



Multisite recurrence of dermatofibrosarcoma protuberans with high-grade fibrosarcomatous transformation: a case report

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Abstract

Background. Dermatofibrosarcoma protuberans (DFSP) is a rare, locally aggressive soft tissue sarcoma with a high recurrence rate. The fibrosarcomatous transformation of DFSP (FS-DFSP) is associated with increased metastatic potential and poorer prognosis. Management typically involves wide surgical excision with clear margins, but recurrent and metastatic cases require a multidisciplinary approach, including radiation therapy and systemic treatment.

Case presentation. We report a 37-year-old female with a history of recurrent DFSP involving the left posterior chest wall. She underwent multiple surgical resections in Egypt (2017, 2019, 2022), with a subsequent recurrence requiring wide local excision and adjuvant radiation therapy (50 Gy in 25 fractions) in 2022. In October 2023, evaluation revealed a recurrent 10×8 cm mass infiltrating the posterior chest wall. MRI confirmed the involvement of the underlying ribs and intercostal muscles. She underwent extensive resection with plastic reconstruction in November 2023, with pathology confirming Grade 3 FS-DFSP (14×13×6 cm) with negative margins. Postoperatively, wound healing progressed to 80%, and adjuvant AIM chemotherapy was planned. However, in November 2024, imaging detected a new 8.8×9×8 cm mass in the pancreatic head involving the superior mesenteric vein. Biopsy confirmed DFSP recurrence. The patient underwent ERCP with stent placement and was re-evaluated for further surgical and oncological management.

Conclusion. This case highlights the aggressive nature of FS-DFSP with multisite recurrence, necessitating ongoing surveillance and a multidisciplinary treatment approach. Despite repeated resections and adjuvant therapies, disease progression underscores the need for novel therapeutic strategies to improve long-term outcomes in high-grade DFSP.

Keywords: Dermatofibrosarcoma protuberans, fibrosarcomatous transformation, recurrent sarcoma, chest wall tumor, pancreatic metastasis.

For citation: Rawia Mubarak Mohamed, Ashraf ALakkad, Babitha Alingal Mohammed, Hazem Muhammad ALmasarei. Multisite recurrence of dermatofibrosarcoma protuberans with high-grade fibrosarcomatous transformation: a case report. *Clinical review for general practice*. 2025; 6 (12): 37–43 (In Russ.). DOI: 10.47407/kr2025.6.12.00728

Многоочаговый рецидив взбухающей дерматофибросаркомы с трансформацией в высокодифференцированную фибросаркому: клинический случай

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Аннотация

Введение. Взбухающая дерматофибросаркома – это редкая локально агрессивная саркома мягких тканей с высокой частотой рецидивов. Трансформация в фибросаркому при взбухающей дерматофибросаркоме связана с повышением метастатического потенциала и худшим прогнозом. Лечение обычно предполагает широкое иссечение с отступом от краев, однако в случаях рецидивов и метастазирования необходимо использовать мультидисциплинарный подход, в том числе лучевую терапию и системное лечение.

Клинический случай. Представлено описание случая 37-летней пациентки с рецидивом взбухающей дерматофибросаркомы в задней грудной стенке слева. Она перенесла несколько операций по иссечению опухоли в Египте (в 2017 г., 2019 г. и 2022 г.), после которых имел место рецидив, в связи с чем в 2022 г. потребовались широкое иссечение опухоли и адъювантная лучевая терапия (50 Гр, 25 фракций). В октябре 2023 г. обследование выявило рецидив – образование размером 10×8 см, проникающее в заднюю грудную стенку. Магнитно-резонансная томография подтвердила поражение нижележащих ребер и межреберных мышц. В ноябре 2023 г. пациентке выполнили обширную резекцию и реконструктивно-пластическую операцию, патологоанатомическое исследование подтвердило трансформацию в фибросаркому при взбухающей дерматофибросаркоме 3-й степени злокачественности (14×13×6 см) с негативными краями резекции. В послеоперационном периоде после заживления раны на 80% была запланирована адъювантная АИМ-химиотерапия. Однако в ноябре 2024 г. томография выявила новое образование размером 8,8×9×8 см в головке поджелудочной железы, распространяющееся на верхнюю брыжеечную вену. Биопсия подтвердила рецидив взбухающей дерматофибросаркомы. Пациентке выполнили эндоскопическую ретроградную холангиопанкреатографию и стентирование, после чего ее обследовали на предмет дальнейшего хирургического вмешательства и лечения рака.

