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Case Report

Dermatofibrosarcoma protuberance of the skin of the breast: A case study and review of the literature

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ABSTRACT

Introduction and importance: DFSP is a soft tissue sarcoma that originates from dermal fibroblasts, that usually occurs in trunk or extremities and can invade the subcutaneous tissue. It rarely occurs in the breast with an incidence of 0.8–4.5% % per million population. It usually presents as a firm, well-defined, mobile, nontender mass. The diagnosis is confirmed by histopathology and immunohistochemistry. The recommended treatment is wide local excision. Adjuvant radiotherapy or imatinib should be considered in special cases. The recurrence-free survival and overall survival are good.
Case presentation: In this study, we present a 48-year-old woman with a growing tumor in her left breast.
Clinical discussion: The patient underwent breast-conserving surgery and reconstruction.
Conclusion: The diagnosis of DFSP was confirmed after a precise histological assessment.

1. Introduction

Dermatofibrosarcoma protuberans (DFSP) is a rare low-grade fibroblastic mesenchymal tumor that originates from the dermal fibroblasts and tends to be locally aggressive. It was first described in 1924 by Darier and Ferrand as a recurrent dermatofibroma and then in 1925 by Hoffmann as DFSP [1,2]. It represents about 6% of all soft tissue sarcomas [3] and can occur in all parts of the body, especially in the trunk and the limbs [4,5]. DFSP of the breast is rare and there are few cases in the literature about it. Its annual incidence is about 0.8–4.5% per million population, and its mostly seen presentation is an exophytic nodule in cutaneous tissue [6,7]. The recommended treatment for these patients is surgical excision with a 2–3 cm margin to reduce the recurrence rate. The reported cases' prognosis is good in the presence of clear margins [8]. The aim of this study is to present a case of DFSP of the breast, its clinical course, and its management. We also carried out a comprehensive search of the literature for cases of DFSP of the breast reported in the recent 20 years; the results are presented.

2. Presentation of case

The patient is a 48-year-old woman who presented with a skin lesion of the right breast that had existed for a year and had had slow growth during that period. On examination, she had a red nodular lesion with a definite border measuring approximately 2 × 4 cm in the lower inner quarter of the right breast. She did not mention any personal or family history of breast cancer (Fig. 1) but had a past history of papillary thyroid carcinoma and had undergone total thyroidectomy and iodine therapy.

On mammography, a dense lesion with definite borders was identified in the subcutaneous area. There was no intramammary lesion or microcalcification on mammography (Fig. 2). In order to confirm the diagnosis, a core needle biopsy was done, and the pathology report indicated DFSP. The patient underwent excision of the mass with 3 cm margins, followed by oncoplastic reconstruction. Macroscopically, there was a cream pink dermal mass of 4 × 2 × 2 cm with subcutaneous thickening. Neoplastic cells were seen in mammary ducts/lobules and mammary fat having honeycomb patterns. Histological analysis showed a hypercellular mass having an infiltrative growth pattern, centered in the breast parenchyma and a uniform spindle cell proliferation with fibro collagenous stroma. Neoplastic cells showed wavy nuclei. No sign of necrosis and atypical mitoses was seen. In immunohistochemistry, the cells stained positive for CD34 and S100, whereas they were SMA and Desmin negative; Ki 67 was 5%. Based on histological findings and IHC,

