Dupuytren's disease

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Abstract

Dupuytren's contracture is a common disorder in which proliferation of the palmar fascia leads to contractures of the fingers causing significant loss of hand function. In the early stages Dupuytren's presents with palmar nodules, which are best managed non-operatively, usually with observation. There is little evidence that any non-surgical intervention improves the long-term outcome. If the disease progresses to form cords with contractures that restrict hand function, surgical intervention is indicated. The surgical options range from percutaneous needle fasciotomy, through regional fasciectomy to dermofasciectomy and skin graft. The bigger procedures have a better chance of achieving a full correction of the deformity and a lower risk of recurrence, but the morbidity is greater and the recovery period longer. The choice of treatment is a balance between risks and benefits and should be a shared decision between the patient and surgeon.

Keywords Dermofasciectomy; Dupuytren's disease; fasciectomy; fasciotomy; hand surgery

Introduction

Dupuytren's contracture is a common disorder in which proliferation of the palmar fascia leads to nodules, which can progress to cords, causing contractures of the fingers with a significant loss of hand function.[1] The pathogenesis of Dupuytren's is complex, with proliferation and thickening of the palmar and digital fascia causing firm sometimes painful nodules, puckering of the skin and cords. The cords often contract with time, leading to contractures of the fingers. The fixed flexion of the fingers affects hand function with difficulty using the hand for activities of daily living, particularly wearing gloves, personal care (washing your face) and social situations (handshakes). As the disease progresses, the patient is unable to place their hand flat on a table (the Hueston test).[2]

Signs, symptoms and course

Dupuytren's disease classically presents with tender nodules in the hand. The disease usually progresses in a predictable pattern leading to contractures; however, the rate of this progression varies considerably.[1]

Initially patients may notice pitting or thickening of the skin of the palm or an increase in the subcutaneous tissues with an associated loss of the overlying skin mobility. This puckering and dimpling may become noticeable, which some patients find aesthetically displeasing.[3] An example of early disease can be seen in Figure 1.

The fascia then thickens, forming nodules, usually in the palm around the mid-palmar crease, but sometimes over the palmar surface of the proximal phalanx of the finger, which may become fixed to the skin and the deeper facial structures. Sometimes the disease may present with Garrod's knuckle pads over the dorsal surface of the proximal interphalangeal (PIP) joints; these are inconvenient but not disabling. The nodules are often tender, particularly during the early proliferative phase of the disease.[4]

Most patients will then develop tight fascial bands called cords, which thicken and contract over time. This leads to contractures with fixed flexion deformities in the fingers and a loss of finger extension of the metacarpophalangeal and proximal interphalangeal joints.[1] This impairs hand function. These contractures often progress and if left untreated may leave the fingers clasped tightly in the palm with a profound functional impairment and difficulties with hand hygiene.

Figure 2 shows a more severe example of Dupuytren's disease. These severe cases may lead to permanent joint contractures. In bilateral disease, the severity is often asymmetrical. Dupuytren's is a clinical diagnosis with a very limited role for any special investigations.[3]

Dupuytren's diathesis is a more aggressive form of Dupuytren's disease, which tends to present earlier, progress more rapidly and carries a higher recurrence risk following treatment. It is characterized by a positive family history, more severe disease affecting more digits or with bilateral involvement, with an earlier onset and faster progression. These patients may also present with ectopic manifestations: Garrod's disease with disease of the dorsal knuckle pads, Peyronie's disease of the dartos fascia of the penis and Ledderhose disease of the plantar fascia.[1,2]

Pathophysiology

Aetiology

Dupuytren's disease is a progressive fibroproliferative disorder of the palmar and digital fascia, in which myofibroblasts (contractile fibroblasts with intracellular actin filaments and extracellular