Заключение. Представленный случай демонстрирует агрессивный характер трансформации в фибросаркому при многоочаговом рецидиве взбухающей дерматофибросаркомы, обуславливающий необходимость наблюдения и применения мультидисциплинарного

подхода к лечению. Прогрессирование заболевания, несмотря на повторные иссечения и курсы адъювантной терапии, подчеркивает необходимость применения новых терапевтических стратегий для улучшения долгосрочных результатов при выходящей дерматофибросаркоме высокой степени злокачественности.

Ключевые слова: выходящая дерматофибросаркома, трансформация в фибросаркому, рецидив саркомы, опухоль грудной стенки, метастазы в поджелудочной железе.

Для цитирования: Равия Мубарак Мохамед, Ашраф Альаккад, Бабита Алингал Мохаммед, Хазем Мухаммад Альмасареи. Многоочаговый рецидив выходящей дерматофибросаркомы с трансформацией в высокодифференцированную фибросаркому: клинический случай. *Clinical review for general practice*. 2025; 6 (12): 37–43 (In Russ.). DOI: 10.47407/kr2025.6.12.00728

Introduction

Dermatofibrosarcoma protuberans is a type of skin cancer that tends to be locally aggressive [1]. It recurs in the same area but doesn't usually spread to other parts of the body [2]. Taylor first recognized this tumor in 1890, and then Darrier described it in 1924, but Hoffman coined the term "dermatofibrosarcoma protuberans" in 1925 [3, 4]. Reports indicate that it affects various body surfaces, primarily the trunk (42–72%), then the extremities (16–30%), and less frequently the head and neck (10–16%) [2]. Even though it makes up less than 0.1% of all malignant neoplasms, it is actually the most common skin sarcoma, accounting for nearly 1% of all soft tissue sarcomas, over 1% of all malignant tumors in the head and neck, and 7% of all head and neck sarcomas [5].

Patients with the fibrosarcomatous variant have higher recurrence rates, and the severity of cancer tends to increase with multiple recurrences [6]. This lesion is characterized by a storiform growth pattern of spindle cells found in the deep dermis [6]. The COL1A1-PDGFB fusion gene appears in about 85–90% of the cases [7]. The epidermis above DFSP appears thinner and might be separated from the tumor by a Grenz zone, based on histological examination [8]. Dermatofibrosarcoma protuberans begins as a nodule or an unclear dermal plaque that spreads into the subcutis or occasionally into skeletal muscle, showing a distinctive "honeycomb" pattern [5]. The epidermis typically isn't affected [5]. The immunohistochemical analysis shows CD34 positivity, which indicates the development of dendritic cells (fig. 1) [5]. In this case, the pancreatic lesion represents an exceptionally rare metastatic site for DFSP, likely via hematogenous spread. This highlights the need for systemic evaluation even in tumors historically known for local aggressiveness. Myoid nodules are observed more often in the fibrosarcomatous variant of DFSP [9].

About 85–90% of all DFSPs are considered low-grade tumors [10]. A small portion, about 10–15%, includes a high-grade fibrosarcoma component [10]. This transformation, which appears in over 5% of tumor volume, is marked by a greater likelihood of local relapse and distant metastasis [11]. Complete surgical resection is considered the best treatment for local DFSP [12]. However, the minimum resection margin required for local control is still not clearly defined. It's really challenging to achieve local control of the tumor [13]. For instance, after regular surgery, DFSP comes back in about 20% of cases, but with Mohs surgery, the recurrence rates drop to less than 1% [14].

Additionally, the unique villous pattern that extends into the subcutaneous fat, fascia, and muscles, while still keeping healthy tissue intact during resection, poses a significant surgical challenge (fig. 2) [15]. If complete excision isn't achieved, it can result in local recurrence [15]. This case

report presents a case of recurrent dermatofibrosarcoma protuberans with high-grade fibrosarcomatous transformation.

