Assessment of clinical severity in Dupuytren's disease

Every clinician should be aware of the key features in the history and examination of the patient with Dupuytren's disease to know when surgical referral is appropriate and to inform the patient about causation and prognosis. This article outlines the most relevant questions to ask and provides a systematic approach to the clinical examination.

The curious condition of the hand first operated upon by Baron Guillaume Dupuytren will be encountered by all doctors, who may be poorly placed to provide the patient with accurate information on causation and prognosis since much of the information in medical texts is at best lacking an evidence base, and at worst frankly wrong. Every doctor should know how to inform his/her patient about the relevance of this condition to general health and when to refer for a surgical opinion. The general physician will benefit from knowing the evidence basis for all of the suggested associations.

Originally termed Dupuytren's contracture, the term Dupuytren's disease has been more widely adopted as the fingers are not always flexed. Dupuytren's disease is a common connective tissue disorder that affects the hands, principally affecting northern European Caucasians (Bayat and McGrouther, 2006). It is not clear how common Dupuytren's disease is but, dependent upon the particular population group, prevalence rates are usually cited between 4% and 20% (Hindocha et al, 2006a), reaching as much as 70% of a population followed over 30 years in some Northern European countries, e.g. Denmark (Mikkelson, 1977a).

Typical presentation is of a palmar nodule located near the distal palmar crease. Over a variable time period, the nodule progresses to form a fibrous cord which extends from the palm into the digits causing metacarpalphalangeal joint (MCPJ) contracture. The nodules or cords may start in the digits or extend from the palm leading to proximal interphalangeal joint (PIPJ) contracture and in the absence of surgery resulting in permanent, irreversible flexion contractures of the affected joints.

The optimum time to operate is when there is functional deformity or clear progression of MCPJ and PIPJ contracture (Bayat and McGrouther, 2006). The functional outcome depends upon the duration and severity of contracture; PIPJ contractures have poorer outcomes as a result of contracture of the check-rein ligaments, delicate structures located proximal to the palmar plate which normally telescope and extend on joint motion (Honner et al, 1971). Indicators for surgical correction include progression of the contracture within the preceding year and functional disability (Bayat and McGrouther, 2006).

Once seen for the first time, most clinicians are able to diagnose straightforward cases, but specific clinical signs can be helpful in unusual cases and other features are relevant to prognosis. The 'Dupuytren's diathesis' is a constellation of features which can be considered adverse prognostic indicators. The features include: bilateral disease, ethnicity, the presence of what are often termed 'ectopic lesions' (plantar, penile and knuckle lesions) but which may alternatively be viewed as separate diseases in their own right, and positive family history (Hueston, 1974, 1990). More recently the diathesis factors have been refined to include: family history of one or more siblings or parents with the disease, male gender, and age of onset younger than 50 years (Hindocha et al, 2006b).

Dupuytren's disease is also associated with epilepsy, diabetes mellitus, and frozen shoulder with the disease course in each being variable. Diabetic patients appear to display more widespread nodules than usual. Other statistical associations are controversial, such as relationship to alcohol or tobacco. Many teetotal non-smokers have the condition. There is no simple cause and effect relationship and there is no proven link to work, injury, or the much quoted but never substantiated 'vibrating tool use'. The patient with Dupuytren's disease will suffer an unpredictable disease course with potentially devastating loss of function. It is therefore imperative that clinicians have clear guidance on severity assessment and are confident about when referral to a specialist hand surgeon is appropriate.

Patient often arrive for the outpatient appointment armed with printouts from the internet, usually giving an unbalanced view of the condition and offering novel unproven treatments. The doctor needs to detect the optimism, pessimism, guilt, anxiety and unrealistic expectations which this information is likely to have engendered, and restore a state of balanced realism.

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The focussed medical history

The most clinically relevant factors to elicit from the history are as follows:

Ethnicity, gender and age of onset

Ethnic origin has been described as the most important single element determining incidence (Hueston, 1990). Northern European Caucasians are more likely to express the disease than other races (Hindocha et al, 2006a), especially when compared to blacks where the disease is rare (Mennen, 1986). However, the incidence in Asian subcontinent people living in Birmingham is higher than anticipated (Srivastava et al, 1989) suggesting environmental factors may have some relevance. The disease is more common and more severe in males, with the typical presentation of a man in his fifth decade, but there are reported cases in children and onset is possible at any age in adult life (Hughes et al, 2003). Females tend to present later with a peak incidence between the age of 60–70 years and they tend to have less severe disease.