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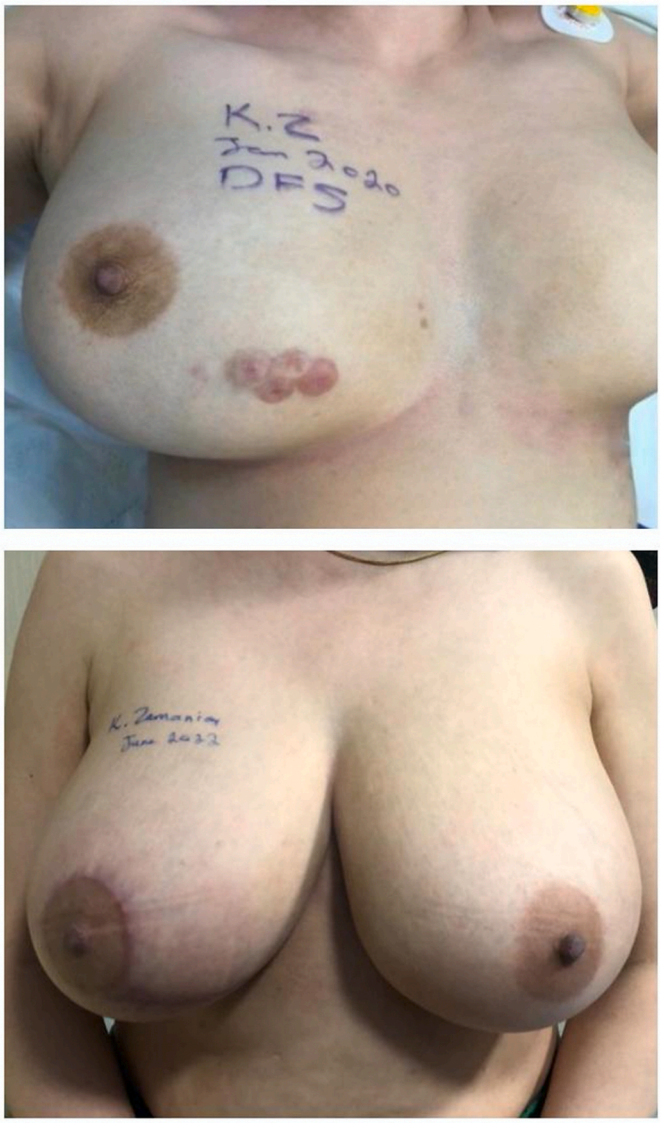

Fig. 1. breast lesion before surgery and the cosmetic result after surgery.

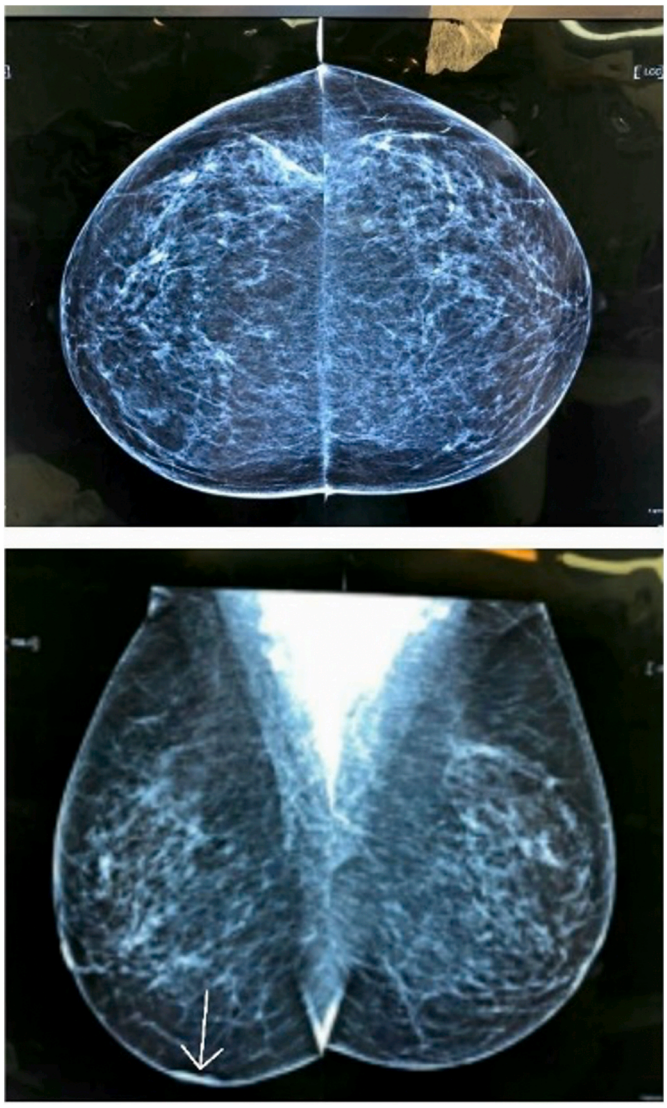

Fig. 2. Mammography CC and MLO view.

the final diagnosis was DFSP and the margins were free of disease. In the follow up after one year, there was no recurrence and the patient was very satisfied with the cosmetic result.