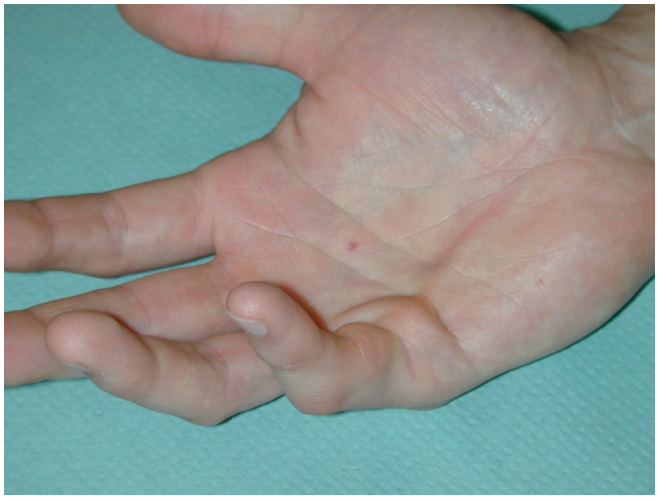

Figure 1 Early Dupuytren's disease with thickening of fascia and skin puckering.

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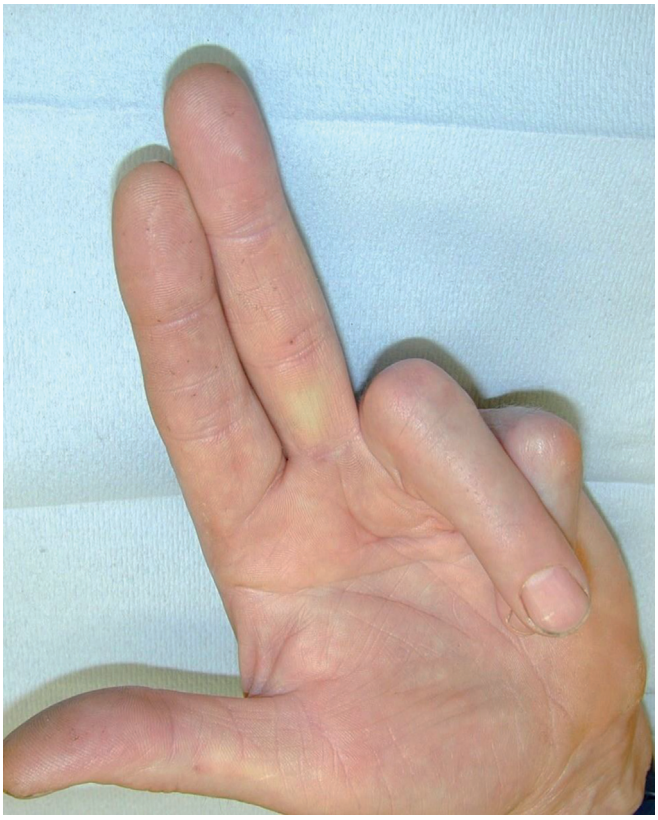

Figure 2 Severe Dupuytren's disease, with significant contracture of the ring and little fingers.

fibronectin) proliferate abnormally causing uncontrolled growth in the palmar fascia, with nodule formation.[5]

The resulting fibrosis and low-grade inflammation are associated with macrophage and lymphocyte recruitment. The nodules are highly cellular, with large numbers of myofibroblasts, macrophages and lymphocytes seen on microscopic studies.[4]

Tissue necrosis factor signalling though the Wnt signally pathway promotes the differentiation of fibroblasts into myofibroblasts, which are contractile. High levels of proinflammatory cytokines have been detected in Dupuytren's tissues. Dupuytren's has also been associated with impaired release and degeneration of TGF-beta.[6]

The proliferation of myofibroblasts causes a change in the ratio of type 3 collagen to type 1 collagen within the fascia, with a fibroplastic hyperplasia. This leads to thickening and a resulting contraction of the longitudinal palmar fascia. These tight facial bands, called cords, lead to the development of the characteristic contracture and are formed from predominantly mature fibrillar type 1 collagen with very few cells.[7]

Luck's classification

In 1959 a histological staging system for Dupuytren's was described, classifying the disease into three stages. This is shown in Table 1.[5]

Cord formation

Normal palmar fascia becomes pathological cords. Thickening of the palmar fascial bands forms pretendinous cords, superficial to the flexor tendons in the palm. Pretendinous cords are the most commonly identified and are often contiguous with digital cords; they may cause flexion contractures at the metacarpophalangeal joint (MCPJ).