Case presentation

A 37-year-old female patient with a history of sarcoma involving the left posterior chest wall presented for ongoing management of recurrent disease. She underwent surgical resection of the sarcoma in Egypt in 2017, 2019, and 2022. Despite multiple resections, the patient experienced tumor recurrence. Plans were made for wide local excision of the recurrent tumor, including chest wall resection involving the ribs, with subsequent plastic reconstruction.

The patient had a history of multiple local recurrences of dermatofibrosarcoma protuberans (DFSP) since 2014. In 2019, PET/CT revealed metastatic lesions in the lungs, for which she underwent metastatectomies. In 2022, a local recurrence at the previous left posterior chest wall surgery site led to a wide local excision, followed by adjuvant radiation therapy (50 Gy in 25 fractions), completed on August 1, 2022 (fig. 4).

Later on October 13, 2023, the patient was evaluated in the plastic surgery clinic, following a referral from the thoracic surgery department for further assessment and surgical planning. The multidisciplinary team scheduled the excision and reconstruction surgery for November 23, 2023. On examination, a recurrent sarcoma measuring approximately 10×8 cm was noted on the left posterior trunk. MRI of the chest with contrast revealed a large, heterogeneously enhancing lobulated mass within the subcutaneous tissue of the left posteroinferior chest wall, measuring approximately 12×7×10 cm (fig 5). Abnormal signal intensity with some enhancement was observed in the posterior aspects of the underlying ribs (7–9) and adjacent intercostal muscles, but there was no evidence of spinal extension.

The most recent resection on November 23, 2023, confirmed a unifocal tumor measuring 14×13×6 cm, identified as Grade 3 DFSP with high-grade fibrosarcomatous transformation. Pathology findings included a mitotic rate of 20 mitoses per 10 high-power fields, 30–40% necrosis, negative margins, and no evidence of lymphovascular invasion (pT3). Histological analysis conducted on 23 November 2023 showed an infiltrative spindle cell proliferation with storiform architecture and CD34 positivity with adjacent areas of fascicular and herringbone growth pattern and extensive necrosis (fig. 3).

On March 8, 2024, the patient underwent a split-thickness skin graft procedure. By August 27, 2024, plastic surgery was completed without major complications. The patient was doing well and underwent daily wound dressings. Upon review with the Doctor, it was noted that wound healing was at 80%, and further assessment was planned

for the following day to determine the timing of chemotherapy initiation. The oncology team discussed the AIM chemotherapy regimen (doxorubicin, ifosfamide, mesna) with the patient, explaining its rationale due to the high-grade transformation and systemic spread. The preliminary plan was to commence chemotherapy on April 15, 2024, pending wound healing progress. A MUGA scan and port placement were ordered. Additionally, the patient was referred to gynecology for fertility preservation counseling before initiating chemotherapy due to the potential risk of infertility, as she is gravida 0. She was scheduled for a follow-up on April 15, 2024, to reassess wound healing and finalize chemotherapy initiation.

The patient was evaluated on multiple occasions, with vital signs recorded on March 26 and March 27, 2024. On March 27 at 11:15 AM UAE time, the patient's temperature was 36.7 °C (tympanic), with a peripheral pulse rate of 88 beats per minute and a respiratory rate of 18 breaths per minute. Blood pressure measured via machine was 129/78 mmHg, with a mean arterial pressure of 95 mmHg. Oxygen saturation was recorded at 99%. The previous day, at 14:29 PM, the patient's temperature was slightly higher at 36.8 °C, with a pulse rate of 98 beats per minute and an identical respiratory rate of 18 breaths per minute. Blood pressure at that time was slightly elevated at 132/86 mmHg, with a mean arterial pressure of 101 mmHg. Oxygen saturation was 98%.

Anthropometric measurements on March 27 indicated that the patient had a height of 165 cm and a weight of 111.5 kg, resulting in a body mass index (BMI) of 40.96 kg/m², classifying them as having severe obesity. The body surface area (BSA) was calculated at 2.26 m², with an ideal body weight of 56.91 kg and an adjusted body weight of 78.75 kg. These measurements highlight a significant discrepancy between actual and ideal body weight, emphasizing the need for weight management interventions. The patient's obesity was considered in planning for chemotherapy dosing and potential wound healing delays.