Symptoms and disease locations in the hand

The most common presenting feature is the appearance of a solid painless nodule in the palm. However, the usual description of Dupuytren's disease as a painless condition may be incorrect as 5% of patients complain of local tenderness (Mikkelson, 1977a). Clinicians should therefore ask patients about tightness, tingling and pain in the area overlying the skin pit or nodule. This is particularly relevant to early disease where burning sensations, itching and tension may be precursors of disease onset in some (Bayat and McGrouther, 2006).

The disease manifests bilaterally in most patients and there is no firm relation to handedness (Hart and Hooper, 2005). The most commonly affected digits are the ring and little fingers, followed by thumb, middle finger and index (Hughes et al, 2003). Close inspection of the other hand is required in patients who present with unilateral disease as knuckle pads (nodules over the dorsal surface of the PIPJs, also termed Garrod's pads) may be apparent. The patient is generally unaware of these, and even where they are noticed, patients usually ascribe them to traumatic causes. Some of these patients may go on to develop palmar lesions in the seemingly unaffected hand and they should be aware of this.

Dupuytren's disease of the radial aspect of the hand refers to lesions arising from the thumb and the first web space (Milner, 2003). Patients can maintain adequate function even in the face of limited ranges of thumb movement and so they may present later. Radial Dupuytren's disease is considered uncommon but could be underreported if clinicians are not looking for it (Tubiana et al, 1982).

Rate and course of disease progression

Some individuals develop contractures in months, while others can take several years (Leclercq, 2000). Overall, the aggression of contracture is influenced by the age of onset and the presence of diathesis factors. Younger patients are likely to have worse disease progression, although the course in the individual is very variable.

Family history of Dupuytren's disease

Patients should be asked if they are aware of other family members who are known to have this condition and at what age their disease began. Family history is significant when more than one close relative has had the disease and more than one generation is involved. Few will know the medical details of their ancestors, or even siblings, and all studies note underreporting of incidence (McFarlane and Botz, 1990).

Perhaps the greatest significance of family history, however, is the way in which it will condition the patient's expectations of disease progression and treatment outcomes. Patients' descriptions of the operations on relatives will vary from 'simple' to 'horrendous' and the patient should appreciate that the course of disease and treatment will be different for every patient, every hand and even every digit. Their experience is unlikely to parallel that of a relative for better or worse. Considerable surgical experience will be necessary to predict future progression with any likelihood of accuracy.

Associated conditions

The patient may have any of the following:

- Insulin-dependent diabetes mellitus
- Epilepsy and medication
- Frozen shoulder, carpal tunnel or trigger finger.

Dupuytren's disease in the diabetic patient has a reported prevalence between 3 and 32% (Noble et al, 1984; Geoghegan et al, 2004) and as with so many epidemiological studies in Dupuytren's disease the diagnosis depends on the observer and the criteria for diagnosis in the early case. In diabetics the distribution of Dupuytren's disease is atypical, generally less severe, often radially located involving the middle and ring finger with or without ulnar side involvement. It is usually mild, nodular in form and often does not require surgery (Hart and Hooper, 2005). Knuckle pads, nodules and tethering are much more common (Noble et al, 1984). Surgical complications such as haematoma, delayed healing, infection and skin sloughing may occur more frequently in diabetics.