Luck's classification: A classification described in 1959, with three stages of disease: proliferative, involutional and residual

Stage	Description
Proliferative stage	Characterized by the presence of large numbers of myofibroblasts and immature fibroblasts. This hypercellular stage is extremely vascular with minimal extracellular matrix. Clinically this coincides with nodule formation.
Involutional stage	Microscopically a dense myofibroblast network is seen, with alignment along tension lines and increased collagen production, with increasing ratio of type 3 to type 1 collagen.
Residual stage	This stage is largely acellular, with disappearance of myofibroblasts leaving fibroblasts as the remaining cell line. The residual tissue is collagen-rich dense scar tissue.

Table 1

At the palmodigital junction, around the metacarpophalangeal joints, natatory and spiral cords are found. Natatory cords are found in the web space running transversely. Natatory cords are responsible for webspace contractures and originate from the natatory ligaments.[5] Digital cords lead to contractures of the interphalangeal joints. The pathoanatomy in the finger is complex.

Digital fascial bands may also be affected, with the central cord continuing from the pretendinous cord, attaching to the flexor tendon sheath over the middle phalanx. The lateral sheet thickens to a lateral cord attaching to the flexor structures or Grayson's ligament causing flexion at both the proximal and distal interphalangeal joints. The lateral cord may also displace the neurovascular bundle due to its large size.[5]

Spiral cords are less common, but are important as they are technically challenging to remove due to their relationship to the neurovascular bundle. Recently the origin of these spiral cords has been investigated, describing a palmodigital spiralling sheet closely involved with the neurovascular bundle starting superficial to the neurovascular structures at the MCPJ before passing deep the bundle. This sheet becomes diseased, forming the spiral cord; it causes displacement of the neurovascular bundle.[8] Understandably this displacement complicates surgical removal of the cord and changes the expected anatomical position of vital structures.[8]

Distal interphalangeal joint (DIPJ) contractures are unusual, but are typically caused by a retrovascular cord, a thickening and contracture of the retrovascular band, running dorsal to the neurovascular bundle; incomplete removal of this cord can prevent full correction of DIPJ flexion.[5] Notably, Dupuytren's disease is said to spare both Cleland's ligament and the transverse ligaments of the palmar aponeurosis.

Free radical hypothesis

It has been hypothesized that free radicals may cause the development of Dupuytren's disease through stimulation of fibroblast proliferation and collagen deposition. Oxygen free radicals are generated when microvessels narrow, which may happen due to comorbidities such as smoking, diabetes and aging.[1,9]

Incidence

Dupuytren's is predominantly a disease of the elderly, with an increased incidence in Western European men. Dupuytren's is four times more common in men. 12% of 55-year-olds, and 29% of 75-year-olds will develop Dupuytren's disease; 20%–40% of these patients develop a contracture within 7–18 years.[10]

Historically, Dupuytren's was thought of as a 'Viking disease', with the hypothesis based on the prevalence of the disease in people of Scandinavian or Norse genetic descent. However, a genetic study using the British population in 2020 concluded that there was no evidence for a link to Viking ancestry.[11] Further work highlighted the presence and prevalence of Dupuytren's disease in the Japanese population, adding further evidence against the Viking disease hypothesis.[12]

Risk factors

Risk factors for Dupuytren's disease have been well documented and the main risk factor for developing the disease is genetic, with up to 80% heritability, however the complex genetics are still being understood, with recent genomic meta-analysis highlighting 56 genetic loci related to Dupuytren's disease.[13,14]

Modifiable risk factors include excessive alcohol use (conveying a 2–5× risk), diabetes (a 2–3× risk) and smoking (a 3× risk).[15] There is evidence of a link between Dupuytren's disease and seizure disorders, HIV, epilepsy, carpal tunnel syndrome, RA and vascular disease. Vibration exposure has also been identified as a risk factor.[1]

Some studies have suggested that Dupuytren's disease carries an increased overall mortality of about 22% across ages and 70% higher mortality for patients whose Dupuytren's disease came on before the age of 60.[16]

Prognosis

The disease process of Dupuytren's disease is currently incurable, however there are treatments that can help to release the contractures and restore hand function. There is a tendency for the disease to recur in treated digits and extend to other digits.[17] Approximately 10% of afflicted patients experience spontaneous disease regression.[3,18]

Surgical release improves hand function, but the cords can reform causing further deformity. Recurrence rates vary significantly based on management modalities and other factors including patient characteristics and disease factors, with recurrence rates after surgery reported of between 0 and 85%.[19,20]