We performed a comprehensive literature search for the DFSP of breasts from 2000 to 2022, and the result returned 28 cases. The location, clinical examination, and imaging findings of all these cases are summarized in Table 1 in order of publication year.

The methods were stated in accordance with the SCARE 2020 guidelines [9].

3. Discussion

DFPS may occur throughout life from the age of 2–75 years old, and the mean age of the disease is around 40 years old [37]. Clinical presentation of DFSP in the breast is similar to its presentation in other parts of the body. It appears as a firm, red or brown-red, well-defined nodule which becomes irregular with further growth. There is no well-known risk factor for DFSP except for some cases reported to arise within a surgical scar, or in the field of irradiation [38]. one study reported a history of estrogen replacement therapy, and another study reported a case of DFSP in the breast during pregnancy [39]. There was a history of papillary thyroid carcinoma in our patient, she had undergone total thyroidectomy and radioactive iodine therapy in the previous year; however, there is no report of the association of thyroid cancers or radioactive iodine treatment with DFSP in the breast or in any other area in the literature.

DFSP of the breast usually appears as a well-defined superficially located benign tumor in ultrasonography and mammography but in some cases, it is ill-defined or has lobulated borders. It may have variable internal echo patterns, and acoustic enhancement may be seen [40]. In our patient, the mammography showed a dense lesion with definite borders in the subcutaneous area.

Core needle biopsy, punch, and excisional biopsy may all be diagnostic but fine needle aspirations are not useful because diagnosis should be confirmed by histopathology and IHC. In histological examination the amount of mitosis is variable and spindle cells are seen as a spiral pattern in the dermal or subdermal layers. This tumor includes various histologic morphologies, including sarcomatous transformation [40].

Immunohistochemical studies have reported positive reactions for CD34 in all cases, and vimentin in a few ones. Also, staining for S-100, actin, SMA, desmin, and CD31 is usually negative. Cytokeratin, factor XIII A, EMA, HMB45, ER, PR, brst-2, CD31, CD68, CD99, c-kit, and BCL-2 have been investigated in some studies and were reported as negative [41].

Table 1
Clinical examination, location, and imaging findings of breast dermatofibrosarcoma protuberance.

