The International Journal of Lower Extremity Wounds
XX(X) 1–4

DOI: 10.1177/1534734613502044
ijl.sagepub.com
SAGE

Squamous Cell Carcinoma and Ledderhose Disease: A Case Report

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Abstract

Ledderhose disease is disorder of the plantar aponeurosis. This disease is not so common and can be tackled with a surgical or conservative approach. A case of a 73-year-old man came to our attention who had a 26-year history of painless bilateral plantar nodules coalescing into an indurated mass. An ulcerative nodule had been noted in the last 16 months on the right foot, in the absence of trauma, not responsive to conservative treatment, so we decided to perform a biopsy. The histopatologic examination showed squamous cell carcinoma, with warty, well-differentiated, low-grade malignancy. Surgical treatment was suggested, so, in pneumoischemia, we made a surgical incision including the skin lesion. Then we proceeded to sculpture the anterolateral thigh fasciacutaneous flap to obtain adequate soft tissue coverage. The tumor was completely removed. Current reconstructive possibilities comprise a good anatomofunctional recovery even in the case of large demolition requests for the therapy of advanced cases of the disease described in this article. Correlation between Ledderhose disease and the formation of malignant tumors has not been made as yet, but perhaps an element that could unite these pathologies can be researched in the lively cell proliferation that characterizes both. It would be interesting to analyze the biological substrate, as well as the systemic and local levels, in patients where both diseases are manifested.

Keywords

Ledderhose disease, Dupuytren disease, La Peronye disease, squamous cell carcinoma

Ledderhose disease, or plantar fibromatosis, is a disorder of the plantar aponeurosis.[1] Plantar fibromatosis is a benign, locally infiltrative neoplasm of the plantar fascia of unknown cause. Twenty-eight percent of patients have associated Dupuytren's disease in the hand. This disease is not so common[2,3] and can be tackled with a surgical or conservative approach.

We describe a particular case of a man affected by Ledderhose disease, who developed squamous cell carcinoma arising on a plantar fibromatosis.

A 73-year-old man came to our attention after a 26-year history of painless bilateral plantar nodules coalescing into an indurated mass (Figure 1). He also had a clinical and histological diagnosis.

The patient also had penile fibromatosis (La Peronye's disease) and Dupuytren's contracture of the hand, both requiring surgical corrections. No family history and no predisposing factors associated with fibromatosis were shown. No diabetic disease was found. He came for the first time to our attention with clinical presentation of thickening of the plantar fascia of both feet, with multiple subcutaneous nodules coalescing into plaques (Figure 1).

An ulcerative nodule had been noted in the last 16 months on the right foot, not responsive to conservative treatment. He was affected by a persistent ulceration on the right plantar fibromatosis, in the absence of trauma. For this reason, an intralesional biopsy was performed, with inguinal lymph nodes echography.

The histopatologic examination showed squamous cell carcinoma, with warty, well-differentiated, low-grade malignancy. However, echography showed only inflammatory lymph nodes bilaterally. Ankle magnetic resonance imaging (MR) and computerised tomography (CT) scan of the lower limbs were perfomed.

Materials and Methods

Surgical treatment was suggested, so he underwent surgical excision of the lesion: a skin-lozenge of 9.7 × 6.8 × 1.3 cm, with an ulcer lesion of 8.5 cm maximum diameter. To obtain adequate soft tissue coverage, surgeons made an anterolateral free-flap, taken from the left thigh. In pneumoischemia, we made a surgical incision including the skin lesion. We proceeded to

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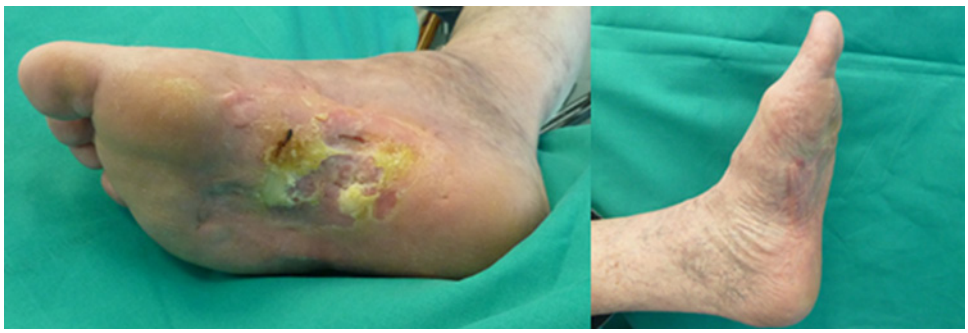

Figure 1. Right and left foot.

