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Title

Dupuytren's Disease Infiltrating a Full Thickness Skin Graft

Abstract

Although the role of the skin in the development and propagation of Dupuytren's disease remains unclear, dermofasciectomy and full thickness skin grafting (FTSG) appears to delay recurrence. In 2011, a 71-year-old left-handed retired man presented with recurrent Dupuytren's disease in his dominant hand. He originally underwent a primary dermofasciectomy and FTSG in 1991 for Dupuytren's disease involving the palmar skin. Twenty years later, the left middle finger was drawn into flexion by a recurrent cord, and the old graft and adjacent palmar skin were clinically involved by fibromatosis. We performed a revision dermofasciectomy and FTSG. Microscopic analysis of the excised graft demonstrated dense infiltration of the entire skin graft by Dupuytren's disease, with areas of active and burnt-out fibromatosis distinct from hypertrophic scarring. This report of Dupuytren's fibromatosis infiltrating a skin graft raises the questions about the pathophysiology of Dupuytren's disease.

Introduction

Dupuytren's disease is thought to begin within the palmar aponeurosis, progressing axially along fascial bands to infiltrate deeper structures and the overlying skin. Dermal fibromatosis has been demonstrated within the palmar skin of patients with both primary and recurrent Dupuytren's disease[1-3]. Also, dermofasciectomy with full thickness skin graft (FTSG) reconstruction appears to alter the course of recurrence[2,4]. However, the role of the skin in the propagation and recurrence of Dupuytren's disease remains a matter of debate.

We describe a case of Dupuytren's disease infiltrating a skin graft and discuss its significance in our understanding of the behaviour of this common condition.

Case Report

In January 1991, a 71 year old retired left-handed male underwent primary dermofasciectomy of the left palm and middle finger for a metacarpophalangeal joint (MCPJ) contracture of 30º with clinically tethered and deficient overlying skin, attributed to Dupuytren's disease. At the time, the skin from the distal palmar crease to the proximal interphalangeal joint (PIPJ) crease was excised along the midlateral line and the defect reconstructed with a FTSG harvested from the ipsilateral medial forearm. Post-operatively, approximately 20% of the graft failed at its proximal margin and this was allowed to heal by secondary intention, resulting in a tethered scar. This symptomatic scar was excised in 1992 and the defect healed by secondary intention. Twenty years later, the patient presented with a recurrent contracture of the left middle finger and a new contracture in the ipsilateral thumb (Figure 1). Both his family history and medical history was unremarkable. He had never smoked and consumed minimal alcohol. He took no regular medication.

On examination, there was a pretendinous cord originating in the palm along the 3rd ray and investing the PIPJ of the middle finger. The overlying skin graft and adjacent native palmar skin was tethered to the cord. The middle finger flexion contracture was 51° at the MCPJ and 62° at the PIPJ. The left thumb had a prominent Type 3 cord[5] resulting in an abduction deficit of 35°. There were no palpable nodules. The feet and genitals were normal.

In April 2011, a revision dermofasciectomy to the left palm and middle finger and fasciectomy to the ipsilateral thumb was performed under general anaesthesia. The dermofasciectomy involved excision of the graft as well as all the skin and subcutaneous tissue from the palmar crease to the distal interphalangeal joint (DIPJ) crease, along the midlateral lines. The specimen was sent for histological analysis. Despite releasing the accessory collateral ligaments and volar plate, there was a residual flexion deformity of 20° at the PIPJ. A FTSG was harvested from the ipsilateral medial forearm and inset with 5-0 Vicryl rapide® and a tie-over sponge dressing. Fasciectomy of the thumb was performed through a curvilinear incision in the web space which was closed directly. The hand was wrapped in soft dressing and elevated in a Bradford sling.

At 2 weeks, there was 100% graft take, the middle finger MCPJ straightened fully, while the PIPJ continued to demonstrate an extension deficit of 20°. For the first 4 weeks, he received physiotherapy without splinting and nurse-led wound care. He was discharged after 14 months of surgeon surveillance. His result at 3 years following the revision surgery is shown in Figure 1.

Histological assessment of the operative specimens showed that the FTSG was densely infiltrated by Dupuytren's fibromatosis in both the active (proliferative) and burnt-out (involutional) stages (Figure 2).

Discussion

This case shows evidence of Dupuytren's fibromatosis invading non-glaborous skin, which represents atypical disease behaviour and a potentially interesting avenue for future research.

In 1969, Hueston proposed that recurrent Dupuytren's disease should be managed by skin excision and replacement[6]. The benefit of skin excision was explained by Logan and colleagues when they showed Dupuytren's fibromatosis within the dermis and epidermis of patients with recurrent disease, resulting in their conclusion that the only way to adequately treat disease of this magnitude was by radical excision[1,7]. Recently, our group has also shown that Dupuytren's fibromatosis also exists in the skin of patients without clinically apparent skin involvement[3]. Consequently, it has become accepted that the skin plays a role in disease recurrence and extension[1], although the underlying pathogenesis remains unclear. The outcomes of most case series have suggested that dermofasciectomy and FTSG reconstruction may delay the onset or reduce the risk of disease recurrence[4-10]; however, they are underpowered retrospective observational studies and subject to substantial risks of bias and confounding. Intuitively, excision of all diseased tissue should result in a longer disease free interval, whether dermofasciectomy confers any benefit remains uncertain. In our unit, we perform dermofasciectomy and FTSG reconstruction for any recurrent disease or primary disease which clinically involves the skin (defined by pitted deficient skin tethered to a nodule or cord).

The validity of the diagnosis of fibromatosis based on the sequence of events and microscopy alone could be questioned, because the differentiation hypertrophic scarring from Dupuytren's fibromatosis histologically is difficult. The biochemical and immunhistochemical markers of Dupuytren's disease are well described[8] but these are not in routine clinical use and were not used in this case given the archetypal microscopic appearances.

We believe that the burden of skin disease in Dupuytren's fibromatosis is greater than currently perceived and clarifying this could be helpful in better understanding the disease. Complete

excision through the removal of all diseased tissue, as well as a margin of healthy tissue, is the model applied to many surgically treated conditions including fibromatosis in other anatomical locations. However, the same notion has never been discussed with respect to Dupuytren's disease. Varian et al (1990) support the concept of complete excision because they described recurrent cords beneath a FTSG which they attribute to incomplete clearance[9]. Although it seems intuitive to speculate that radical excision may improve disease control[1,2,4,6,7,9], we believe that further prospective clinical research is necessary to better understand this common condition.

Figure Legends

Figure 1. Top row: Preoperative photographs. Lower row: Three years post revision dermofasciectomy with a healed full thickness skin graft.

Figure 2. These are micrographs of the skin graft excised from the palm and stained with H&E. Upper row: low magnification showing fibromatosis in the dermis which is distorting the normal architecture of the skin, obliterating the rete ridges and displacing adnexal structures. Middle row: medium magnification showing a zone of active fibromatosis (arrowheads) in the reticular dermis. The adipocytes (arrows), which would have been on the underside of skin graft, are shown to be encased by the fibromatosis, displacing them into the dermis, away from the subcutis. Lower row: high power magnification showing densely proliferating myofibroblasts typical of Dupuytren's disease.

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