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Breast Phyllodes Sarcoma in a 12-Year-Old Girl: A Case Report

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Abstract

Introduction Phyllodes sarcomas are rare malignant tumors of childhood and adolescence. **Objective:** To analyze the epidemiological, diagnostic and therapeutic management of this pathology, highlighting its challenges in an undermedicalized country. **Case presentation**: We report the case of a 12-year-old girl who presented with phyllodes sarcoma of the left breast. Medical nomadism and traditional treatment contributed to late management. In her case, we performed a simple mastectomy with passage into healthy zones, confirmed after histological analysis. Follow-up was straightforward until the last news, two years after our care. **Conclusion:** Phyllodes sarcoma is rare but can occur in children. Any mammary nodule in children should be referred to a specialist. Traditional treatment should be avoided.

Keywords

Phyllodesarcoma, Pediatrics, Mastectomy

1. Introduction

Mammary pathology in children and adolescents is known to be essentially benign. However, malignant etiologies may be encountered in the course of breast tumors at this age. Their incidence is estimated at 0.08 per 100,000, with a ratio of less than 1% for sarcomas [1]. Breast sarcomas are a heterogeneous group including fibroepithelial proliferations commonly referred to as phyllodes sarcomas (PS) [2]. SPs are therefore extremely rare malignant breast tumours in the paediatric population.

A recent retrospective review conducted in the USA confirms the rarity of Pediatric Phyllodes Sarcoma (PPS), with an estimated frequency of less than two cases per year [3]. Again according to this study, PPS have a variable, non-specific

clinical profile, similar to adult female phyllodes sarcoma. However, the rarity of breast cancer at this age often leads to misdiagnosis. Only histological examination can clarify the diagnosis of PPS. Treatment of PPS is based on surgery, and overall survival appears to be better in young patients than in the adult population, according to *Yunxiao* [3].

We report an unusual case of a mastectomy without axillary curage performed for a voluminous phyllodes sarcoma of the left breast in a 12-year-old child with a history of medical nomadism. The aim of this rare case was to discuss the epidemiological, diagnostic and therapeutic aspects of the management of this pathology, highlighting its challenges in an under-medicalized country.

2. Case Presentation

2.1. Anamnesis

K.M. was a 12-year-old schoolgirl who had not yet gone through menarche. She was the last child and only girl in a sibling group of 4 children. She had no particular background. She lived with her parents in a rural area 432 km from the economic capital Abidjan. Her personal and family senological history was unremarkable. The girl had been evacuated to the Gynaecology and Obstetrics Department of the Treichville University Hospital for a presumed large abscess of the left breast. On admission, the history revealed a retroareolar nodule that had appeared in the left breast six months previously. The nodule was painless and increasing in volume. The parents stated that they had consulted a general practitioner as there was no paediatrician in the area. The GP concluded that the abscess was an uncollected breast abscess and prescribed an unspecified course of antibiotics. Given the unfavorable course of the disease despite compliance with the treatment prescribed by the doctor, the parents would have resorted to traditional topical treatment. The appearance of ulceration-like skin changes and a monstrous increase in breast volume would have prompted a further consultation with the general practitioner, who promptly referred her to us for further treatment.

2.2. Diagnosis

On physical examination, the patient's general condition was average, with infectious facies. In the left breast, there was a large, firm mass measuring 23 cm in long axis, with loss of substance that had washed away the areola-mammary plate (**Figure 1**). A 2 cm mobile adenopathy was palpated in the homolateral axillary fossa. The right breast and its lymph nodes were normal. No imaging examinations had been performed due to the parents' poverty. We therefore suggested superinfection of either a giant adenofibroma or a breast carcinoma, but were not convinced of a malignant etiology.