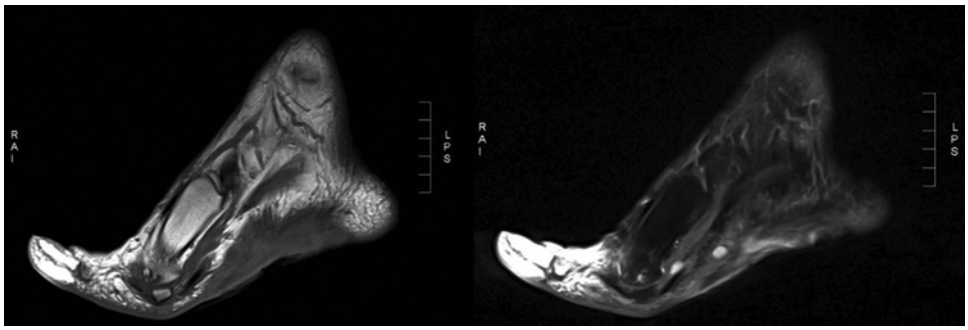

Figure 2. T1 (left) and T2 (right) MR scan of the ankle.

identify vascular axis posterior tibial, posterior tibial artery isolation, and concomitant veins as recipient vessels. Then we proceeded to sculpture the anterolateral thigh fasciacutaneous flap and the isolation of the vascular pedicle based on the descending branch of lateral femoral circumflex artery, through muscle dissection. We performed a terminal-lateral microvascular anastomosis (1-2) between the descending branch of the left lateral femoral circumflex artery and the branch of the right posterior tibial artery, and a T-T of comitantes veins (Figure 3).

Results

The definitive histological exam (Figure 4) confirmed the previous squamous cell carcinoma diagnosis and proved that the tumor had been completely removed.

Discussion

Ledderhose disease is named Dr Georg Ledderhose,[3] who described the condition for the first time in 1894. It is characterized by local proliferative disorder of mature fibroblasts in the plantar fascia. Dupuytren's disease is a similar condition, causing the contracture of aponeural fascia of the hand and the progressive retraction of fingers. It is sometimes associated with other forms of fibromatosis such as La Peronye's disease, or knuckle pads. Only 25% of patients show bilateral involvement.[4]

In the plantar fibromatosis, unlike Dupuytren's contracture of the hand, flexion contracture of the toes is rare.[5,6] However, it is listed as a "rare disease" by the Office of Rare Disease of the National Institutes of Health, which means that it affects less than 200 000 people in the Unites States.[7]

The true incidence of plantar fibromatosis is unknown. De Bree et al[8] reported an incidence of 1 in 100 000. White people are affected more often than any other ethnic group from the sixth decade onwards.[9] It is well recognized in adults and is very rare in children.

Plantar fibromas are well defined and most commonly seen in the medial or central divisions of the plantar fascia. The diagnosis is usually a clinical one and rarely requires confirmation. Fibromas are usually asymptomatic, but if they are located on the medial band, they can produce the compression of the proper digital branch to the great toe and can be painful when walking as the nodule is in contact with the shoe or floor. They can be single or multiple and they can invade the dermis and sometimes flexor tendon sheath and become larger with time.

Platelet-derived growth factor and transforming growth factor-β and others growth factors have been suggested to contribute to the etiology of palmar fibromatosis.[10,11]

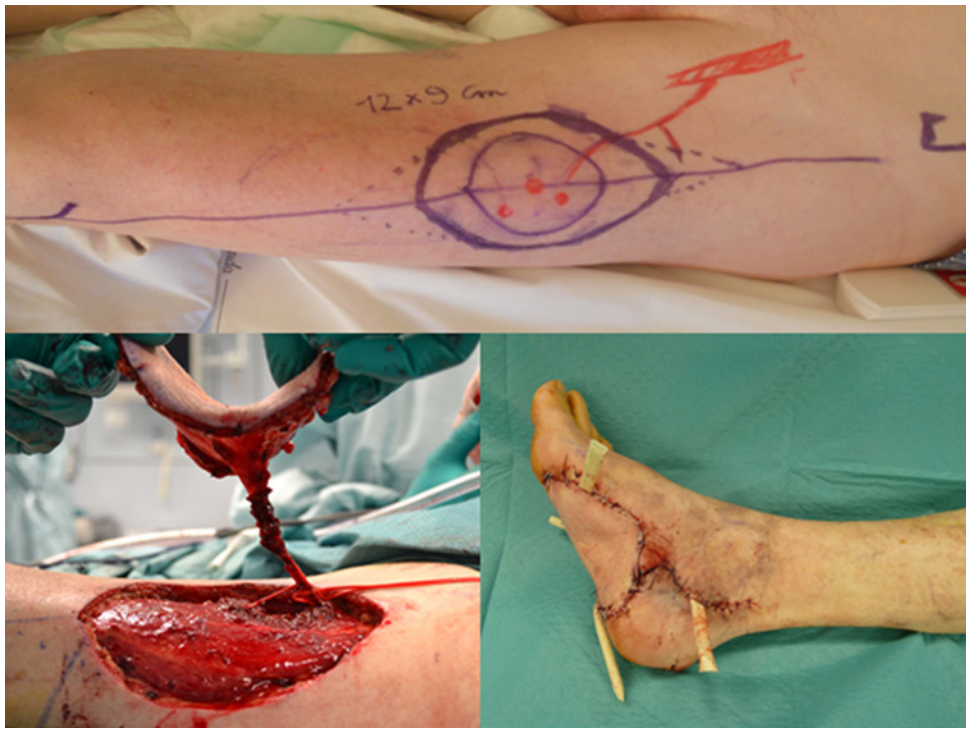

Figure 3. Surgical procedure: anterolateral free-flap for tissue coverage.