During the physical examination, the patient was alert and oriented, with no acute distress. The eyes showed equal, round, and reactive pupils to light, with intact extraocular movements and normal conjunctiva. Head, ear, nose, and throat (HENT) examination revealed a normocephalic appearance, moist oral mucosa, and no signs of pharyngeal erythema. The neck was supple, and non-tender, with no carotid bruits, jugular venous distention, or lymphadenopathy.

Respiratory examination demonstrated clear lungs to auscultation, non-labored respirations, equal breath sounds, and symmetrical chest wall expansion. Cardiovascular assessment showed a normal heart rate and regular rhythm, with no murmurs, gallops, or edema. The gastrointestinal examination revealed a soft, non-tender, and non-distended abdomen with normal bowel sounds and no organomegaly. The genitourinary examination was unremarkable, with no costovertebral angle tenderness.

Neurologically, the patient was alert and oriented, with normal sensory and motor function and no focal deficits. Cognitive and speech assessment indicated clear and coherent speech, with functional cognition intact. The psychiatric

Fig. 1. Diffuse CD34 positivity noted in the neoplastic cells.

Рис. 1. Диффузно расположенные CD34-положительные опухолевые клетки.

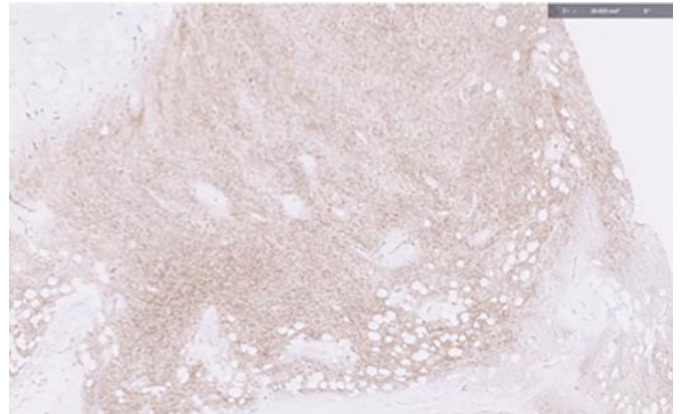


Fig. 2. At low power, lesion appears as a poorly-circumscribed, infiltrative spindle cell neoplasm within the dermis, often extending into the subcutaneous fat.

Рис. 2. При низкой мощности образование выглядит как веретенчатая опухоль с нечеткими краями и инфильтративным ростом в дерме, во многих местах распространяющаяся в подкожно-жировую клетчатку.

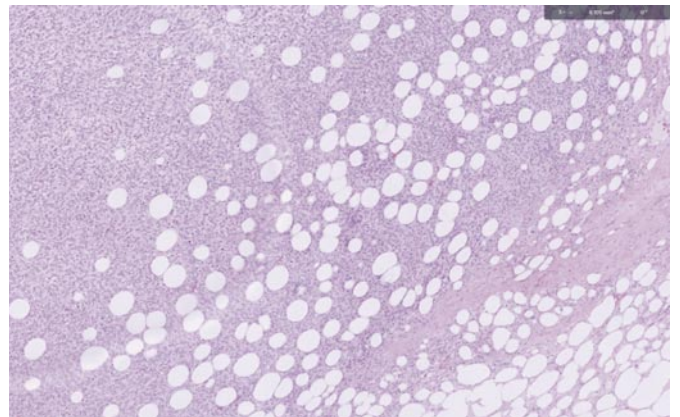
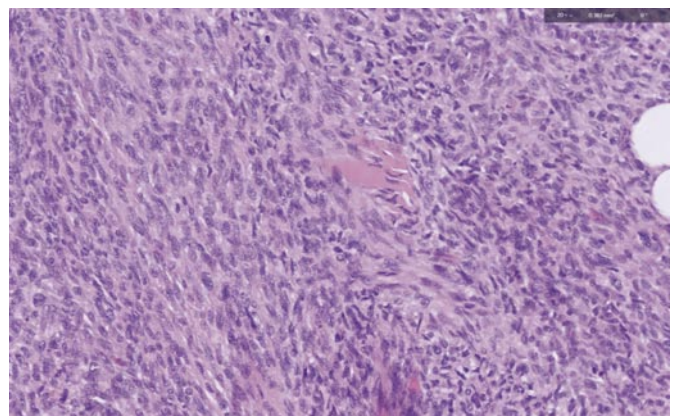


Fig. 3. Focal fascicular architecture with loss of characteristic storiform pattern. Increased cellularity with increased mitotic activity.

