Giant Cell Tumor of Tendon Sheath Developed over Chimeric-Free Latissimus Dorsi and Serratus Anterior Muscle Flaps

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Abstract

Keywords

- giant cell tumor of tendon sheath
- proteus syndrome
- chimeric-free flap
- microsurgery

This article describes a rare case of giant cell tumor of the tendon sheath (GCTTS) that was developed over the substance of chimeric-free latissimus dorsi and serratus anterior muscle flaps performed for lower limb reconstruction. To our knowledge, development of GCTTS over a free flap is first described in the literature. A 71-year-old woman was presented with a large protuberant ulcerated tumor mass that was developed over the substance of chimeric free muscle flaps at the foot and ankle. We performed an extensive tumor resection, and the pathology report confirmed the presence of a primary giant cell tumor. The patient was advised to have a below-knee amputation. However, the patient refused the amputation, and 4 months later, she was presented with a metastatic mass proximally at the upper thigh. We believe that the GCTTS was associated with the chronic inflammation of the soft tissue and bones along with the recurrent episodes of infection, mainly due to proteus mirabilis and proteus syndrome (PS). PS may lead to the development of malformations and overgrowth of different tissues in unusual locations. In cases resistant to antibiotics, the radical surgical debridement should be considered as the most effective treatment.

Introduction

Giant cell tumor of the tendon sheath (GCTTS) is referred as a benign solitary tumor that usually arises in the limbs. It is less common at the foot where the biological behavior and risk of local recurrence have not been defined.¹ This article describes a rare case of GCTTS that was developed over chimeric-free latissimus dorsi and serratus anterior muscle flaps performed for lower limb reconstruction.

Case Report

A 71-year-old woman was presented with a large protuberant ulcerated tumor mass that was developed over the substance of chimeric-free latissimus dorsi and serratus anterior muscle flaps at the medial aspect of the foot and ankle, a

reconstructive procedure that was performed 5 years ago. As such the time interval between the primary surgery and development of GCTTS was 5 years. The tumor gradually increased in size over a period of 6 months (► **Fig. 1**). The patient had discomfort and increasing pain at rest, while she was able to walk.

The patient had a history of lower limb trauma in childhood, involving the bones and soft tissue of the distal third of the left tibia, ankle, and dorsum of the foot. This old injury left a complex pathologic situation at the lower leg, ankle, and foot, characterized by bony and ankle joint fusion, skin and soft tissue of poor quality, recurrent episodes of cellulitis, chronic ulcers, and verrucous lesions, requiring change of dressings regularly (► **Fig. 2**). Initially the patient and his relatives were advised to undergo a radical procedure including bone and soft tissue debridement leaving a degree of bone shortening, and a

Fig. 1 A large protuberant ulcerated tumor mass was developed over the substance of chimeric-free latissimus dorsi and serratus anterior muscle flaps at the medial aspect of the foot and ankle.

was covered by a chimeric-free latissimus dorsi and serratus anterior muscle flaps (►Fig. 3A, B). The histology report at that time revealed fibrous and necrotic tissue with areas of chronic inflammation. During the period after surgery, the patient took many courses of antibiotics and followed up by specialists. Additionally, she was admitted repeatedly because of recurrent episodes of soft tissue infections.

Initially punch biopsies were taken from the tumor. The pathology report revealed benign neoplasm with features of GCTTS. We performed an extensive tumor resection down to periosteum, and the defect was covered with split-thickness skin grafts. The pathology report confirmed the presence of a primary giant cell tumor, a multinodular encapsulated mass with multinuclear osteoclastic type giant cells, varying degrees of hyalinization, mitosis, increased mitotic activity, nuclear diversity, and atypia (►Fig. 4A, B). These features correlate rather with malignant change and are associated with high risk of tumor recurrence and metastasis. The patient had full-body computed tomographic (CT) scans, which did not show any metastasis, whereas the magnetic resonance imaging (MRI) did not show any bone invasion. The case was discussed with oncologists. Further extensive resection that could provide oncologic safe margins seemed impossible due to the tumor location and the patient's general condition. Therefore, the patient was advised to have a below-knee amputation. Unfortunately the patient refused the amputation. Four months later, she was presented with a

Fig. 2 Skin and soft tissue of poor quality at the lower leg and foot, with recurrent episodes of cellulitis, chronic ulcers, and verrucous lesions requiring change of dressings regularly.

complex orthopaedic and plastic surgery reconstruction that was not accepted. Therefore, we proceed to a generous soft tissue debridement and a “superficial” bony debridement, recognizing the limitations and that the whole pathology might not be treated. The large soft tissue defect involving the distal third of the tibia, ankle, and dorsum of the foot

mass proximally at the upper thigh. A biopsy was performed and confirmed the presence of metastatic disease. Moreover, the CT scan revealed the presence of a nodule at the lung. Thereafter, the patient was under the care of oncologists. To the best of our knowledge, the development of GCTTS over a free flap is first described in the literature.

