CASE REPORT – OPEN ACCESS

International Journal of Surgery Case Reports 80 (2021) 105644

Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

The malignant degeneration of a phyllode tumour in man: A case report

Mohamed Boudou [a,*], Rachid Jabi [a], Abdelali Guellil [a], Anass Haloui [b], Mohammed Bouziane [a]

[a] Department of Visceral Surgery and Digestive Oncology A, Mohammed VI University Hospital, Oujda, Morocco
[b] Department of Anatomopathology, Mohammed VI University Hospital, Oujda, Morocco

ARTICLE INFO

Article history:
Received 10 January 2021
Received in revised form 8 February 2021
Accepted 8 February 2021
Available online 13 February 2021

Keywords:
Male
Breast
phyllodes
Malignant
Case report

ABSTRACT

INTRODUCTION: Malignant phyllodes tumours of the breast represent less than 1% of all breast cancers.

Few cases of phyllodes tumours have been reported in men.

CASE PRESENTATION: We present the case of a 60-year-old man who was operated on one year ago for a breast tumour that had undergone a lumpectomy with an anatomopathological study in favour of a grade 2 phylloid tumour. He was admitted to hospital with a palpable mass in his right breast. The lumpectomy enlarged to the right pectoralis major muscle was then performed with clear surgical margins.

Microscopic examination revealed high-grade malignant phyllodes.

Postoperatively, after 3 months, the patient was given a breast MRI and a PET/CT scan which returned without abnormalities.

The patient is followed for eight months and has shown no signs of recurrence.

DISCUSSION: Malignant phyllodes tumours of the breast show clinical and mammographic signs comparable to those of benign lesions.

The diagnosis is confirmed by histology, treatment is based on surgery, which may be a large lumpectomy or mastectomy, and the prognosis depends on several factors, the most important of which is the margin for surgical resection.

CONCLUSION: The best treatment is a wide local excision with a safety margin of 1 cm, unless it is metastatic.

Early diagnosis and surgery improves the prognosis.

1. Introduction

Malignant phyllodes tumour are rare malignant tumours of the breast, they belong to the group of phyllodes tumours which can be benign, borderline or malignant with a very rare incidence in men and few cases reported in the literature, generally preceded by gynecomastia. The best treatment is wide local excision with a safety margin of 1 cm.

All three pathologies (benign, borderline and malignant) were reported in men [1].

The anatomopathological analysis of tumours requires a reference anatomopathologist and an immunohistochemical study for confirmation.

Through this clinical case and a review of the literature, we wish to highlight the circumstances of the occurrence of these tumours, the difficulties of clinical and paraclinical diagnosis as well as the therapy.

This work has been reported in line with the SCARE criteria [2].

2. Case presentation

A 60-year-old man, currently smoking 15 cigarettes per day, recently cut down from 22, without any family history, operated 1 year ago for a breast tumour having undergone a lumpectomy with an anatomopathological study in favour of a grade 2 phyllode tumour with unpredictable evolutionary potential in the right breast.

He was recently admitted for local recurrence with a palpable mass, located at the superior lateral quadrant of the right breast.

On clinical examination, the patient was hemodynamically and respiratorily stable, apyretic, weight =85 kg, height = 1.75 m, and BMI: 27, 8.

The senological examination showed a lumpectomy scar about 7 cm long, a nodule of the upper lateral quadrant of the right breast about 4 cm long, with irregular margins, painful on palpation,

* Corresponding author.
E-mail address: boudoumohamed0@gmail.com (M. Boudou).

https://doi.org/10.1016/j.ijscr.2021.105644

Table 1
Histopathological criteria for diagnosis of phyllodes tumour.

Criteria	Benign	Borderline	Malignant
Stromal cellularity and atypia	Minimal	Moderate	Marked
Stromal overgrowth	Minimal	Moderate	Marked
Mitoses/10 high power fields	0–4	5–9	≥10
Tumor margins	Well circumscribed with pushing tumour margins	Zone of microscopic invasion around tumour margins	Infiltrative tumour margins