	First author	Year published	Age (year)	Gender	Size (cm)	Side, Location	Physical examination	Mammography	Ultrasonography
1	Romero [10]	2002	56	female	4.8	Upper and lower inner quadrant of right breast	A non-ulcerated,well-circumscribed, multilobe, nodular lump	A dense, well-circumscribed, nodular lesion	Two adjacent heterogeneous, nodular masses
2	Park [11]	2011	28	female	2	Lower inner quadrant of left breast	A smooth mobile keloid-like reddish mass surrounded by brownish plaque-like cutaneous thickening		
3	Rani [12]	2013	14	female	1.5	around areola of Left breast l	A Firm and nontender mass		
4	Chan [13]	4014	41	female			An exophytic, lobulated tumor, brownish in the periphery and lighter in the center		
5	Kinney [14]	2016	26	female	3	Lower inner quadrant of left breast	A keloid-like, purple-hued, protuberant, and irregular mass		
6	Hoseinpour [15]	2016	18	female	3	Axillary tail of right breast	A well-defined mildly tender firm mass in the dermis		A cyst with thickened wall, located near the axillary tail, which continued to the dermis
7	POHLODEK [16]	2017	43	female	6	Upper and lower inner quadrants of left breast	A superficial, skin-infiltrating, prominent lump	A circumscribed round and partially lobulated radiopaque lesion with sharp contours	
8	Zhao [17]	2017	29	female	3	upper inner quadrant of left breast	A mobile, hard and irregular margins lump in subcutaneous	A high density oval mass with well-defined lobulated margins	A mixed echogenic mass, with partially defined margin and abundant blood flow signal at the margins
9	Hernández [18]	2017	56	female	4.8	Upper and lower inner quadrants of right breast	A non-ulcerated, well-circumscribed, multilobe, nodular lump	A dense, well-circumscribed, nodular lesion	Two adjacent heterogeneous, nodular masses
10	Burud [19]	2017	47	female	2	Upper outer quadrant of right breast	An oval-shaped, well circumscribed, , mobile, firm and fixed lump	A fairly well-defined lesion	An ill-defined heterogeneous lesion pre-dominantly hyper echoic with increased vascular flow
11	Noh [20]	2017	43	female	1.4	upper inner quadrant of left breast	A firm, non-tender mass with overlying erythematous skin changes	no abnormal lesions	An oval shaped low echoic mass in the dermal layer, with surrounding increased subcutaneous fat echogenicity
12	Dhakal [21]	2018	48	male	13	left breast	A nodular,non-tender and mobile mass		
13	Küçük [22]	2018	43	female	10	upper outer quadrant of right breast	A mobile, firm and lobulated margins mass	A high opacity nodules with smooth margin	A solid nodule with smooth margins and hypoechoic quality
14	Sookar [23]	2018	33	female	1.6	Lower inner quadrant of left breast	An irregularly demarcated, firm and tan-brown mass		An area of skin thickening
15	Bouhani [24]	2019	44	male	4	Upper quadrant of left breast	A firm, tender, mobile mass that invaded the skin	An indistinct limited mass with central vascularization	A nonhomogeneous and hyperdense lesion
16	Vecchio [25]	2019	41	female	5	between upper quadrants of right breast	A solid, multinodular lesion, with irregular margins	A well-circumscribed, round to oval homogeneous mass, with multilobulated borders	A well-circumscribed, round to oval homogeneous mass, with multilobulated borders
17	Farsi [26]	2020	35	female	3	Upper inner quadrant of left breast	A nodular,pinkish, mobile and non-tender skin lesion	A lobulated mass	A subcutaneous lobulated mass showed increased vascularity
18	Ezejiofor [27]	2020	13	male		upper outer quadrant extending to the inner upper quadrant	A nodule and areas of hyper- and hypo-pigmentation of the overlying skin		
19	Ng [28]	2021	30	female	3	Upper outer quadrant of left breast	A palpable, mobile and non-tender mass		A hypoechoic focal lesion
20	Emegoakor [29]	2021	28	female	9	upper outer quadrant of left breast	A firm, well defined nodular skin lesions		
21	DIMAS [30]	2021	52	female	4	upper outer quadrant of right breast	A light-reddish, delineated exophytic, nodular skin mass	A dense, broad-based, well circumscribed, cutaneous lesion	
22		2021	37	female	5				

(*continued on next page*)

Table 1 (*continued*)

	First author	Year published	Age (year)	Gender	Size (cm)	Side, Location	Physical examination	Mammography	Ultrasonography
	Batista Aquino [31]					lower inner quadrant of right breast	An exophytic and ulcerated lesion		An exophytic and vascularized nodule
23	Ćirilović [32]	2021	66	male	4.5	Upper inner quadrant	A palpable nodule beneath the skin	A hyperdense mass without calcifications	A well-defined hypoechoic lesion with sharp and smooth edges
24	Kim [8]	2021	45	female	4	upper inner and upper outer quadrants of right breast	A movable, hard, subcutaneous lump		An oval-shaped hypoechoic mass with hyperechoic rim in the subcutaneous layer and increased vascularity with several microcalcifications
25	Arslan [33]	2021	22	female	7.5	lower inner quadrant of right breast	A new breast mass		An encapsulated, hypoechoic well- circumscribed, oval-shaped mass
26	Saikia [34]	2022	40	male	6.5	central part of left breast	A firm, mobile, nodular, non-tender, ulcerated swelling		
27	Rezaee [35]	2022	42	female	4		A new breast mass	An oval-shaped lesion with halo	An echo-complex, non-homogenous mass
28	Prabhu [36]	2022	55	male	5	Upper outer and upper inner quadrants of left breast	A nodular swelling and smaller satellite nodules, red and shiny skin over the nodules		

Routine initial staging is not recommended unless metastasis is suspected, which is a rare event that is usually hematogenous and mostly involves the lungs; occurring after several recurrences [42].