2.3. Management

Trimming was performed with multiple biopsies in healthy areas for anatomopathological examination. Ten days later, the histological result indicated a

phyllodes sarcoma. Following a multidisciplinary oncology consultation, the decision was made to perform a mastectomy without delay. Enlightened information had been given to the parents, who had agreed to the mastectomy, and the extension work-up had not been carried out, again due to the parents' indigence. Histological analysis of the mastectomy specimen showed proliferation, confirming a phyllodes sarcoma (Figure 2). Exeresis margins were healthy

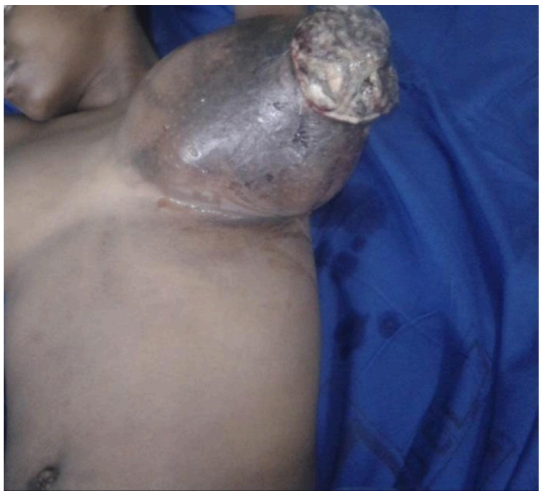

Figure 1. Large tumor in left breast.

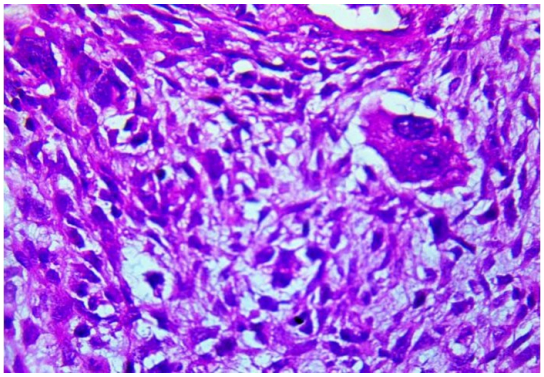

Figure 2. (HEX400): Phyllode sarcoma with the presence of marked cytonuclear atypia and multinucleated giant cells.

2.4. Follow-Up and Prognosis

The immediate post-operative course was straightforward, and the parents were offered psychological support. Complete healing was achieved after 21 days (Figure 3). Follow-up was uneventful until the patient was lost to follow-up two years later.

3. Discussion

3.1. Epidemiology

This case raises a number of questions concerning this unusual case of phyllodes

sarcoma in a child. In the breast, there is a group of fibroepithelial proliferative lesions (*i.e.* both epithelial and stromal tissue) represented essentially by adenofibromas in almost 97.5% of cases, and phyllodes in 2.5% [2]. Adenofibromas are frequent tumours in children and young teenagers (4 = Aworolo), while phyllodes are mainly found in adult women from the fourth decade of life onwards ([2] [3]. According to the WHO classification, three types of phyllodes tumors can be distinguished according to their evolutionary potential: grade I PTs corresponding to benign tumors, grade II PTs designating tumors bordering on malignancy or borderline, and finally grade III PTs, which are malignant tumors known as phyllodes sarcomas[2], such as the case we present in this very young patient. In the USA between 1976 and 2015, out of a total of 2773 patients with phyllodes sarcoma, Yunxiao Xiao *et al.* found only 60 patients under 21 years of age [3] [4]. Moreover, the characteristics of their population showed a higher proportion of MS in young black patients (34%) than in adults of the same race (10.4%). Apart from these large series, only clinical cases have been reported for this rare childhood disease. Our 12-year-old patient was the first case of pediatric MS encountered in our practice in 15 years. Other authors before us had also reported cases of phyllodes sarcoma in children or adolescents [5] [6]. Because of their rarity, the etiopathogenesis of MS remains poorly understood, although these tumours are usually observed in women, as in our case.