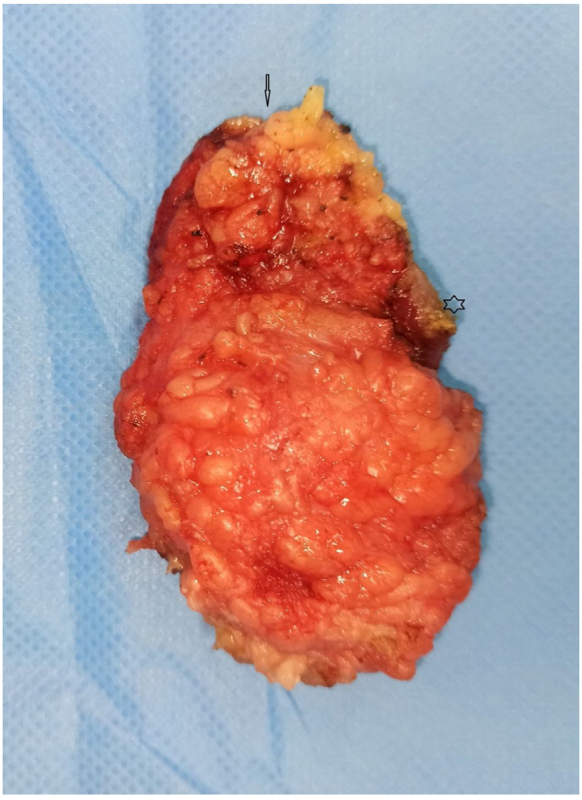

Fig. 1. Breast lumpectomy specimen (arrow: upper pole; star: section slice of pectoral muscle).

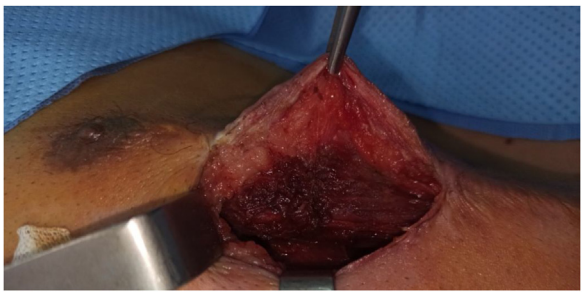

Fig. 2. Incision and the residual cavity after lumpectomy.

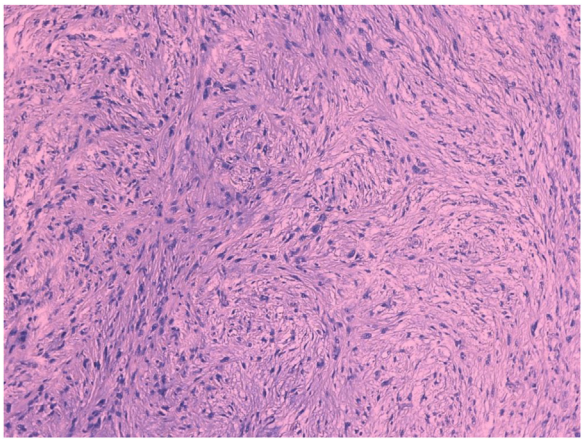

Fig. 3. Tumoral proliferation of spindel cells, essentially arranged in a whorled and a fascicular pattern.

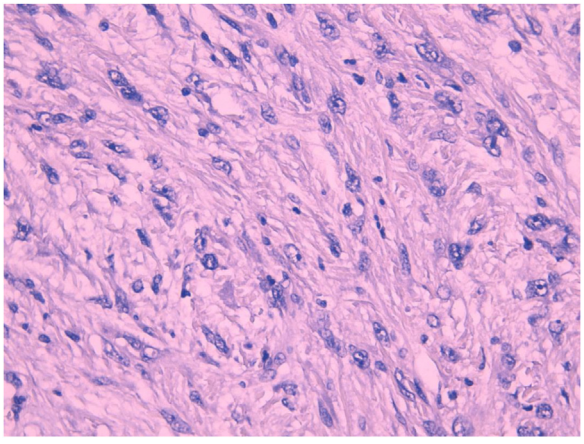

Fig. 4. Histological aspect of tumour cell atypia, with moderate to marked nuclear pleomorphism and strong mitotic activity.

adhering to the deepest part of the body, without any inflammatory signs.

Axillary lymphadenopathy was negative and there were no palpable supraclavicular nodes.

The left breast was normal.

ultrasonography showed a hypoechoic mass with partially defined contours measuring 4.5 × 3.0 cm, located in the upper lateral quadrant, with intra-tumoral micro calcifications, corresponding to a fibroadenoma or phylloid tumour.

The lumpectomy enlarged to the right pectoralis major muscle was then performed, due to infiltration of the pectoralis major muscle (Figs. 1 and 2).

Grossly, the lumpectomy measured 7 × 4 × 3.5 cm. On section, a nodular lesion was observed, measuring 1.2 × 2.2 × 4 cm, of firm consistency and whitish in color. Areas of hemorrhage and necrosis were not seen (Fig. 1).

Histological examination revealed a tumoral proliferation of spindel cells, essentially arranged in a whorled and a fascicular pattern (Fig. 3).

The tumoral cells were highly atypical, showing moderate to marked nuclear pleomorphism, and a brisk mitotic activity. The mitosis was estimated at 22 mitosis/10 HPF (Fig. 4).

