Contents lists available at ScienceDirect



International Journal of Surgery Case Reports





Case report

The devastating impact of unresectable infectious undifferentiated pleomorphic sarcoma in the gluteal region: A case report

Rasoul Goli^a, Aysan Torabzadeh^b, Amireh Hassanpour^b, Alireza Jafarimaraghoush^c, Elaheh Manouchehri^d, Navid Faraji^{e,*}

^a Department of Medical-Surgical Nursing, School of Nursing and Midwifery, Urmia University of Medical Sciences, Urmia, Iran

^b Department of Nursing, School of Nursing and Midwifery, Urmia University of Medical Science, Urmia, Iran

^c Department of Technology and Engineering, Civil Engineering, Islamic Azad University of Tabriz, Tabriz, Iran

^d Department of Nursing, School of Nursing and Midwifery, Kermanshah University of Medical Science, Kermanshah, Iran

e Department of Critical Care Nursing, School of Nursing and Midwifery, Urmia University of Medical Sciences, Urmia, Iran

ARTICLE INFO

Keywords: Sarcoma Malignant fibrous Histiocytoma Tumor Case report

ABSTRACT

Introduction and importance: Undifferentiated pleomorphic sarcoma (UPS), previously known as malignant fibrous histiocytoma (MFH), is a highly aggressive soft tissue sarcoma characterized by its pleomorphic histology and lack of differentiation.

Case presentation: A 35-year-old man visited our oncology department with a complaint of a growing mass in his left buttock area. The mass had been increasing in size for the past six months, affected by local and systemic infection. While it was initially painless, the patient started feeling discomfort during sitting and physical activities a few weeks, but later the complication of tumor became more aggressive.

Clinical discussion: UPS can arise in various anatomical sites, including the extremities, trunk, retroperitoneum, and head and neck region. Clinically, UPS may present as a rapidly growing mass, often with pain and limited range of motion. However, the presentation may vary depending on the site of origin. Treatment for UPS typically involves surgical resection, aiming to remove the tumor completely. Depending on the size, location, and aggressiveness of the tumor, additional treatments such as radiation therapy or chemotherapy may be recommended.

Conclusion: Undifferentiated pleomorphic sarcoma (UPS) represents a rare and aggressive soft tissue sarcoma requiring prompt and accurate diagnosis for appropriate management. With its non-specific clinical presentation and histological features, UPS can be challenging to differentiate from other soft tissue tumors.

1. Introduction

Sarcomas, which are relatively uncommon cancers, arise from bones and soft tissues such as fat, muscle, blood vessels, nerves, deep skin tissues, and fibrous tissues [1]. Undifferentiated pleomorphic sarcoma (UPS), formerly known as malignant fibrous histiocytoma, is a complex and heterogeneous soft tissue sarcoma [2]. UPS accounts for a significant portion of adult soft tissue sarcomas, representing approximately 20 % of all malignant mesenchymal tumors [3]. UPS of the buttock is a relatively rare presentation, with limited literature available on its clinical characteristics and management strategies [4].

Undifferentiated pleomorphic sarcoma is a rare and aggressive soft tissue sarcoma that can develop in various anatomical locations [5]. One such location is the buttock, which presents unique challenges in terms of diagnosis, treatment, and prognosis. UPS of the buttock is characterized by a high risk of local recurrence and distant metastasis, leading to significant morbidity and mortality [6]. Despite advancements in diagnostic techniques and treatment modalities, the management of UPS in the buttock remains complex and controversial. By understanding the specific challenges posed by this particular anatomical site, healthcare professionals can optimize patient care and outcomes for individuals affected by this aggressive malignancy. This work has been reported in line with the SCARE criteria [7].

https://doi.org/10.1016/j.ijscr.2024.109592

Received 30 January 2024; Received in revised form 23 March 2024; Accepted 26 March 2024 Available online 27 March 2024

^{*} Corresponding author. E-mail address: navidfaraji1993@gmail.com (N. Faraji).

^{2210-2612/© 2024} The Authors. Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

2. Presentation of case

A 35-year-old male presented to our oncology department with complaints of a progressively enlarging mass in the left buttock region. The patient reported that the mass had been growing over the past six months (Fig. 1). Initially, it was painless, but in a few weeks, he experienced discomfort during sitting and physical activities. He denied any systemic symptoms such as weight loss, fever, or night sweats. Past medical history was unremarkable, and there was no family history of cancer. Radiological imaging, including computed tomography (CT), was performed to assess the extent and nature of the mass (Fig. 2). The images revealed a large, infiltrative mass measuring in the left gluteal region. The tumor appeared to arise from deep tissue layers and infiltrated surrounding structures, encroaching upon the sciatic nerve. No distant metastases were observed at the time of initial evaluation.