Staging of the disease

The disease is usually staged according to the severity of the contracture and the number of digits affected. Tubiana described the first staging system, and subsequent modifications of the Tubiana staging can be used to classify Dupuytren's disease based on the degree of deformity, measured as the angle at the metacarpal phalangeal and interphalangeal joints, as shown in Table 2. The modified Tubiana staging incorporates the original total flexion deformity measurement and additional variables, such as the number of surgical interventions and total number of affected joints, have also been incorporated (shown in Table 3).[21]

Management

Management options for Dupuytren's contracture include physiotherapy, steroid or collagenase injections, radiotherapy and surgical intervention. Surgery for Dupuytren's disease to 'straighten a bent finger rather than cure the disease'. There are several types of surgical interventions, which can be divided into those aiming simply to divide the Dupuytren's cords (a fasciotomy) and those which remove part or all the Dupuytren's disease (a fasciectomy). In some severe cases the skin is removed and replaced with a graft a dermofasciectomy. Poorer outcomes are seen in patients with interphalangeal joint fixed flexion deformities than in those with only the metacarpophalangeal joints affected.

Tubiana staging

Stage	Deformity
0	No lesion
N	Palmar nodule without presence of contracture
1	Total Flexion Deformity 0 to 45
2	Total Flexion Deformity 45 to 90
3	Total Flexion Deformity 90 to 135
4	Total Flexion Deformity >135

Table 2

Modified Tubiana staging

Criteria	Score
Surgical procedures	Total number of operations for left and right hand
Recurrence	Total number of recurrences for left and right hand
Number of digits affected	Total for left and right hand
Number of nodules	Total for left and right hand (palmar/digital)
Number of pits	Total for left and right hand
Garrod's pads	1
Ledderhose's disease	1
Peyronie's disease	1
Bilateral or unilateral	1 for unilateral and 2 for bilateral
Stage 1 – Total Flexion Deformity of each digit 0 to 45	1
Stage 2 – TFD 45 to 90	2
Stage 3 – TFD 90 to 135	3
Stage 4 – TFD greater than 135	4

Table 3

Correction of the contractures allows patients to regain lost function. Recurrence rates are difficult to predict for individual patients, but are generally lower after more extensive surgery, and are further reduced by skin excision and grafting. It is generally considered that there is no benefit from very early surgery.

The NHS in England has criteria for treatment and service commissioning, which include the degree of deformity and its impact of function. An extension deficit of at least 30 degrees and an inability to put the hand flat on a table with functional restrictions are required before surgical management is funded. There is a useful NHS England Dupuytren's decision tool which guides patients through the available treatment options for their disease.[22]

Injection therapy

Injections can be useful particularly in the earlier stages of Dupuytren's disease; steroid injections have been shown to reduce the activity of the myofibroblasts *in vitro*.[23] However, they are not currently recommended by the NHS due to the lack of evidence of any effect upon the long-term outcome of the disease. Steroid injections can help with painful nodules but have no effect on contracture.[24]

Collagenase injections use a genetically engineered enzyme from *Clostridium histolyticum* to soften and disrupt the affected cords, allowing for manipulation of the digit to rupture the cords and correct contractures. Collagenase injections are well tolerated, with lower complication rates and recurrence rates than surgery. There is a risk of skin splits and injury to tendons and nerves. Commercial considerations have led to its withdrawal from the European market.[25]

Anti-tumour necrosis factor (adalimumab) injections have been proposed. Randomized controlled trials have shown improvements in the nodule hardness and size with no significant complications, with effects lasting up to nine months after the final injection. There is also some evidence that it slows the progression of disease.[26]

Percutaneous needle fasciotomy

Percutaneous needle fasciotomy (PNF) is a minimally invasive procedure that corrects contractures by perforating pretendinous palmar cords using a needle under local anaesthetic. A forceful hyperextension of the affected joint is performed by the surgeon, rupturing the cord and restoring passive extension.[27] This procedure is considered to be safe, with a low complication rate. The patient is awake and able to provide feedback as well as actively move their flexor tendons. Recovery is quick with patients often returning to work within a few days. Costs are low, with very limited equipment and staffing required, however the recurrence rates after PNF are very high, with 60%–85% recurrence within 2–3 years.[27,28]