Immunohistochemically, the tumoral cells expressed CD34 (Fig. 5).

The histological and immunohistochemical appearance has given rise to two diagnostic possibilities. Malignant phyllodes tumour and undifferentiated pleomorphic soft tissue sarcoma. The diagnostic decision of Malignant phyllodes tumour was made after

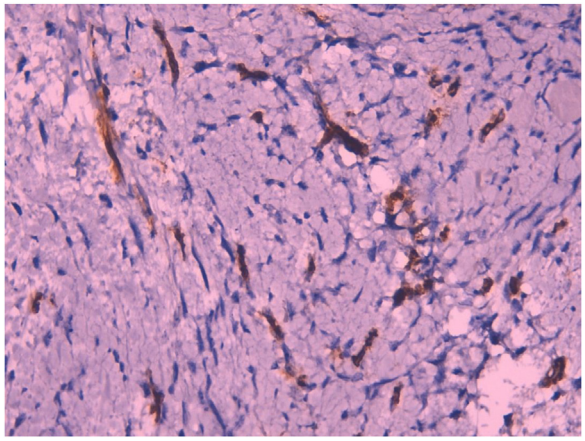

Fig. 5. Immunohistochemically, the tumoral cells expressed CD34.

taking into consideration the clinical and paraclinical data of the patient.

The post-operative period was marked by the appearance of a lymphocele, which was treated by puncture-aspiration on three occasions with good clinical evolution.

After 3 months of surgery, the patient was given a breast MRI and a PET/CT 18F-FDG scan, which returned without abnormalities.

3. Discussion

Malignant phyllodes tumour of the breast is rare, represent less than 1% of all breast cancers.

[1]. It differs from primitive breast sarcomas by the presence of both epithelial and connective elements.

The predominance of the conjunctive component is a criterion of malignancy which defines grade III; it is the malignant phyllodes tumour [3].

As with other malignant breast tumours, the incidence of malignant phyllodes tumours of the breast is much higher in women than in men.

The age of appearance of this tumour is between 45 and 55 years for Malard et al. [3], which is a young age compared to epithelial breast cancers.

The clinical distinction between a phylloid sarcoma and a fibroadenoma is difficult [4].

The tumour is found in 30 % of cases in the superolateral quadrant, its size can vary from a few centimetres to several tens of centimetres in long axis [4], it is multilobular and not fixed at the cutaneous or deep level [4].

Node involvement is rare in sarcomas, as it rarely involves metastasis to the nodes, but it can occur in 8% of cases [3].

The study by Yilmaz et al. [5], in which mammograms were performed on 31 patients, revealed a significant hyperdensity of phylloid tumours in relation to the surrounding mammary parenchyma. For Stebbing and Nash, mammography showed a typically benign opacity without microcalcifications in six out of eight cases of malignant phylloid tumours [1].

Ultrasonography gives low-echogenic images with regular contours and no posterior shadow cone [5]; for Stebbing and Nash, two malignant tumours were reported as benign by ultrasound among 13 patients who were ultrasonographed [1].

Cytology by fine needle cytopuncture doesn't allow the diagnosis to be made. Alam et al. [6] report a certain preoperative diagnosis by cytopuncture in only 17 % of cases.

The diagnosis of phylloid sarcoma is histological [5], by microbiopsy or macrobiopsy or on a lumpectomy sample as in our case or mastectomy.

Phylloid tumours of the breast can be classified into three categories: benign, borderline and malignant, depending on the histological characteristics of the tumour which include margins, cellularity of the stroma, disproportionate proliferation of stroma elements, tumour necrosis and mitotic index.

The most commonly used classification is that proposed by Azzopardi [7] (Table 1).

Immunohistochemistry is currently an indispensable tool for confirming histological type [6].

Dacic et al. [8] have shown that the positivity of Ki67 and stromal P53 was more often associated with high-grade breast phylloid tumours; therefore, Ki67 expression may help to distinguish between benign and malignant phylloid tumours in cases of difficult diagnosis.

Surgery is the gold standard in the treatment of malignant phyllodes tumours, be it a mastectomy or a large lumpectomy [9].

In an American study, published in 2006, which monitored 821 patients with non metastatique phylloid tumours over 21 years, they had either a mastectomy or a wide lumpectomy; the authors concluded that mastectomy does not provide better survival than wide lumpectomy [10].

As with other soft tissue sarcomas, the quality of the surgical removal margins is the main predictor of local recurrence.

The study of the margins of the surgical specimen is very important, especially in the case of conservative partial surgery [5].

Axillary curage does not bring any benefit in terms of survival; it can be performed if the adenopathies are clinically palpable [9].

Pandey et al. [11] studied the interest of postoperative radiotherapy on 12 patients with a high-grade phyllode tumour.