Рис. 3. Очаговая пучковая архитектура с утратой характерного «муарового» рисунка. Повышенная клеточность и высокая митотическая активность.



evaluation showed a cooperative patient with an appropriate mood and affect, as well as normal judgment.

The patient had a known history of G3 dermatofibrosarcoma protuberans with high-grade fibrosarcomatous trans-

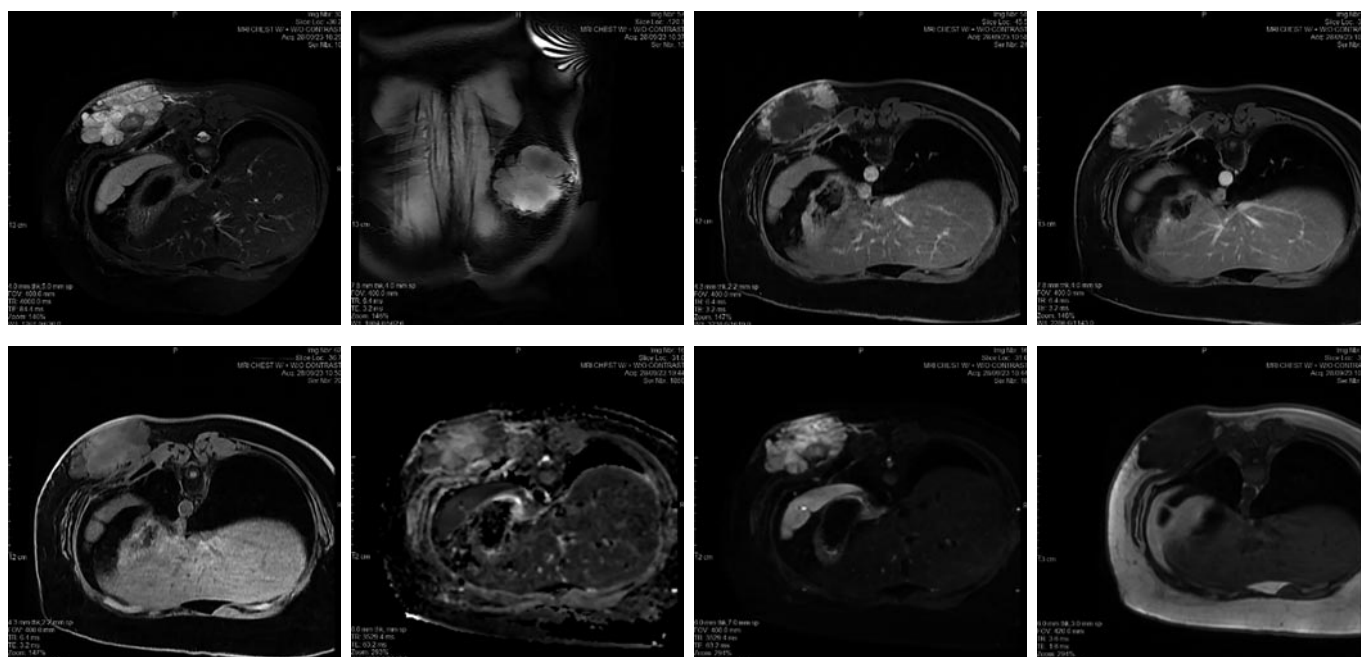
Fig. 4. Status post previous left chest wall surgery. CT of the chest wall, performed on 28/09/2023, reveals a large soft tissue mass arising from the left posterior-inferior chest wall, measuring 5 cm (anteroposterior) ×11 cm (transverse) ×10 cm (craniocaudal).

Рис. 4. Состояние после операции на грудной стенке слева. Компьютерная томография грудной стенки, выполненная 28.09.2023, выявила крупную опухоль мягких тканей в нижней части задней грудной стенки слева размером 5 см (переднезадний размер) ×11 см (поперечный размер) ×10 см (краниокаудальный размер).



Fig. 5. MRI performed on 28/09/2023 reveals a large, heterogeneously enhancing lobulated mass with a non-enhancing central portion, involving the subcutaneous tissue of the left posterior inferior chest wall and measuring approximately 12×7×10 cm.