Wide local excision with a 2–3 cm margin is the recommended treatment for DFSP. This is feasible in the breast by using oncoplastic techniques, as done in our patient. For large bulky tumors, mastectomy is the best option [43,44]. When the lesion is present in areas such as the face, neck, and scalp where removal of the lesion with a wide margin is not possible due to cosmetic results, the Mohs microsurgery (MMS) method can be used. The advantage is that the margins are identified during the operation and more healthy tissue is maintained. This method might also be appropriate for breast lesions [45]. There is only one case of Mohs surgery reported for cutaneous metastasis of breast cancer [46]. Case reports have also successfully performed nipple sparing mastectomy followed by immediate reconstruction [47].

The recurrence rate after excision is estimated to be 20–50%, this high rate may be the result of the infiltrating growth pattern of the tumor. Most recurrences seem to occur within the first 3 years after resection but there is a case report of recurrence after 26 years [48].

Adjuvant radiation therapy is not recommended in DFPS of the breast except for those with undesirable prognostic factors such as high mitotic counts and positive margins, or fibrosarcomatous features [49,50]. In a meta-analysis of 167 DFSP patients undergoing adjuvant radiotherapy, Chen et al. stated that adjuvant radiotherapy could be performed for all patients undergoing surgical excision without considering the surgical margin status, although data of long term follow up of cases of DFSP with negative margins is not reported [51]. There is no study on the result of adjuvant radiotherapy in DFSP of the breast. In our patient, radiotherapy and oncology consultation did not recommend any adjuvant treatment because of wide negative margins in the resected specimen.

Imatinib mesylate, an inhibitor of the platelet-derived growth factor receptor–associated tyrosine kinase, has been demonstrated to be effective in the treatment of some advanced cases of DFSP, and might be useful for tumors originating in the breast; especially if translocation (7; 22) (q22; q13) is detected [52]. A systematic review of 152 cases of DFSP of different body parts, with advanced local or metastatic disease treated with imatinib, showed that this medicine was an effective treatment, and a complete response was seen in 2.5% of patients and a partial response in 55.2% [53,54].

Recurrence-free survival that has been reported for all DFSP cases is as long as 15 years [8].

4. Conclusion

DFSP of the breast is a rare occurrence and there are only some case reports on this subject in the literature. It seems that its clinical presentation and treatment are identical to DFSP in other parts of the body. There is limited data about adjuvant therapy for breast DFSP but radiotherapy and imatinib are recommended in selected cases.

Ethical approval

All procedures performed in this study involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

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Author contribution

Dr. Ramesh Omranipour: Conceptualized and designed the study, drafted the initial manuscript, and reviewed and revised the manuscript.

Dr. Sadaf Alipour: Designed the data collection instruments, collected data, carried out the initial analyses, and reviewed and revised the manuscript.

Dr. Leila Haji Maghsoudi: Coordinated and supervised data collection, and critically reviewed the manuscript for important intellectual content.

Registration of research studies

1. Name of the registry: Tehran University of Medical Sciences. Unique Identifying number or registration ID:

2. Hyperlink to the registration (must be publicly accessible).

Guarantor

Leila Haji Maghsoudi.

Consent

Not applicable.

Consent to participate

from the under 16 years old was given by a parent or legal guardian.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Availability of data and material

Data sharing is not applicable to this article as no datasets were generated or analyzed during the current study.

Consent for publication

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.

Research registration

N/A.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Declaration of competing interest

The authors deny any conflict of interest in any terms or by any means during the study.

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