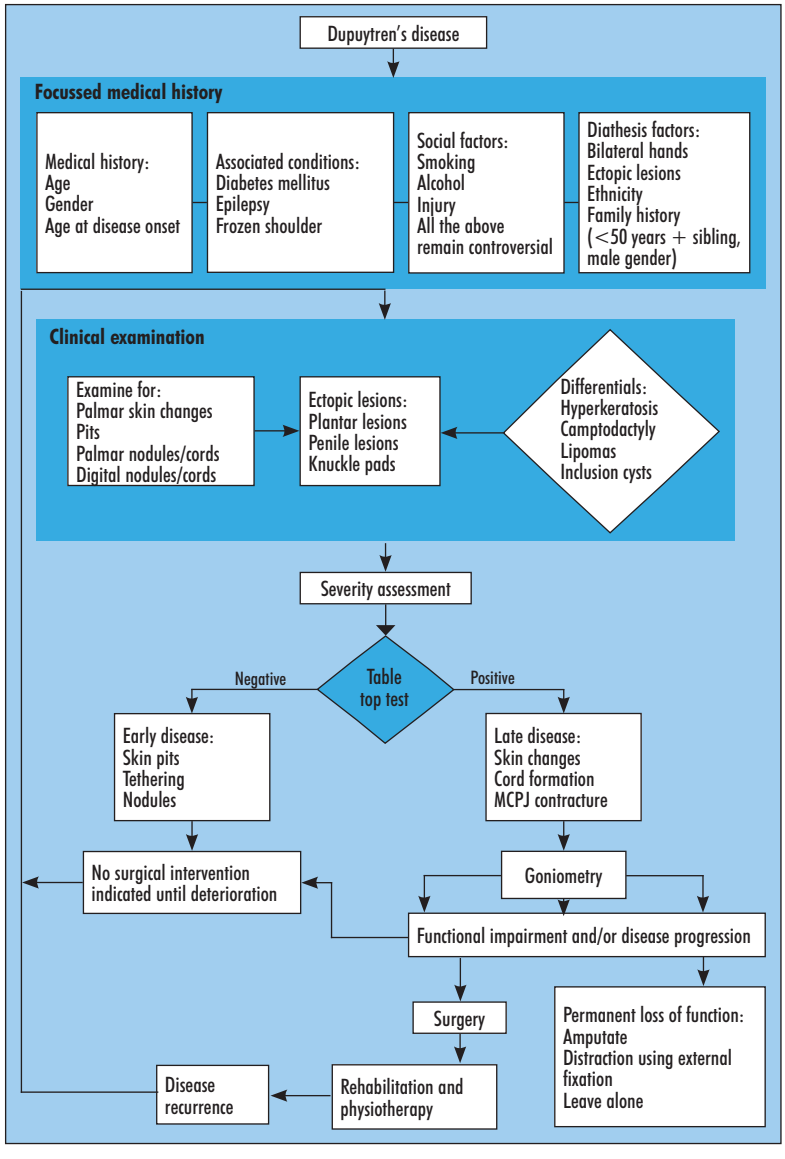
Epileptics tend to present with symmetrical, bilateral disease with a course that is not generally more severe or progressive than in non-epileptic patients (James, 1969). Incidence of Dupuytren's disease increases with duration of epilepsy, but is not thought to be linked to the severity of seizures or the disease course (Hurst and Badalamente, 1990). Epileptics have a higher incidence of plantar nodules and knuckle pads, suggesting either a predisposition of the individual to fibrosing conditions or that the fibrotic process has been induced by antiepileptic medication (Hart and Hooper, 2005). Different drugs have been implicated but especially phenytoin (Arafa et al, 1992).

Any of a number of upper limb maladies may occur in Dupuytren's disease more frequently than by chance, such as trigger finger or carpal tunnel syndrome. Dupuytren's disease is eight times more common in those patients with frozen shoulder (Smith et al, 2001), and frozen shoulder may follow surgery for Dupuytren's disease.

Manual labour and injury

There is no evidence that manual labour or manual recreations induce Dupuytren's disease (Hueston, 1990). A genetically predisposed individual may attribute the onset to a specific injury. Sixty five per cent of Dupuytren's disease cases are said to present within a year of injury, infection or surgery to a previously healthy hand and so there may be an association, but this is likely to be occurring in individuals who are genetically susceptible to Dupuytren's disease formation. Many patients become aware of palmar nodules after an injury but this may be explained by the injury drawing their attention to it (Leclercq, 2000). It is important to try and establish the point at which changes in the hand occurred, but this question is often met with imprecise recollections particularly as skin changes are an early and subtle feature of the disease.

Figure 1. Severity assessment and management plan pathway. MCPJ = metacarpalphalangeal joint.



Alcohol consumption and smoking

Although widely quoted in medical textbooks, associations of smoking, alcohol and Dupuytren's disease are controversial with different studies presenting conflicting results. Epidemiological studies have provided conflicting evidence in support of the relevance of alcohol and smoking as risk factors in Dupuytren's disease causation and progression (Noble et al, 1992; Gudmundsson et al, 2001; Godtfredsen et al, 2004). Clinically, it is worth establishing smoking and alcohol intake for general health evaluation but there is no evidence that cessation of either will modify the course of the disease. On current evidence, alcohol and smoking should not be thought of as actual 'risk' or 'causative' factors.

The clinical examination

The clinical examination should begin with the hands and it is a matter of judgment whether to extend this to examination of the feet for plantar nodules since the latter are unlikely to require intervention (*Figure 1*).