Percutaneous needle fasciotomy is best utilized for patients with mild to moderate contractures with a well-defined cord; it is less effective in patients who will lack compliance, have more severe disease or who have secondary joint changes.[27]

Limited or segmental fasciectomy

Limited fasciectomy is the surgical removal of a segment of a Dupuytren's cord through a skin incision in the palm or finger overlying a palpable cord. This procedure can be a very effective and safe treatment, interrupting the continuity of a cord and removing a section of diseased tissue, restoring extension and improving contracture as shown in Figure 3. Further work may be required to reduce tension on the skin and allow closure.[29]

Limited fasciectomy has a lower recurrence rate than percutaneous needle fasciotomy, but has greater morbidity, longer recovery and slightly increased risks of complications including infection, bleeding, neurovascular damage and tendon injury.[28] Studies have shown limited fasciectomy to be safe even in recurrent disease with repeat limited fasciectomy.[29]

Digital and palmodigital fasciectomy

In more severe or widespread disease, a more extensive fasciectomy may be required with resection of the majority of the

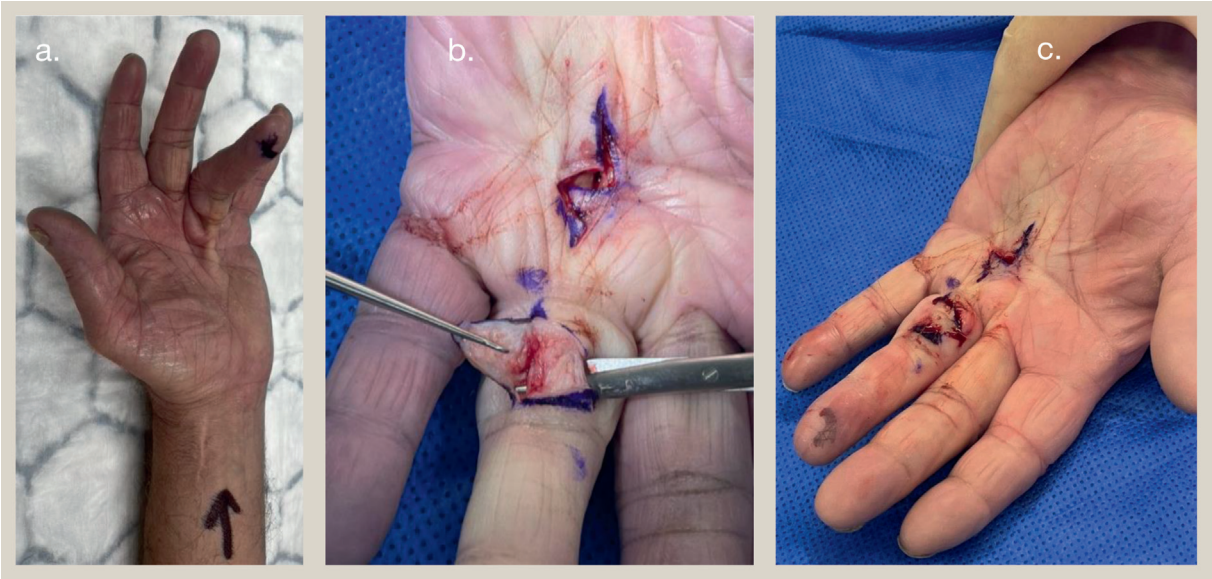

Figure 3 Limited fasciectomy for Dupuytren's disease. (**a**) The preoperative deformity, (**b**) an intraoperative image showing the incisions and surgical procedure, (**c**) Postoperative improvement in deformity.

disease performed through a larger incision as shown in Figure 4. Straight incisions are prone to contracture if used on the palmar surface of the finger and are closed with a Z-plasty, alternatively a Brunner incision is used to avoid this. The cords are excised in the palm and into the affected digit. The cords are usually resected *en bloc* to aid the identification and removal of the diseased fascia. Additional care is needed to identify and protect the neurovascular bundles due to the close adherence of the cords to these key structures, and there is a risk that a spiral cord may displace the NV bundle.[30]