Рис. 5. Магнитно-резонансная томография, выполненная 28.09.2023, выявила крупное образование с дольчатым строением и неравнорным накоплением контрастного вещества, с не накапливающим контрастное вещество участком в центре, распространяющееся в подкожно-жировую клетчатку нижней части задней грудной стенки слева, размером около 12×7×10 см.



formation, status post resection (fig 6). Wound healing was noted to be at 80%, and further evaluation with Doctors was planned for the following day to determine the timeline for initiating chemotherapy. The patient was counseled regarding the AIM regimen in the adjuvant setting, consisting of six cycles, and consent was obtained for treatment initiation on April 15, 2024, pending wound healing status.

To facilitate chemotherapy, a Multi-Gated Acquisition (MUGA) scan and PORT placement have been ordered. Additionally, the patient was referred to gynecology to discuss fertility preservation options prior to chemotherapy, given the associated risk of infertility and her Go status.

The patient was scheduled for chemotherapy starting April 15, 2024, with a five-day regimen every three weeks

for six cycles. Anticipated side effects included immune suppression, fatigue, nausea, vomiting, and the potential need for medical leave during treatment. The patient was reassessed at the next visit to confirm wound healing progress before proceeding with chemotherapy.

Later, a CT scan of the chest, abdomen, and pelvis performed on 20/11/24 revealed a normal abdominal wall and liver with no detectable focal lesions (fig. 7–9). The gallbladder had been surgically removed, and there was mild dilation of the intrahepatic biliary radicles. A new interval finding of a heterogeneous mass measuring 8.8×9×8 cm was noted in the liver hilum, inseparable from the pancreatic uncinate process, involving the superior mesenteric vein and compressing the descending part of the duode-

Fig. 6. Status post resection of a left posterior chest wall tumor and chest wall reconstruction. CT performed on 08/02/2024 demonstrates post-operative changes in the left posterior chest wall and ribs, with no evidence of recurrent disease.

Рис. 6. Состояние после иссечения опухоли нижней части задней грудной стенки слева и реконструктивной операции на грудной стенке. Компьютерная томография, выполненная 08.02.2024, демонстрирует послеоперационные изменения задней грудной стенки и ребер слева, а также отсутствие признаков рецидива заболевания.



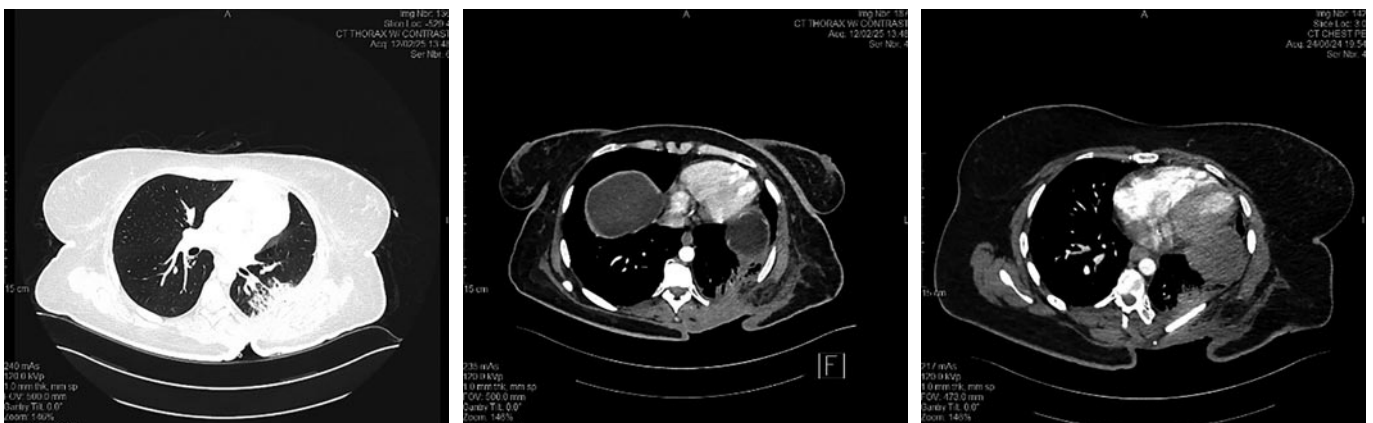
Fig. 7. Abdominal CT performed on 28/11/2024 reveals a newly detected heterogeneous mass measuring 8.8×9×8 cm in the liver hilum, inseparable from the pancreatic uncinate process. The mass involves the superior mesenteric vein and compresses the descending part of the duodenum.