Sites of skin changes (pits and tethering), bands or nodules

Palmar skin creases, pits, tethering and nodules are the earliest manifestations of the disease. The degree of skin involvement is important to document and the simplest means of doing so is by photography. Skin pits are a reliable indicator of Dupuytren's disease as they are rarely associated with other conditions (Rayan, 1999) (*Figure 2*). A lump in the distal palm is a characteristic sign of Dupuytren's disease and other causes of lumps are rare (*Figure 3*) (Johnson, 1980).

Establishing a diagnosis of Dupuytren's disease in the early stages can be difficult, particularly when the only signs are thickened skin or prominent bands of fascia. Dupuytren's nodules are frequently located on the ulnar aspect of the palm, proximal or distal to the distal palmar crease. Clinicians should consider the likelihood of other diagnoses such as the common hyperkeratosis, callous formation or the much less common lipomas, inclusion cysts, flexor tendon pulley ruptures or rheumatoid arthritis (Lennox et al, 1993; Hughes et al, 2003). Distinguishing Dupuytren's disease from a callous can be difficult but an occupational history that reveals manual labour helps (*Figure 3*).

Knuckle pads, plantar lesions and Peyronie's disease

Knuckle pads (Garrod, 1893) form part of the ectopic manifestations of Dupuytren's disease and are typically located on the dorsal aspect of the PIPJs and on the

thumb (Hueston, 1984). They have been reported in up to 54% of patients (Hughes et al, 2003). They can vary in size from patient to patient. Patients are often unable to give a precise account of the onset of the pads as they do not cause functional problems (Mikkelson, 1977b). The presence of knuckle pads should alert the clinician to the possibility of Dupuytren's disease (*Figure 4*).

Plantar nodules (Ledderhose disease) appear often as painless lumps (rarely painful) in the arch of the sole along the medial border of the plantar fascia (*Figure 5*). Unlike palmar disease, the overlying skin is usually mobile and the lesions do not progress further to lead to contracture of the toes (Rayan, 1999). Patients frequently do not notice the condition.

Peyronie's disease is characterized by the presence of plaques on the dorsal aspect of the penis and is most common in middle age with a prevalence of 0.3–3% (Hauck and Weidner, 2001). Peyronie's disease occurs in Dupuytren's patients more frequently than by chance alone (Hinman, 1983). In the acute phase pain, flaccidity and penile curvature on erection are commonly reported; later, the disease may spontaneously regress in 50% of patients (Hauck and Weidner, 2001). The plaques are best palpated when the penis is flaccid. The majority of Dupuytren's patients do not have Peyronie's (or Ledderhose disease) and as there is no prophylactic treatment, it is a matter of clinical judgment and presence of positive family history of Dupuytren's disease whether to enquire into these statistically associated conditions, which may only serve to increase anxiety without contributing to the management of the hand problem (Wooldridge, 1988).

Digital nodules and cords

It is common for palmar involvement to progress into the digits but the disease can begin and remain in the digits (Rayan, 1999). Digital cords commonly occur on the palmar aspect of the proximal phalanx with extensive skin involvement. They produce a proximal traction on the PIPJ through attachments to the middle phalanx, leading to some degree of PIPJ flexion contracture. Clinicians should be mindful when examining the little finger that flexion caused by 'congenital' (it may not appear until teenage years) camptodactyly (flexion of the PIPJ) can also be mistaken for Dupuytren's disease. However, it is likely that camptodactyly will have been present during childhood and nodules will be absent.

Joint contracture

MCPJ deformity results from the contracture of a palmar cord. In the early phases of the disease, loss of the normal ability to passively hyperextend the MCPJ may be a clinical feature, later progressing into a permanent lack of extension, irrespective of the adjacent joint position (*Figure 6*).

The table top test devised by Hueston can be a useful means of establishing the progression of contracture. The test is positive when the patient is unable to lay their hands, palm down, flat on a table top. Patients can often identify this point in time although many will be unable to state when their disease commenced (Hueston, 1982).

It is important to identify which joints are affected because established PIPJ contracture confers poorer prognosis and functional outcomes are less favourable (Honner et al, 1971). There are also increased postoperative com-

Figure 2. Early skin pits.

Figure 3. The Hugh Johnson sign.

Figure 4. Knuckle pads.

Figure 5. Plantar lesions.

Figure 6. Palmar and digital nodules as well as palmar cord causing flexion contracture of metacarpalphalangeal joint of the middle finger.

plications associated with PIPJ contractures of 60° or more (Bulstrode et al, 2005). With longstanding contractures it is difficult to correct the joint stiffness which accompanies long-term joint immobility. Fixed flexion contracture of the PIPJ is the most problematic contracture to correct surgically (James, 1969).