The aim of a palmodigital fasciectomy remains the same, to restore the function in the affected finger, and the more widespread resection reduces the risk of recurrence. This more extensive procedure is usually recommended in more severe or recurrent Dupuytren's disease, while digital fasciectomy is often required for contracture at the proximal interphalangeal joint. Risks remain similar but, overall, this is a more complicated and risky procedure with neurovascular bundles more at risk due to their proximity.[1]

Dermofasciectomy and skin grafting

Surgery for severe contracture is challenging, and complicated by restricted access to the area as well as associated skin contractions, which may well need skin grafting. In dermofasciectomy, a more radical resection is undertaken, with areas of involved skin also resected; the resulting skin defect requires skin grafting.[1]

Skin grafts are usually full thickness and can be harvested from the ipsilateral forearm with satisfactory cosmetic results. They are often required in more extreme contractures, where the skin is heavily involved with the disease. Skin grafting carries its own additional risk and graft take failure may lead to further operations or morbidity.[30] A 'firebreak graft' has been hypothesized to reduce the rate of recurrence, with a full thickness skin graft from an unaffected area placed within a flexion crease to reduce disease recurrence by preventing consistent cord formation.[31] A recent systematic review has indicated that skin grafting reduces the recurrence rate of Dupuytren's disease, though recurrence rates varied significantly.[30] Dermofasciectomy is indicated in younger patients, more severe disease and in surgery for recurrence.

Fasciotomy vs fasciectomy

Multiple comparisons of the effectiveness of fasciotomy and fasciectomy has been done, with systematic reviews identifying variation in the reported outcomes. Fasciectomy is more effective in correcting contractures than fasciotomy. The average recurrence rates for fasciotomy are considerably higher (62% vs 39%) at 4 years.[32] However, the morbidity of treatment is greater.

Amputation

The last resort for the management of Dupuytren's disease is amputation of the finger. This may be an appropriate choice in severe or recurrent cases. Amputation may be indicated in severely contracted fingers causing functional loss or pain, after failed previous interventions or when the finger has become compromised due to neurovascular injury or other severe complications.[1] Metacarpophalangeal joint amputation or even more proximal amputations eliminate the risk of recurrence. However, the amputation carries a risk of a painful neuroma or phantom pains occurring.[33]

