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Case Report

A rare case of a giant ulcerated benign phyllode tumor

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ABSTRACT

Phyllode tumors are rare fibroepithelial neoplasms divided into three histological grades according to their potential for malignancy. Low grade tumors are usually smaller with a slower evolution.

We present here the case of a 40 year old women presenting an augmentation over one month of the volume of her right breast with an ulcerated mass measuring 25 cm. The clinical characteristics were in favor as a malignant tumor. The radiological findings were not contributive and the pathologic examinations of the biopsies were benign. A therapeutic and diagnostic mastectomy was thus performed and the final pathologic examination confirmed a benign phyllode tumor.

These rare tumors are very deceitful for the radiologic investigations are poorly contributive. Likewise, the pathologic examination can be incorrect. Biopsies must be repeated and a mastectomy performed if any doubts remain.

1. Introduction

Mammary phyllode tumors are rare fibroepithelial neoplasms occurring predominantly in middle-aged women (45–50 years old) and account for 1% of all primary breast tumors [1]. These tumors are histologically very similar to fibroadenomas except for the presence of leaflike projections in the stroma and an increased stromal cellularity [2]. They are classified in three subgroups: benign, borderline and malignant. Usually, malignant phyllode tumors have a very rapid and aggressive evolution whereas benign tumors are smaller with a slower evolution. However, low and intermediate grade phyllode tumors have a risk of local recurrence of 15 and 17% and a potential for metastasis of 0.1% and 0.2% [3]. The key problem is the pre-operative diagnosis. Indeed, the sensitivity of needle core biopsy and imaging for the diagnosis of phyllode tumors are only to 63% and 65%, respectively [4].

We report here the case of a patient presenting an unusual form of giant benign phyllode tumor with all the clinical and radiological findings matching a malignant, aggressive tumor.

2. Clinical case

A 40 year-old Filipino woman consulted for a rapid augmentation of the volume of her right breast over the past 6 weeks. This patient had no prior illness. She had never undergone surgery and was nulligest. No case of breast, ovarian or colo-rectal tumor was found in her family. Upon physical examination, we found a mass of 25 cm, adherent to the pectoral muscle with an ulcerated outgrowth over 5 cm large of the interior-superior quadrant (Fig. 1, Image D and E). The left breast was normal with an 85D cup. There were no palpable axillary or sus-clavicular nodes. The ultrasound performed found a heterogeneous vascularized mass of 20 cm with a suspect axillary node of 10 mm level 1 of Berg classification (Fig. 1, Image A). The mammography was impossible because of the size and the procedure was too painful. Likewise, the patient could not undergo a conventional MRI since the tumor was too large to use a breast antenna. The classic body MRI confirmed the presence of a 23 cm × 14 cm × 23 cm mass with a heterogeneous T2 signal and important vascularization after injection compatible with a sarcoma (Fig. 1, Image B and C). The tumor was fixed to the pectoral muscle but no sign of invasion was found. The thoraco-abdomino-pelvic scan detected no metastasis.

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Fig. 1. Image A. Ultrasound of the right breast showing the regular borders of the mass and the intense internal vascularization. Image B and C. Injected T2 axial (B) and sagittal (C) MRI showing the heterogeneous aspect of the mass and the well-circumscribed edges with no infiltration of the pectoral muscle. Images D–F. Initial photographs of the mass (D and E). Pre-operative photography, a week later, showing the rapid evolution and the spread of the ulceration (F). Image G and H: Macroscopic photography of the mass (G). Microscopic view with intra-canalicular architecture and typical leaf-like projections (hematein-eosin-saffron, original magnification ×20) (H).

The cytological examination of the axillary node found no malignancy. Pathological examination of an ultrasound-guided needle core biopsy showed a fibro-epithelial neoplasm compatible with an adenofibroma or a benign phyllode tumor. No significant stromal atypia was found and the mitotic index was low. A cutaneous biopsy on the ulcerated part of the mass was performed and did not identify any malignant cells. The pathologic examination was on behalf of a cellular fibroma or a phyllode tumor of low grade. A TEP-scan was urgently carried out and revealed a strong heterogeneous fixation on the breast (SUV 8.4), with suspicious axillary and pectoral nodes. No other distant metastasis was found.

Finally, all the clinical and radiological findings pointed towards an aggressive tumor such as a sarcoma whereas the histological diagnosis was consistent with a low grade phyllode tumor. The only reassuring element was the well-circumscribed aspect of the mass with no infiltration of the muscle. Because of the discrepancy and the difficulties to determine the adequate surgical procedure with the potential necessity of a flap, the committee of oncology recommended a surgical biopsy. The biopsy was performed on the most suspicious area on the TEP scan and another biopsy was done on the most ulcerated part of the breast. Once more, the histologic examination revealed a low grade phyllode tumor. The neoplasm had continued to increase in size and had a local infection treated by antibiotics. Another ulceration appeared on the surgical biopsy site confirming the aggressiveness of the neoplasm (Fig. 1, Image F).

It was thus decided to perform a therapeutic and diagnostic mastectomy. Breast conservation was impossible due to the size of the tumor. We performed a simple mastectomy with an elliptical incision. Only an abdominal advancement flap was necessary. The suspicious axillary node was removed and the frozen sections were benign. No axillary dissection was carried out. Immediate breast reconstruction was not carried out since the nature of the mass was still uncertain and adjuvant treatment such as radiotherapy or re-operating for wider margins was not ruled out.