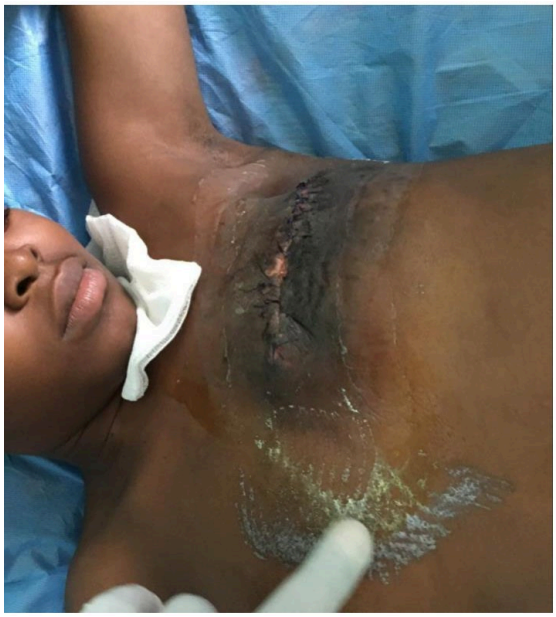

Figure 3. Mastectomy scar on day 14.

3.2. Diagnosis

Diagnosis of MS is difficult compared to that of adenofibromas. The clinical signs of MS are non-specific, and there are no pathogenetic signs for this disease. In children, over 90% of breast tumours are benign [4]. In the presence of a mammary tumour, it is usual to first evoke an adenofibroma, or even a juvenile

adenofibroma if tumour growth has been rapid [4]. Although malignant etiologies do exist at this age, they are rarely evoked during a clinical examination, due to their rarity. However, if clinical signs seem suggestive, such as the presence of pain, large tumour size or necrosis, breast carcinoma may be evoked, as in our case, since it is by far the most frequent malignant cause of breast tumour in children [4]. Rarely, phyllodes sarcoma is immediately evoked. Tekbas in Turkie suggested a giant adenofibroma in the presence of a large breast mass in a 14-year-old girl, which later turned out to be a breast sarcoma [7].

The clinical features of breast sarcomas include rapid tumor growth and pain, which are found in 30% of cases [2]. The mean tumour size at diagnosis is 4.3 cm, but may be larger [8]. These signs were present in our young patient, in whom pain had progressively set in, with a voluminous 23 cm mass developed over six months. These late signs could be related to the medical nomadism found in our patient's care. In addition, our patient had not benefited from the use of traditional African medicine. This practice, which is frequent in our country, should be the subject of ongoing awareness-raising campaigns to warn the population of the limitations of this medicine for childhood breast tumour disorders.

During the management of our care, we did not perform any imaging examinations due to the family's indigence. Although the tumour was obvious on inspection and not deeply fixed on palpation, a thoracic CT scan would have been appropriate. This would have enabled the ratio of the mass to the chest wall to be assessed. Ultrasound and MRI are the imaging examinations of choice for mammary exploration in paediatric patients, but they only show the usual signs of lesions presumed to be benign [6]. In our practice, if ultrasound remains an accessible examination, this is not the case for MRI, which costs around US$200.

3.3. Treatment

The treatment of breast sarcoma in young girls does not differ from that of adult women. The aim is local control of the disease, and surgery remains the standard treatment. Healthy tumour margins remain the main prognostic factor in this disease. According to the guidelines of the National Comprehensive Cancer Network (NCCN) in the USA, this surgical excision should be wide, with healthy margins of at least 1 cm, and without the need for axillary curage [3] [8]-[10]. This surgical condition would allow better local control, thus avoiding recurrence. The prognosis of this pathology remains mainly linked to the quality of surgical excision. For Yunxiao Xiao, young age seems to be a protective factor in the prognosis of MS [3]. Other therapeutic means are used in the treatment of breast sarcoma, notably radiotherapy and systemic treatment with cytotoxic agents, but their impact on overall survival seems uncertain [2] [8].

The current state of knowledge about pediatric MS suggests the need for large-scale prospective multicenter studies of this disease, including our African regions. This would help to better understand the etiopathogenesis and management of phyllodes sarcoma in the paediatric population.

4. Conclusion

Mammary tumors in children are generally benign; however, the possibility of a malignant etiology such as phyllodes sarcoma remains. The mere presence of a palpable lesion at this age should, in our context, lead to a de facto referral to a specialist for early and appropriate management. In addition, we need to continue raising parents' awareness of the harmful use of traditional medicine for breast pathologies in general, and children's pathologies in particular.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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