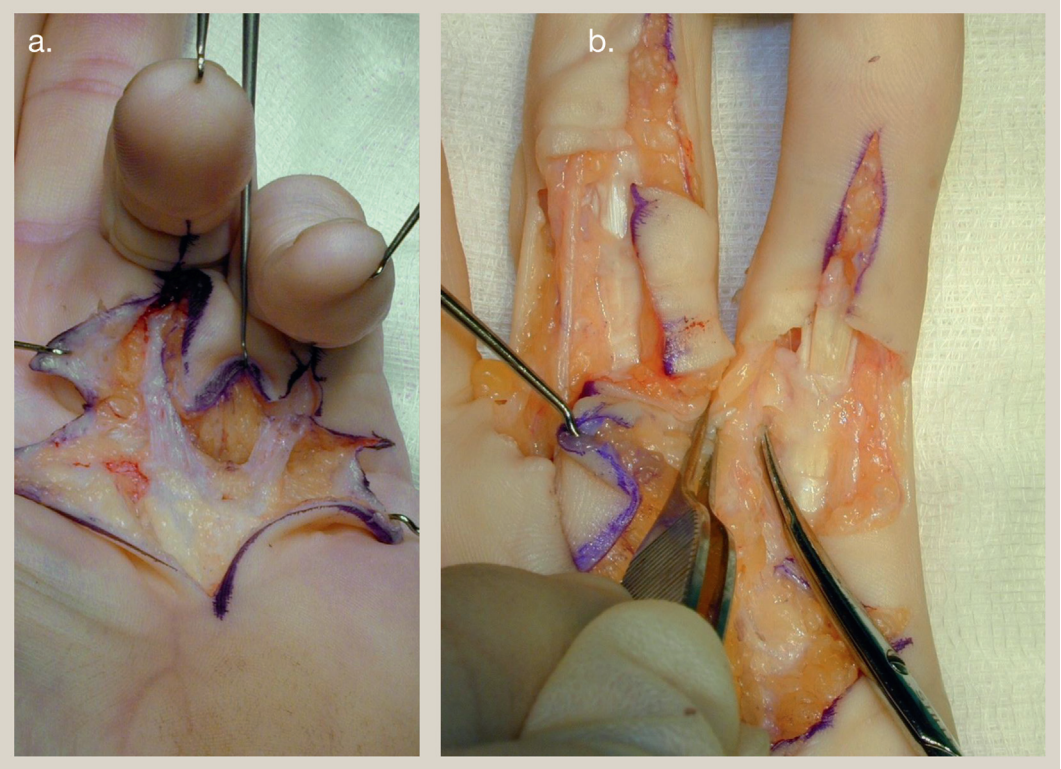

Figure 4 Palmodigital fasciectomy for Dupuytren's disease. (**a**) Intraoperative image showing the appearance of cords in the palm. (**b**) Intraoperative image showing the improvement of deformity with fasciectomy.

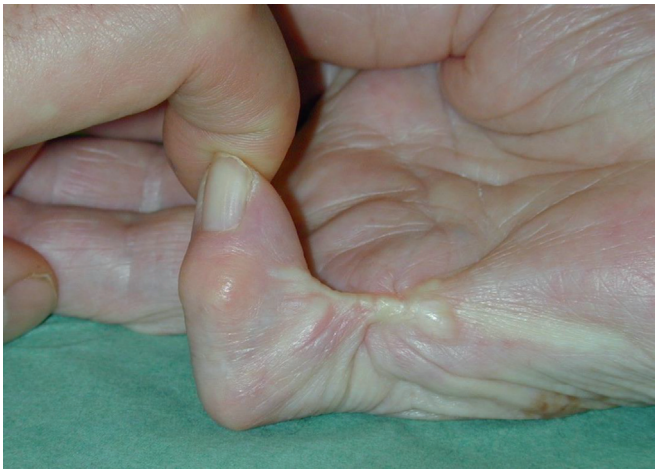

Figure 5 Recurrent Dupuytren's disease with significant deformity and evident skin loss and scarring.

Revision for recurrent contracture

Revision surgery for recurrent Dupuytren's contractures is technically challenging, complicated by multiple factors. Following previous surgery, patients may develop dense scarring, which increases the complexity of dissection and alters and obscures normal planes, with associated deformity as seen in Figure 5. Neurovascular bundles are often difficult to identify and may have moved from their expected anatomical positions.[1]

Revision surgery is also complicated by structural joint contractures, and skin shortages leading to larger areas needing skin grafting.

Radiotherapy

Radiotherapy may be utilized for Dupuytren's disease, with low-energy x-rays used to soften thickened tissue and prevent worsening of the disease, slowing or stopping the progression. This involves multiple doses of radiation over a couple of short courses. Symptoms have been seen to improve and the size of nodules have been seen to reduce. Side effects are mainly from radiation burning in the form of red, dry, itchy skin. Radiotherapy does not correct established contractures and the extent to which it slows the progression of disease is as yet unproven.[9]

Risks

Risks involved in Dupuytren's surgery vary with different surgical techniques, but surgery poses a risk to the neurovascular bundles, and flexor tendon sheath with common issues including stiffness, numbness and cold intolerance. A rarer yet debilitating complication of chronic regional pain syndrome has also been reported.[1,29] More extensive surgery has been shown to have greater morbidity and risks.[34]

Conclusion

Dupuytren's contracture is a common disorder in which proliferation of the palmar fascia leads to contractures of the fingers causing significant loss of hand function. In the early stages, Dupuytren's presents with palmar nodules, which are best managed non-operatively, usually with observation. There is little evidence that any non-surgical intervention improves the long-term outcome. If the disease progresses to form cords with contractures that restrict hand function, surgical intervention is indicated. The surgical options range from percutaneous needle fasciotomy, through regional fasciectomy to dermofasciectomy and skin graft. The bigger procedures have a better chance of achieving a full correction of the deformity and a lower risk of recurrence, but the morbidity is greater and the recovery period longer. The choice of treatment is a balance between risks and benefits and should be a shared decision between the patient and surgeon. ◆

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Practice points

- Importance of clear understanding of the pathophysiology of Dupuytren's
- Understanding of the displacement of neurovascular bundles due to spiral cord formation
- Use of the NHS Dupuytren's decision-making tool to guide and educate patients about the management of Dupuytren's disease
- Treatment options for Dupuytren's disease