Рис. 7. Компьютерная томография брюшной полости, выполненная 28.11.2024, выявила новое неоднородное образование размером 8,8×9×8 см в воротах печени, которое составляет единое целое с крючковидным отростком поджелудочной железы. Образование распространяется на верхнюю брыжеечную вену и вызывает компрессию нисходящей части двенадцатиперстной кишки.



Fig. 8. Chest CT performed 12/2/2025 following the resection of left posterior chest wall tumour with chest wall reconstruction, open wound with partially exposed lower lobe, no significant changes of noted Post-operative pulmonary and soft tissue scarring with soft tissue thickening.

Рис. 8. Компьютерная томография грудной клетки, выполненная 12.02.2025 после иссечения опухоли задней грудной стенки слева и реконструктивной операции на грудной стенке: открытая рана с частично открытой нижней долей. Не отмечено существенных изменений послеоперационного рубцевания легких и мягких тканей с уплотнением мягких тканей.



num. There was mild prominence of the pancreatic duct, while the pancreas itself appeared normal in size and density. The spleen, adrenal glands, and kidneys were normal, with no focal lesions or signs of perinephric fluid accumula-

tion. The urinary bladder was underfilled, with an inflated Foley's catheter balloon. The uterus was anteverted with a tiny subserosal myoma at its fundus, while the right and left ovaries measured 49×30 mm and 30×20 mm, respectively.

Fig. 9. Abdominal CT performed 12/2/2025 with no significant changes of pancreatic head/uncinate process mass lesion with related changes with CBD stent.

Рис. 9. Компьютерная томография брюшной полости, выполненная 12.02.2025: отсутствуют существенные изменения образования в головке/крючковидном отростке поджелудочной железы, изменения, связанные с билиарным стентированием.



Timeline of clinical events	
Схема истории болезни	
Date	Event
2014	First diagnosis of DFSP, left posterior chest wall
2017	Surgical resection in Egypt
2019	Local recurrence; resection and lung metastatectomy
2022	Recurrent tumor; wide local excision + radiation (50 Gy)
Oct 2023	Recurrence confirmed by MRI
Nov 2023	Wide local excision, chest wall resection, reconstruction
Mar 2024	Skin graft procedure
Apr 2024	Scheduled chemotherapy (AIM) and fertility preservation referral
Nov 2024	New pancreatic mass identified
Dec 2024	Biopsy confirmed DFSP recurrence
Feb 2025 No recurrence at the primary surgical site. The pancreatic metastasis was stable.	

Minimal free pelvic fluid was noted on the left side. The gastrointestinal tract showed no abnormal wall thickening, and no abnormally sized or appearing lymph nodes were detected. The abdominal vessels were normal, without stenotic plaques, and the mesentery and peritoneum appeared unremarkable. No signs of bone destruction were observed.

A new interval finding of a pancreatic head mass was identified, involving the superior mesenteric vein and compressing the descending duodenum. There was no definitive sign of local recurrence of the previously resected left posterior chest wall tumor. An ERCP was performed on 29/11/24 with the placement of a plastic stent. A biopsy review on 11/12/24 confirmed that the pancreatic head mass exhibited features favoring dermatofibrosarcoma. She was scheduled for evaluation by the plastic surgery team to assess a wound defect on the upper back. After the plastic surgery, the patient recovered completely.

Discussion

In the present case, a 37-year-old female was found to have recurrent dermatofibrosarcoma protuberans (DFSP) at the left posterior chest wall area accompanied by several local recurrences and the metastatic spread to the lungs and

pancreas. Simultaneous to these resections between 2014 and 2023, the patient underwent wide local excision, adjuvant radiation therapy, and intended chemotherapy, but the disease recurred. The most recent findings include a pancreatic mass confirmed as DFSP with an aggressive disease course and fibrosarcomatous transformation.