The excised specimen weighed 4000 g and measured 29 cm × 24 cm × 14 cm (Fig. 1, Image G). The final pathological examination concluded to a benign phyllode tumor (Fig. 1, Image H). The resection margins were less than a millimeter. The axillary node was free of malignant cells. A mammary reconstruction was proposed to the patient after at least a follow-up of 6 months.

3. Discussion

Many features in this case are very unusual for a typical benign phyllode tumor. Classically, patients present a firm, well-limited, round, painless mass [5]. Here, the patient presented an ulcerated, bleeding, infected mass. Moreover, the patient described a rapid evolution in less than a month. We witnessed this dramatic evolution where another ulceration appeared in less than a week on the site of the surgical biopsy. There is little data available on how fast a benign phyllode tumor can evolve but most agree it takes several months [6–9]. The final measurement confirmed a tumor of 29 cm. Usually, these tumors measure 4 cm and a mass measuring over 10 cm is qualified as a giant tumor. Barrio et al. had found that a tumor size over 3 cm was significantly more frequent in malignant tumors [1].

The radiological findings could not conclude on the grade of the neoplasm. The tumor was very heterogeneous, vascularized with an axillary lymph node. Yet, the mass remained well circumscribed with very regular edges and no invasion of the pectoral muscle. No radiologic pathognomonic signs have been described to differentiate the grade of the tumor [10]. The pre-operative MRI remains essential to guide the surgical treatment. The invasion of the pectoral muscle and the adjacent structures can change the eventual reconstruction.

Because of all these clinical and radiological abnormalities, we insisted and repeated the histologic exams. Core needle biopsies have been found to have sensitivity and a positive predictive value very variable from one study to another. The positive predictive value of the core needle biopsy can differ from 52.7% to 87% [3,11,12]. Due to the heterogeneous nature of phyllodes tumors, it is known that the choice of the location for the biopsy is essential and the results can strongly vary from one sample to another [13]. More importantly, since the imagery is of little added value, it is necessary to perform macro-biopsies guided by an ultrasound with as much histological material as possible. We thus performed a surgical biopsy in order to confirm our clinical suspicion of malignant tumor.

The pathologic confirmation of a phyllode tumor is usually made using HES only (hematein-eosin-saffron) coloration while the use of immunohistochemistry (IHC) is less common [14]. IHC is mainly used in differentiating phyllode tumors from sarcomas or fibroepithelial tumors. Cimino-Mathews found that cytokeratins labeled 21% of malignant phyllodes tumors (focal) and 100% of sarcomatoid carcinomas. Likewise, focal p63, p40, and cytokeratin labeling can be seen in malignant phyllodes but never in low grade fibroepithelial tumors [15]. Its role in differentiating grade is less established. Indeed p53, CD117, EGFR, VEGF, CD10 and Ki67 correlate with grade but must be used with caution [14,16].

The demographic characteristics of our patients were classic. Asian women tend to be younger on average when diagnosed with a malignant phyllode tumor [1]. They also present a higher recurrence rate [5]. The presence of hypertrophic lymph nodes is not typical in a low grade phyllode tumor. When present, they are usually inflammatory and less than 1% are metastatic [5]. Our patient presented a reactive lymph node, free of malignancy. Hence, lymph node dissection is not systematic since the spread of the disease is hematological rather than following a lymphatic route. Other similar cases of giant benign phyllode tumors have been issued. Yan et al reported the case of a 54 year-old woman presenting a 22 cm well circumscribed benign phyllode tumor. However, no ulceration was present and the evolution had been progressive over the past six months [9]. Reports of giant benign ulcerated phyllode tumors measuring less than 25 cm have been published but non were as large as our patient's tumor [6–8]. One case was particular similar to our patient. Islam et al reported the case of a 45 years old patient presenting a neglected 50 cm ulcerated mass of the right breast. The histopathology, reviewed by two senior pathologists, found a benign phyllode tumor. The patient however presented a massive pleural effusion several month lateral with malignant cells revealed at cytology [6].

Even though it is still debated, actual guidelines recommend an excision with one-centimeter margins for a low grade and borderline phyllode tumor [17]. French recommendations accept an in sano resection for low grade phyllode tumors [3]. A conservative treatment can be discussed if the cosmetic results are satisfying with no impact on the disease free survival [18]. In our case, only a total mastectomy could be considered. We discussed an immediate breast reconstruction, but our clinical suspicion of malignancy was on behalf of a radical treatment with a differed reconstruction. If the margins had been invaded a revision surgery would have been needed with repercussions on the reconstruction.

4. Conclusion

This case clearly demonstrates that phyllode tumors can show unusual clinical features, mimicking malignant tumors. Imagery can seldom conclude of the grade on the tumor. Likewise, biopsies can misdiagnose a malignant tumor. Yet, there needs to be an accurate diagnosis in order to treat the patient accordingly. Research must focus on new techniques such as intravoxel incoherent motion magnetic resonance imaging and immunohistochemistry to offer a precise diagnosis per-operatively to the patient.

There is no clear recommendation on the modalities of the follow[HYPHEN]up for low grade phyllode tumors. Because of the high risk of local recurrence (15%), recent French guidelines recommend a biannual clinical examination with an annual mammography and breast sonography for ten years [3].

Disclosure of interest

The authors declare that they have no competing interest.

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