The patient was treated with an incisional biopsy of the mass, and subsequent histopathological examination was conducted. The biopsy revealed a highly cellular malignant tumor composed of pleomorphic spindle and ovoid cells with irregular and hyperchromatic nuclei. Mitotic activity was markedly increased, and necrosis was noted. Immunohistochemistry results were inconclusive, with the tumor showing no definitive differentiation markers (Fig. 3). The final diagnosis was established as undifferentiated pleomorphic sarcoma (UPS) of the buttock.

Following the diagnosis, the patient commenced a systemic chemotherapy regimen consisting of a combination of doxorubicin and ifosfamide. A total of 12 chemotherapy sessions were administered to attempt to control the disease and prevent further growth. However, subsequent imaging studies revealed a limited response to chemotherapy, with the tumor exhibiting aggressive growth despite the treatment.

As the disease progressed despite chemotherapy, the treatment approach was discussed in a multidisciplinary setting. Considering the location and aggressive nature of the tumor, surgical intervention was deemed high-risk and not feasible due to the potential for functional impairment. Palliative radiotherapy was initiated to alleviate local symptoms and improve the patient's quality of life. Despite this intervention, the tumor continued to grow, causing local tissue destruction and severe pain.

The decision not to perform a debunking operation in this case was primarily due to the aggressive nature of the undifferentiated pleomorphic sarcoma (UPS) and its infiltration into surrounding structures, including the sciatic nerve. Additionally, the high-risk nature of the surgery, potential for severe functional impairment, and the patient's overall health status contributed to the decision. The presence of infectious agents within the tumor added another layer of complexity, potentially complicating surgical intervention and post-operative recovery. Given the limited response to chemotherapy and the rapid progression of the disease, including pulmonary metastasis, the multidisciplinary team opted for palliative radiotherapy to manage symptoms and improve the patient's quality of life.

Further investigations revealed the presence of infectious agents within the tumor. Microscopic examination of tissue samples showed the infiltration of inflammatory cells and the formation of abscesses, suggesting a possible bacterial or fungal infection. The patient also exhibited symptoms such as fever, an elevated white blood cell count, and localized pain, further supporting the notion of an infectious process. These findings underscored the complexity of the patient's condition, as the aggressive growth and pulmonary metastasis of the tumor (Fig. 4) were compounded by the presence of an infectious component,



Fig. 1. Undifferentiated pleomorphic sarcoma (UPS).



Fig. 2. Radiological imaging of undifferentiated pleomorphic sarcoma (UPS).

necessitating a comprehensive approach to care.

Unfortunately, the patient's condition rapidly deteriorated. He began experiencing recurrent episodes of internal bleeding, likely stemming from tumor invasion of blood vessels in the buttock region. Hemorrhagic shock necessitated emergency interventions, including blood transfusions and surgical hemostasis. Concurrently, the metastatic tumor burden in the lungs further compromised the patient's respiratory function, leading to progressive respiratory failure. Intensive care management was initiated, but the patient's condition continued to decline.

Regrettably, despite aggressive management, the patient succumbed to the complications of undifferentiated pleomorphic sarcoma. His cause of death was attributed to massive internal bleeding and respiratory failure secondary to tumor invasion and metastasis.

3. Discussion

Undifferentiated pleomorphic sarcoma is a high-grade malignant tumor that predominantly affects individuals aged 50 to 70 years, with a slightly higher incidence in males. While it can arise in various anatomic locations, its occurrence in the buttock region presents unique diagnostic and management considerations. Tumors in this region often present at an advanced stage due to their deep-seated location and asymptomatic nature in the early stages. This delayed presentation poses challenges in implementing appropriate treatment strategies, ultimately affecting patient prognosis [8].

In the buttock region, undifferentiated pleomorphic sarcoma typically manifests as a painless enlarging mass with nonspecific symptoms such as discomfort during sitting or physical activities. Due to its deep location, the tumor may go unnoticed until it reaches a substantial size [9]. Diagnostic imaging, such as computed tomography (CT) or magnetic resonance imaging (MRI), plays a crucial role in mapping the extent of the disease and guiding the biopsy procedure. Nevertheless, the imaging findings of UPS in the buttock region can mimic other benign and malignant neoplasms, leading to challenges in accurate diagnosis [10].

Histologically, undifferentiated pleomorphic sarcoma exhibits pleomorphic spindle or ovoid cells with a high degree of nuclear atypia. This pleomorphism often defies lineage identification, making the tumor "undifferentiated" [11]. The immunohistochemical profile varies widely, further complicating diagnosis. UPS can express markers associated with fibroblasts, myofibroblasts, and histiocytes. However, the absence of specific immunohistochemical markers makes accurate diagnosis challenging in some cases [12].