There is little agreement on the timing of surgery in relation to the severity of contracture. MCPJ contractures can generally be corrected at any stage. By contrast the PIPJ contracture is more difficult to correct, Nevertheless there is evidence that operation when the joint is 30° contracted will leave some patients with increased contractures, whereas leaving the operation until the joint is severely flexed will almost always have a poorer outcome. Rather than basing the decision on an arbitrary degree of flexion it is better to base the decision to operate on the answers to two questions: first is there a functional disability and second is the contracture progressive? If there is PIPJ contracture, it is best to measure this and ideally to review the situation in 3 months' time.

Classification systems and measurement of contractures

Several classification systems have been described in the literature. That described by Tubiana (2000) incorporates both descriptions of individual digital flexion and the distribution of lesions in the hand, ensuring systematic examination and is outlined here.

Tubiana's system divides the hand into five segments. Each segment is allocated a sign that corresponds to the level of deformity. For each finger the range of deformity is measured using a goniometer and ranges from 0° (complete extension) to 200° (contracture of the digit to the palm). Such a system may have value in analysis of operative outcomes but it is best for the busy clinician to focus on the ranges of motion of the joints and in particular on PIPJ contracture in terms of deciding on the progression of the disease or timing of operation (*Figure 7*).

The presence of knuckle pads can distort goniometry measurements because the device fits imprecisely over the pads, leading to overestimation of contractures. In such instances, the goniometer should be placed on the lateral aspect of the digit, closely following the course of the phalanx. Once the measurements have been taken, the disease can be staged according to the degree of contracture for the joint in each affected digit.

Each ray (digit) is examined, starting with the thumb and each allocated a number that represents the stage depending on the measurement of the contractures determined by goniometry. Nodules without contracture are still assigned a value (0.5). Letters are used to further describe the characteristics of each ray, for example, P for palmar lesions, D for digital and H for hyperextension of the distal interphalangeal joint. For those instances where the extension deficit of the PIPJ exceeds 70°, the + sign follows D. There are further letters to describe the evaluation of the postoperative hand, details of which can be found elsewhere (Tubiana, 2000). The maximum score for each hand is 23, indicating that the four fingers and the thumb are contracted into the palm.

Conclusions

Dupuytren's disease is a common fibroproliferative disorder affecting primarily the hands of northern European Caucasians. Well-developed cases are generally straightforward to diagnose with the most frequently encountered presentation being that of a male in his fourth to sixth decade with bilateral disease. The presence of a cord usually leads to finger contracture, involving the MCPJ, and/or PIPJ. By contrast, distal interphalangeal joint contractures are rare.

Figure 7. Using a goniometer to measure contractures.

Taking a detailed history is important and can reveal information of prognostic value. Key factors to document include: age of onset, gender, ethnicity, symptoms, rate and course of disease progression, past medical history, positive family history, occupational history, injury and previous hand operations. The term 'diathesis' has been coined to include a grouping of features suggesting a poorer prognosis. Signs elicited in the clinical examination are: the presence of skin changes (pits and tethering), nodule/cord formation, knuckle pads and depending on clinical judgment the presence of ectopic lesions. Adverse prognosis, in terms of onset and extent of joint flexion contractures, is largely influenced by the presence of diathesis factors in a given individual.

The diagnosis of Dupuytren's disease is made on the characteristic clinical features, nodules in the lines of the digital rays in the palm or in the digits being pathognomic of the disease. The digits should be palpated to check for contracted fascial cords. These factors are important not only for establishing disease severity but also for counselling patients with Dupuytren's disease.

The only effective treatment is surgery but not all patients will benefit and there is a significant recurrence rate. The indications for operation are progression of contracture and functional disability and at this stage a hand surgical opinion (plastic or orthopaedic surgeon with a specialist interest in hand surgery) should be consulted at an early stage. **BJHM**

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KEY POINTS

- Most clinicians should be able to make an accurate diagnosis of Dupuytren's disease.
- Important factors in the history include age of disease onset, gender, ethnicity, bilateral disease, rate and course of disease progression, past medical history positive family history, occupational history, injury and previous operations.
- Important factors in the clinical examination are the presence of skin changes (pits and tethering), nodule or cord formation in the palm or digits and the presence of ectopic lesions (penile, plantar, knuckle pads and elsewhere).
- Proximal interphalangeal joint contractures are problematic to correct surgically and are associated with poorer prognosis.
- Surgery is indicated for functional disability or progressive contracture.
- Dupuytren diathesis factors can give an indication of prognosis although there is a great individual variation.