diagnosis of genital mesenchymal tumours is quite challenging due to their rarity and similarity in their anatomic locations and pathological features and the index case was initially confused for a Batholin's gland duct cyst due to its anatomic location as seen in Figure 1. Initial misdiagnoses were also recorded in many of the previous cases (1, 7, 8, 9, 11). All the cases of vulval myofibroblastoma were treated by local excision (2, 5, 10, 11) which was what was also offered in the index case.
Our case has been followed up for two years and there was no recurrence. This also is in tune with the low recurrence rate of the tumour as reported in the case series as only one case of recurrence had been recorded so far in the literature (1, 2, 3, 4, 5).

Conclusion
Myofibroblastoma of the lower genital tract are rare but rarer are those involving the vulva. They are generally more common among the peri-menopausal and post-menopausal women and this was the first case to be diagnosed in an adolescent. Differential diagnoses abound and there is a need for closer attention to tumours of the lower genital tract to enhance its prompt diagnosis

List of Abbreviation
SMF: Superficial Myofibroblastoma

Declarations
Ethical approval and consent to participate
The mother gave informed consent for the surgery. The patient and her mother also permitted the publication of the case in a medical journal for the advancement of medicine.

Consent for publication

Availability of data and materials
All essential data supporting the findings of this case are available within the article. Additional data are available upon request from the corresponding author.

Competing interests
The authors declare no conflict of interest.

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Authors' contributions
AOR: Managed the case, Literature search, initial manuscript drafting, final proof reading.
OOI: Managed the case, initial manuscript drafting, final proof reading.
OOO: Manuscript editing, manuscript review and final proof reading.
AAO: Literature search, manuscript editing, manuscript review and proof reading.
GEO: Literature search, manuscript editing, manuscript review and proof reading.
AOA: Manuscript editing, manuscript review and proof reading.
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