Effective management of undifferentiated pleomorphic sarcoma in the buttock region necessitates a multidisciplinary approach involving surgical oncologists, radiation oncologists, and medical oncologists [13,14]. Wide local excision with negative margins, accompanied by postoperative radiation therapy, is the mainstay of treatment, and also caring of secondary ulcers as well [15,16]. The proximity of vital structures and the requirement for extensive resections to achieve negative margins often complicate the surgical management of buttock UPS. Additionally, the high local recurrence rate and the potential for distant metastasis contribute to the overall poor prognosis associated with this malignancy [17].

Another important measure in the care of most sarcomas is the management of the wound created by the malignant [18]. Nursing care for wounds encompasses a holistic approach that involves meticulous assessment, documentation, and management of wounds [19,20]. This includes identifying the underlying causes of the wound, implementing appropriate interventions, applying various dressings and topical agents, and monitoring wound progress over time [21]. Nurses also play a significant role in patient education, teaching patients about wound care at home and advocating for their needs [22]. Additionally, they collaborate with other healthcare professionals to coordinate a multidisciplinary approach to wound care, ensuring that patients receive the resources and support they need to manage their wounds effectively [23].

4. Conclusion

This case highlights the aggressive nature of UPS and the challenges in its management. It emphasizes the need for further research to understand the underlying mechanisms and explore new treatment strategies to improve outcomes. Efforts should also be made to identify and manage infectious components in tumors, as they can complicate the clinical course and treatment response.



Fig. 3. The result of histopathological examination.



Fig. 4. Footage of pulmonary metastases.

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.

Ethical approval

A written consent was received from the patient. In such case, the anonymised presentation of case report does not require a separate approvement by the ethics committee.

Funding

This CASE REPORT did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Author contribution

Rasoul Goli, Navid Faraji, Amireh Hassanpour, and Aysan Torabzadeh: Study concept, data collection, writing the paper and making the revision of the manuscript following the reviewer's instructions. Elaheh Manouchehri, and Alireza Jafarimaraghoush: Study concept, reviewing and validating the manuscript's credibility.

Guarantor

Navid Faraji.

Research registration number

Not applicable.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Conflict of interest statement

None.

References

- T.F. DeLaney, D.C. Harmon, K. Sikora, F.J. Hornicek, Soft tissue sarcomas, in: Treatment of Cancer, CRC Press, Nov 24, 2020, pp. 434–458.
- [2] B.C. Anzali, R. Goli, N. Faraji, A. Torabzadeh, Z. Najafzad, S. Nikpey, Invasion of undifferentiated pleomorphic sarcoma (UPS) in breast tissue; a case report study, Int. J. Surg. Case Rep. (107) (Jun 1, 2023) 108307.
- [3] F. Zare, N. Shahbazi, N. Faraji, R. Goli, B. Mostafaei, S. Anari, A cruel invasion of Ewing's sarcoma of the skull: a rare case report, Int. J. Surg. Case Rep. (108) (Jun 6, 2023) 108380.
- [4] T. Sambichler, K. Horny, T. Mentzel, I. Stricker, A. Tannapfel, C.H. Scheel, B. Behle, D.R. Quast, Y.P. Lee, M. Stücker, L. Susok, Undifferentiated pleomorphic sarcoma of the breast with neoplastic fever: case report and genomic characterization, J. Cancer Res. Clin. Oncol. 149 (4) (Apr 2023) 1465–1471.
- [5] B. Babamiri, S. Yavari, S. Nikpey, N. Faraji, R. Goli, K. Rahimi, Undifferentiated soft-tissue sarcoma (STS) in a 34-year-old woman: a case report, Int. J. Surg. Case Rep. (105) (Apr 1, 2023) 108104.
- [6] T. Gambichler, K. Horny, T. Mentzel, I. Stricker, A. Tannapfel, C.H. Scheel, B. Behle, D.R. Quast, Y.P. Lee, M. Stücker, L. Susok, Undifferentiated pleomorphic sarcoma of the breast with neoplastic fever: case report and genomic characterization, J. Cancer Res. Clin. Oncol. 149 (4) (Apr 2023) 1465–1471.
- [7] C. Sohrabi, G. Mathew, N. Maria, A. Kerwan, T. Franchi, R.A. Agha, The SCARE 2023 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. Lond. Engl. 109 (5) (2023) 1136.
- [8] R. Sacco, M. Lalevé, P. Pellegrino, A. Ferro, B. Yasmine, G. André, G. Matthieu, A. Hamza, R. Piana, F. Dujardin, Soft tissue sarcomas of the buttock: a systematic review and meta-analysis, Surg. Oncol. (45) (Nov 7, 2022) 10188.
- [9] A. Sakamoto, T. Noguchi, S. Matsuda, A transposition flap reconstruction after resection of a soft-tissue sarcoma in the buttock, J. Orthop. Case Rep. 11 (10) (Oct 2021) 38.
- [10] H. Tsuchie, N. Miyakoshi, H. Nagasawa, Y. Shimada, Tumor lysis syndrome due to eribulin administration for metastatic undifferentiated pleomorphic sarcoma of the buttock, Acta Med. Okayama 75 (4) (2021) 533–538.
- [11] S. Yadsar, M. Abavisani, R. Lakziyan, Z. Sarchahi, Right ventricular undifferentiated pleomorphic sarcoma: a case report, Ann. Card. Anaesth. 24 (1) (Jan 2021) 102.
- [12] P. Rambhia, S. D'Angelo, P. Shah, High-grade undifferentiated pleomorphic sarcoma presenting as acquired lymphangioma circumscriptum, SKIN J. Cutan. Med. 7 (3) (May 20, 2023) 852–853.
- [13] N. Parizad, N. Faraji, R. Goli, H. Salmanzadeh, B. Mostafaei, M.H. Bisafar, Extensive excisional surgery; the last hope for a patient with human papillomavirus-associated severe head and neck squamous cell carcinoma: a case report, Int. J. Surg. Case Rep. (94) (May 1, 2022) 107049.
- [14] N. Parizad, K. Hajimohammadi, R. Goli, Y. Mohammadpour, N. Faraji, K. Makhdomi, Surgical debridement and maggot debridement therapy (MDT) bring the light of hope to patients with diabetic foot ulcers (DFUs): a case report, Int. J. Surg. Case Rep. (99) (Oct 1, 2022) 107723.
- [15] K. Rahimi, L. Hosseinpour, S.M. Balaneji, R. Goli, N. Faraji, B. Babamiri, Large wound surgery of diabetic foot ulcer with Split-thickness skin graft (STSG), and maggot debridement therapy (MDT): a case report, Int. J. Surg. Case Rep. (104) (Mar 1, 2023) 107947.
- [16] R. Goli, N. Faraji, S. Shakorzadeh, M. Abbasi, R. Abbaszadeh, B. Mostafaei, Treating extravasation injury by honey antibacterial wound dressing in a neonate: a case report, Int. J. Surg. Case Rep. (95) (Jun 1, 2022) 107279.