Fig. 3 (A) A chimeric-free latissimus dorsi and serratus anterior muscle flap. (B) Flap inset was completed.

Fig. 4 (A) The pathology report confirmed the presence of giant cell tumor with varying degrees of hyalinization, mitosis, nuclear diversity, and atypia. Vascularized stroma with focal storiform arrangement of spindle cells. Numerous osteoclast-like giant cells and mononuclear cells with bland and vesicular nuclei. (B) Mononuclear cells were epithelioid or spindled, with eosinophilic cytoplasm, central nuclei, and mitotic figures.

Discussion

GCTTS is a slow-growing benign soft tissue mesenchymal tumor arising from the synovial membrane. The nature is unclear whether these lesions represent reactive lesions or true neoplasm.² There is no exact etiopathology of GCTTS although pathogenetic theories have included trauma, disturbed lipid metabolism, osteoclastic proliferation, infection, vascular disturbances, immune mechanisms, inflammation, neoplasia, and metabolic disturbances.³ The basic theory, as Jaffe et al proposed,⁴ is that of a reactive or regenerative hyperplasia associated with an inflammatory process. It is most commonly found at the flexor aspect of the hand and wrist where local recurrence after excision has been reported in up to 45% of cases.^{1,5} The foot and ankle are rare sites of GCTTS⁶ and may sometimes mimic other lesions such as lipoma, synovial sarcoma, malignant fibrous histiocytoma, synovial cyst, and ganglion. Hence it is important to include this lesion in differential diagnoses, especially if the lesion is found to be anchored to any of the surrounding tendons.⁵ MRI has been proved to be the most useful preoperative investigation as GCTTS has a characteristic appearance that allows planned local excision to be performed.¹

The gross pathologic features include a well-circumscribed lobulated or multi-nodular encapsulated mass with varying degrees of hyalinization. On microscopy, the cellular infiltrate is constituted by macrophage-like mononuclear cells, epithelioid histiocyte-like cells, osteoclast-like giant cells, and xanthomatous cells. Hemosiderin-laden macrophages and cholesterol clefts are also seen.^{5,7} Because of the high incidence of recurrence, radical surgical excision is the treatment of choice.⁸ The most bothering fact to the surgeon is the high rate of local recurrence of up to 45%, after excision.⁹ Osseous involvement, radiologically evidenced by the presence of bony erosions, is a risk factor for local recurrence.¹⁰ There has been no description in literature regarding usage of any local adjuvants, after tumor excision, for preventing recurrence.⁵ Postoperative radiotherapy may have a role in cases in which complete excision may not be possible.⁹ Regular follow-up is necessary in all cases after excision to identify recurrences, if any.⁵

We believe that in our case the development of the GCTTS was associated with the chronic inflammation of the soft tissue and bones along with the recurrent episodes of infection, mainly due to *proteus mirabilis*. Having collected all the wound sub cultures throughout all this period, we documented the

presence of proteus in the vast majority of them. Proteus syndrome (PS) has been well described and is caused by an activating AKT1 mutation (c.49G > A, p.Glu17Lys).¹¹ PS is referred as an extremely rare and complex disease characterized by malformations and overgrowth of different tissues.¹² Many variable features are possible in this mosaic disorder; including disproportionate, asymmetric, and distorting overgrowth; bone abnormalities different from those observed in other disorders; dysregulated adipose tissue including fatty overgrowth and localized fat deposits; and other unusual features.¹¹ Therefore, it is possible that this rare development of GCTTS over the substance of the chimeric flap is associated with PS. Although malignant transformation of GCTTS is rare, a few cases have been reported but not over the substance of a free muscle flap. The presence of the increased mitotic activity in the area of chronic inflammation infection, repetitive minor trauma due to tumor location, and the other pathogenetic factors³ might trigger the transformation of the tumor and the malignant change. It was proved that the eradication of the chronic infection was impossible in spite of the various courses of antibiotics. In this case, the radical surgical debridement should be the most effective way to treat the infection.

In conclusion, GCTTS must be included in the differential diagnosis of a soft tissue lesion around the ankle and may undergo malignant change even though it is rare.³ PS may lead to the development of overgrowth of different tissues such as GCTTS in unusual locations. In cases resistant to antibiotics, the radical surgical debridement should be considered as the most effective treatment.

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None.

Conflict of Interest

None declared.

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