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Bilateral hand dupuytren's disease: Report of a rare case

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Abstract

Dupuytren's contracture was described by Baron Guillaume Dupuytren in 1831 and subsequently, it was a term as Dupuytren's disease (DD). We illustrate a rare case of bilateral DD. A 68-year-old male farmer with no underlying medical illness presented with bilateral hand contractures. It affected his right ring and little finger and left middle and little finger. Further questioning revealed the presence of a neglected gunshot wound involving the right hand. He also had a history of wound debridement for a left-hand abscess, twenty years prior, which had healed via secondary intention. Hand examination showed evident palmar bands over the metacarpals extending to the proximal interphalangeal joints of the affected fingers. Both hands exhibited grade three contractures. Despite the chronicity, high recurrence rate, and history of defaulting follow-up; we still offered him treatment in the form of surgery for his bilateral hand contractures, i.e Dupuytren's release. However, he was not keen on any surgical interventions. This case was worth sharing because DD is usually unilateral, either affecting the right or left hand, seldom both.

Keywords: Dupuytren's disease, Dupuytren's contracture, Bilateral hand, Case report

Introduction

Dupuytren's contracture was named after Baron Guillaume Dupuytren. In his famous lecture, he described the finding of palmar fascia that heal by chronic fibrotic contracture^[1]. As the diagnosis of the disease does not depend on the presence of the contracture itself, hence the term DD was coined^[2].

This benign musculoskeletal fibroproliferative disorder is common, particularly affecting the metacarpophalangeal or the proximal interphalangeal joints. DD usually affecting the ring, small and middle fingers. Many patients have unilateral disease except for a few who have bilateral DD.

Several works of literature agree that there exists a genetic predilection. DD largely occurs in those of North European descent, rarely in Asians. It has been reported to be positively associated with increasing age^[2]. DD is male predominant especially among those over the age of 50 years old. Though the incidence of DD increases with diabetes, smoking, chronic alcoholism, epilepsy, hereditary factors, and certain infections^[2, 3], the exact etiology of DD remains unclear.

DD follows three distinct stages. First is the proliferative phase, second is the involution phase and the third is the residual phase. Histologically, the presence of dense collagenous matrix that predominated by fibroblast cells within the cords of DD which arrange along the longitudinal lines of stress. The collagenous matrix within the cord also shown skew toward the formation of type three collagen more than type one collagen. Hypoxia theory was proposed to explain the pathophysiology of DD. Microvascular narrowing initiates the event leading to hypoxia. This trigger the release of more free radicals by fibroblast that in turn stimulate the proliferation of myofibroblast and consequently contracture of digit^[3].

We illustrate a rare case of bilateral DD that had developed in a Malaysian as a result of a neglected injury and infection.

Case report

A 68-year-old male farmer with no underlying medical illness was referred by a health clinic to the orthopaedic department of the nearby public hospital for bilateral hand contractures. It involved his right ring and little finger and left middle and little finger.

There was an old scar over his right hand denoting a former penetrating injury. It was a remnant of a neglected gunshot wound he acquired 30 years ago. The patient did not want to

disclose the exact mechanism of injury but he informed that he was accidentally shot during an incident involving the taking down of an aggressive cow during a festive season. Wound debridement was done at the local hospital but he could not recall the intraoperative findings. After discharge, he defaulted follow-up and opted for traditional treatment.

Subsequently, this elderly gentleman acquired a left-hand abscess involving the volar aspect of his left hand, ten years later. He denied any precipitating factors. A wound debridement was done. Again, due to non-compliance to post-operative physiotherapy and rehabilitation, healing of the left hand was via secondary intention.

Hand examination showed evident palmar bands over the metacarpals extending to the proximal interphalangeal joints of the affected fingers [Fig. 1]. Both hands exhibited grade three contractures.

Since then, he has retired from farming and wishes to treat his hands. It turned out that he agreed to be referred back to the orthopaedic clinic because he was searching for treatment options for his old contractures. Despite the chronicity, high recurrence rate, and history of defaulting follow-up, he was still offered treatment, i.e. Dupuytren's release. However, again, he was not keen on any surgical interventions. He admits he is already satisfied with his current hand function.

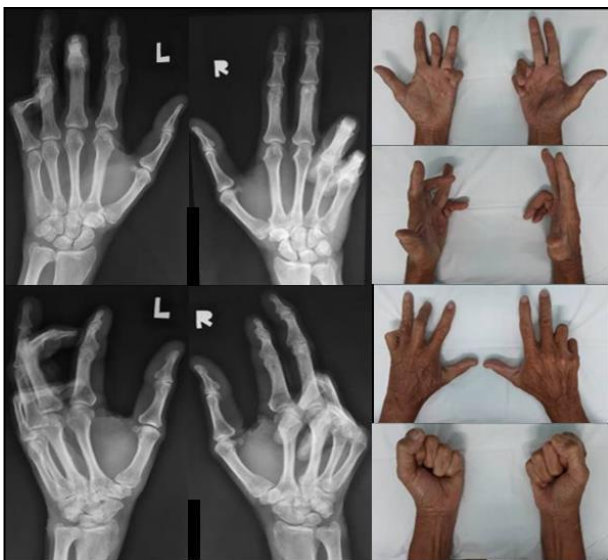


Fig 1: Left: Radiography showing normal carpal bones with no evidence of foreign body or fracture. Right: Note that the right little and ring finger and left little and middle finger are not able to achieve full extension due to the contractures but full hand grip still achievable bilaterally.

Discussion

The patient displayed classical features of DD. Patients may report a decrease in hand range of motion and a loss in hand dexterity. Examination wise, DD usually characterized by the presence of a painful nodule located at the volar aspect of the affected hand and the cords that proximal to the nodule are painless. A positive Hueston tabletop test may suggest DD. The patient was still able to manage his daily routine activities despite the contractures. He claimed he was not able to seek treatment earlier because he was preoccupied with farming work. As the sole breadwinner of his family, leaving work would have meant losing money. The most recent treatment modalities used for the management of DD are extensive percutaneous

aponeurotomy and lipografting, multi-needle aponeurotomy, injecting collagenase *Clostridium histolyticum*, shockwave therapy, and INF-gamma therapy as well as radiotherapy. The treatment option that listed above have their pros and cons and it does not applicable for everyone^[4].

With proper treatment, hand functional outcome is generally good. This case was unique as, regardless of having bilateral hand contractures, the patient still had satisfactory hand function. He had defaulted proper follow-up for both hands but it was unintentional. In a physician's perspective, defaulting is always a negative thing. For patients; particularly our patients, complying with follow-up would be at the expense of sacrificing his time working. Work for some individuals translates into money. In this case, not working would mean financial loss. Every patient has a different socio-demographical, educational, and financial background. Due to this, we should see and understand the patients from their perspective. Over penalizing the patient for defaulting should be avoided.

One study looked into the lives of patients with bilateral DD in which it can be related to the patient in the sense that he presented late and was reluctant to get proper treatment^[5]. They summarized that people with DD often deny the early stages of the disease. Instinctively, they gradually force themselves to adapt to the condition. The condition hits them by surprise when they suddenly have difficulties in completing their tasks or activities. This realization leads them to have actualization of disease. After being aware they have the condition, they seek aid and guidance but are often perplexed on what the problem is and how to manage it. Though the contractures may cause inconvenience, patients are usually ambivalent about whether to surgically treat them and have unanswered concerns or live with it.

This case was worth sharing because DD is usually unilateral, either affecting the right or left hand, seldom both.

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