R. Goli et al.

- [17] N.V. Kamat, L. Million, D.H. Yao, S.S. Donaldson, D.G. Mohler, M. van de Rijn, R. S. Avedian, D.S. Kapp, K.N. Ganjoo, The outcome of patients with localized undifferentiated pleomorphic sarcoma of the lower extremity treated at Stanford University, Am. J. Clin. Oncol. 42 (2) (Feb 1, 2019) 166–171.
- [18] Faraji N, Goli R, Bahrami S. An urgent invasion: lower mandible Ewing's sarcoma and unleashed massive abscess; a case report study. Int. J. Surg. Open.:10–97.
- [19] B. Babamiri, F. Nikkhah, N. Faraji, R. Goli, N.V. Moghaddam, K. Rahimi, Diabetic foot ulcer: successful healing with combination therapy, including surgical debridement, maggot therapy, and negative pressure wound therapy, Int. J. Surg. Case Rep. (110) (Sep 1, 2023) 108695.
- [20] N. Faraji, N. Parizad, R. Goli, F. Nikkhah, M. Golhkar, Fighting diabetic foot ulcer by combination therapy, including larva therapy, Medi honey ointment, and silver

alginate dressings: a case report, Int. J. Surg. Case Rep. (113) (Dec 1, 2023) 109055.

- [21] S. Esmaeili, N. Faraji, S. Bahrami, S. Shaddeli, A. Ahmadi, B. Mostafaei, Deep excision surgery of face due to cruel invasion of basal cell carcinoma: a case report study, Int. J. Surg. Case Rep. (109) (Aug 1, 2023) 108551.
- [22] N. Faraji, R. Goli, P. Mohsennezhad, Y. Mohammadpour, N. Parizad, E. Salamat, R. Pourbahram, S. Bazbandi, NSAID (nonsteroidal anti-inflammatory drugs) induced Stevens Johnson syndrome in a 50-year-old woman: a case study, Toxicol. Rep. (12) (Feb 28, 2024) 289–291.
- [23] K. Hajimohammadi, N. Parizad, M. Bagheri, N. Faraji, R. Goli, Maggot therapy, alginate dressing, and surgical sharp debridement: unique path to save unresponsive diabetic foot ulcer, Int. J. Surg. Case Rep. (111) (Oct 1, 2023) 108907.