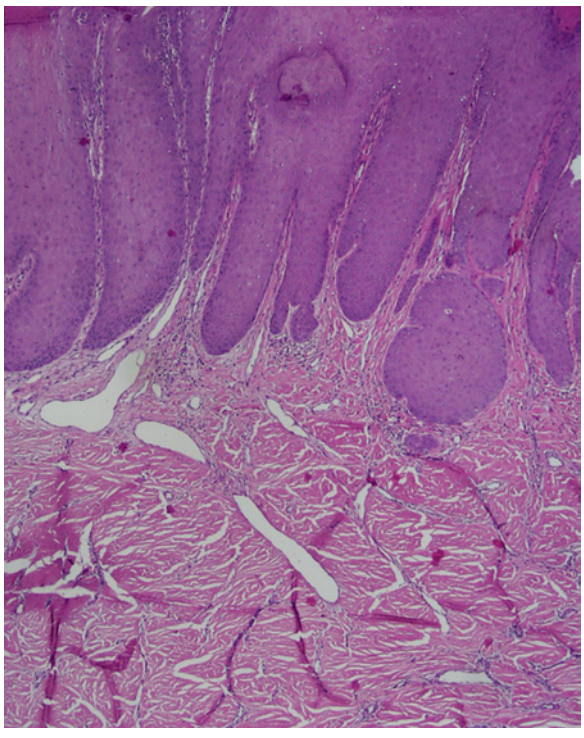

Figure 4. Histologic images of the tissue (HE, 40×).

The clinical classification of plantar fibromatosis has 3 stages: proliferative, involutional, and residual.[12,13] In the first stage there is cellular proliferation, the second stage sees the formation of nodules, and in the third stage there is tissue contraction.

Myofibroblastic proliferation with elongated oval-shaped nuclei and a preponderance of type III collagen are associated with fibromatosis.

The differential diagnosis should include medial plantar nerve entrapment, plantar fasciitis, plantar rupture, tenosynovitis, ganglion cyst, tarsal tunnel syndrome, or other lesions such as fibrosarcoma.[9,14,15]

Conservative treatment includes padding, orthotic support, physiotherapy, and anti-inflammatory drugs, as well as intralesional steroid injections. The surgical options are chosen when conservative measures are ineffective and when the pain persists. The treatment could include local excision, wide excision, and complete plantar fasciectomy. The skin can be separated from the underlying aponeurosis through a zig-zag incision in the plantar aspect of the foot, and the removal of the thickened, cord-like aponeurosis. Is crucial to identify patients who are at increased risk of wound dehiscence and recurrence.[16] Recently, the importance of radiation therapy for primary treatment of Ledderhose disease has been investigated only in a few studies.[17]

Generally skin defects after surgery can require a soft tissue coverage. There are many techniques for obtaining adequate soft tissue coverage: multiple triangular flaps (Z-plasty), free skin grafts, cross-finger flaps, local flaps, or free flaps. In this case we preferred to use a flap microsurgically as a first reconstructive option for the peculiar advantages that this technique offers, such as, only one time reconstructive surgery, a better and more rapid functional recovery, and the ability to recreate the skin and subcutaneous tissue physiologically present in the plantar region by transposition of skin tissue and fascia to cover the loss of substance left after demolition surgery that the disease required. In this way it was possible to obtain a better functionality of the foot, decidedly less difficulty in walking with an increase of resistance to forced sharing.

Modern microsurgery is not averse of the advanced age of patients in need of a major reconstructive surgery. Common disadvantages of surgeries remain in any case, such as vascular and systemic diseases in patients with diabetes mellitus.[18,19]

Furthermore, current reconstructive possibilities comprise a good anatomofunctional recovery even in the case of large demolition requests for the therapy of advanced cases of the diseases described in this article.

Discussion

Plantar fibromatosis is a rare, benign, hyperproliferative disorder of the plantar aponeurosis. The etiopathology of Ledderhose disease is still doubtful. It is likely that the increased production of some cell growth factors may affect the formation of fibromatosis and can determine the progressive worsening of the contracture, as in Dupuytren's disease.[10,11] The correlation between Ledderhose disease and the formation of malignant tumors has not been made yet, but perhaps an element that could unite these pathologies can be researched in the lively cell proliferation that characterizes both.

It would be interesting to analyze the biological substrate as well as the systemic and local levels of the cell growth factors in patients with both diseases manifested. It would thus be possible to find some predispositions that facilitate the degenerative disease processes.

Acknowledgements

The authors are grateful to Dr Matteo Izzo for help with the graphics of this manuscript and to Dr Maggiulli for help with the English language of this presentation in it final version.

Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

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