This seems to increase the five-year survival rate and decrease the local recurrence rate.

Currently, radiotherapy is indicated if the safety margin is less than 10 mm or if there is a local recurrence [9].

Several chemotherapy protocols have been used without any survival benefit.

Doxorubicin has been used successfully in a few studies in cases of inoperable local recurrences or in the presence of metastases [9].

Despite the presence of hormone receptors in phyllodes tumours, hormone therapy has not yet been studied for this type of pathology [4].

Local recurrence appears to be related to the degree of malignancy of the tumour [5] as well as the surgical margins of resection. A higher rate of distant metastases and local recurrence is found when the margins of resection are less than 10 mm [10].

After resection, the prognostic factors for malignant phyllodes tumours correspond to the histological grade, surgical margins and the presence of tumour necrosis [11].

Tumour size may also be a pejorative factor [11].

4. Conclusion

Malignant phyllodes tumour of the breast is rare, it is completely different from epithelial cancer and it can exist in man.

Its epidemiology is particular and it has particular difficulties in clinical and paraclinical diagnosis.

The treatment is based on surgery, which may be a large lumpectomy or mastectomy, lymph node removal is unnecessary, and the prognosis depends on several factors, the most important of which are margins of surgical resection.

Declaration of Competing Interest

No conflict of interest.

Funding

No sources of funding to our research has been needed.

Ethical approval

It's a one case report needing no ethnical approval.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request

Author contribution

Dr. Boudou Mohamed: Have written the article, have consulted the patient, prescribed all of the tests and prepared the patient for surgery and participated in the surgery

Dr Guellil Abdelali: have helped writing the article, data collection.

Dr Haloui Anass: Interpretation of histological data.

Pr Benani Amal(anatomopathology professor): confirm the histological diagnosis

Pr Jabi Rachid: supervised the writing of manuscript.

Pr Bouziane Mohamed (oncology surgery professor): have supervised the writing of the paper, and has been the leader surgeon of the case.

Registration of research studies

Not applicable.

Guarantor

Mohammed bouziane: bouzianemohammed@hotmail.com (oncology surgery professor).

Provenance and peer review

Not commissioned, externally peer-reviewed.

References

[1] J.F. Stebbing, A.G. Nash, Diagnosis and management of phyllodes tumor of the breast: experience of 33 cases at a specialist center, Ann. R. Coll. Surg. Engl. 77 (1995) 181–184.
[2] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, pour le groupe SCARE, The SCARE 2020 guideline: updating consensus surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.
[3] C. Confarvreux, A. Lurkin, N. Mitton, R. Blondet, C. Saba, D. Ranchèe, et al., Sarcomas and malignant phyllodes tumors of the breast: a retrospective study, Eur. J. Cancer 42 (2006) 2715–2721.
[4] S.G. Pollard, P.V. Marks, L.N. Temple, H.H. Thompson, Breast sarcoma. A clinicopathologic review of 25 cases, Cancer 66 (1990) 41–44.
[5] N. Taira, D. Takabatake, K. Aogi, S. Ohsumi, S. Takashima, R. Nshimura, et al., Phyllodes tumor of the breast: stromal overgrowth and histological classification are useful prognosis-predictive factors for local recurrence in patients with a positive surgical margin, Jpn. J. Clin. Oncol. 37 (10) (2007) 730–736.
[6] I. Alam, Z.T. Awad, H.F. Given, Cystosarcoma phyllodes of the breast: a clinicopathological study of 11 cases, Ir. Med. J. 96 (2003) 179–180.
[7] J.G. Azzopardi, Sarcoma in the breast, in: J. Benningron (Ed.), Problems in Breast Pathology, II, WB Saunders, Philadelphia, 1979, pp. 355–359.
[8] S. Dacic, S. Kounelis, E. Kouri, M.W. Jones, Immunohistochemical profile of cystosarcoma phyllodes of the breast: a study of 23 cases, Breast J. 8 (6) (2002) 376–381.
[9] D.K. Blanchard, C.A. Reynolds, C.S. Grant, J.H. Donohue, Primary non phyllodes breast sarcomas, Am. J. Surg. 186 (2003) 359–361.
[10] O.K. Macdonald, C.M. Lee, J.D. Tward, C.D. Chappel, D.K. Gaffney, Malignant phyllodes tumor of the female breast: association of primary therapy with cause-specific survival from the Surveillance, Epidemiology and End Results (SEER) program, Cancer 107 (9) (2006) 2127–2133.
[11] M. Pandey, A. Mathew, J. Kattoor, E.K. Abraham, B.S. Mathew, B. Rajan, et al., Malignant phyllodes tumor, Breast J. 7 (2001) 411–416.