DFSP is a skin tumor that comes from the dermis and is known for its slow growth that spreads into surrounding tissues [16]. It often recurs after surgery but usually doesn't spread to other parts of the body [17]. The way the tumor looks in a clinical setting really depends on how advanced the disease is [17]. At first, it looks pink-to-bluish-red patch on the skin that can be trophic or sclerotic [17]. Over time, it changes into a lumpy nodular mass and eventually becomes an ulcerative, bleeding tumor that sticks out [18]. It feels like it's just on the surface and moves around when touched since it's attached to the skin above, but not to the tissues below [19]. Unfortunately, in the later stage of the tumor, there may be fixation to deeper structures like fascia and muscle [20]. Scalp fixation due to periosteal attachment might happen in the early stages. Telangiectasia can be seen on the surface or around the edges [21].

The tumor grows slowly, so it can take anywhere from weeks to years to develop [21]. It's pretty common for there

to be a delay in diagnosis and for the initial lesion to be misdiagnosed, mainly because there aren't any symptoms present [21].

Immunohistochemical staining shows strong positivity for CD34 (sensitivity 84–100%) and vimentin while being negative for CD44, S-100, staining. Factor XIIIa staining. The positivity of the last marker and stromelysin 3 (ST3) is helpful for distinguishing benign fibrous histiocytoma (dermatofibroma) [22]. In our case, Histological analysis showed an infiltrative spindle cell proliferation with storiform architecture and CD34 positivity with adjacent areas of fascicular and herringbone growth pattern and extensive necrosis (fig. 3).

The literature review shows that DFSP rarely metastasizes, and in case of distant spread, it involves the lungs most commonly. As an example, Lombart et al. report a case of recurrent DFSP of the chest wall that metastasized to the lungs and liver [23]. In contrast to our case, metastases from squamous cell carcinomas of the skin usually occur in the pulmonary system, whereas metastases to the pancreas are a highly unusual site. There are also cases similar to Kimmel et al. in which there was a local recurrence of the disease with no distant metastases despite multiple resections and adjuvant radiotherapy in a DFSP with fibrosarcomatous change [24]. Nikolaus et al. also described a case of 45-year-old male with DFSP of the trunk, treated with wide local excision and adjuvant imatinib therapy for PDGFB overexpression [15].

DFSPs have a really strong tendency to invade the nearby tissue. The typical treatment method for this tumor involves a wide and deep local excision (WLE), which also includes the underlying fascia. In a study observing 159 cases, it was found that even though 99% of the patient's group

had complete surgical resection, the pathologic review revealed that only 93 patients (58%) had negative microscopic margins [25]. Lindner et al. found that removing healthy tissues in a circumferential range of 2.5–3.5 cm helped improve local control of the disease [26]. Most people seem to agree that having 3–5 cm of lateral and deep margins is enough for controlling the disease locally.

Conclusion

This case highlights the challenges in surgery for recurrent DFSP with fibrosarcomatous transformation. Pancreatic metastasis, the unique presentation, and the aggressive recurrence pattern underscore the need for vigilant long-term follow-up and consideration for systemic therapies beyond surgery and radiation. Optimal treatment strategies for high-risk DFSP patients needs further studies.

Conflict of interests. The authors declare that there is not conflict of interests.

Конфликт интересов. Авторы заявляют об отсутствии конфликта интересов.

The contribution of the authors. Ashraf ALakkad made a major contribution to the development of the concept of the article with writing and editing the case report. Rawia Mubarak Mohamed agreed to take responsibility for all aspects of the case report. Hazem Muhammad Almasarei was responsible for the radiological analysis and its interpretation in the article. Babitha Alingal Mohammed assisted in the analysis of histopathology images. All the authors approved the final version of the article.

Вклад авторов. Ашраф Альлаккад внес основной вклад в разработку концепции статьи, а также написал и отредактировал описание клинического случая. Равия Мубарак Мохамед согласилась взять на себя ответственность за все аспекты представленного описания клинического случая. Хазем Мухаммад Альмасареи отвечал за радиологические исследования и интерпретацию их результатов в представленной статье. Бабита Алингаль Мохаммед помогла проанализировать гистологические изображения. Все авторы одобрили финальную версию статьи.

Список литературы доступен на сайте журнала <https://klin-razbor.ru/>

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Поступила в редакцию: 06.10.2025

Поступила после рецензирования: 14.10.2025

Принята к публикации: 16.10.2025

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Received: 06.10.2025

Revised: 14.10.2025

Accepted: 16.10.2025