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## Dupuytren's Disease in an 8 Year-Old

A. R. Marsh and C. P. Kelly  
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### A New Technique for Reduction of Flexor Tendon Avulsions after Delayed Presentation

Dear Sir,

Delayed reduction of avulsed FDP tendons may be complicated by difficulty in passing the tendon through the A4 or, less often, the A2 pulley. The various techniques of doing this were comprehensively reviewed recently by [Kamath and Bhardwaj \(2007\)](#).

We present a simple solution to this problem. A 6Fr urinary catheter is passed in a retrograde direction down the empty tendon sheath to the free end of the tendon. If necessary, the catheter can be inflated to dilate pulleys, as described by [Jarvis et al. \(2002\)](#), and this has proven helpful in 25% of cases. A further catheter is then placed adjacent to the first and both are secured to the tip of the tendon with individual or combined core type sutures ([Figs 1a and b](#)). The tendon is then pulled distally through the pulleys by gentle traction on both catheters ([Fig 2](#)). We have used this technique successfully in eight cases.

a



b

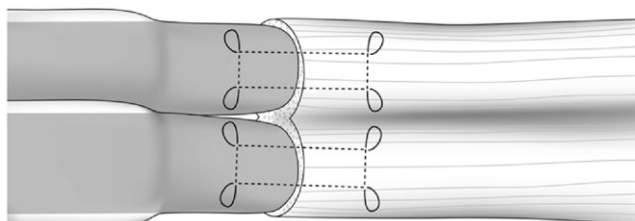


Fig 1 (a) Two 6Fr catheters are passed in a retrograde direction and (b) are sutured to the tip of the tendon.



Fig 2 Gentle traction on both catheters reduces the tendon back down the flexor sheath.

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### Ewing's Sarcoma of the Distal Phalanx of the Little Finger

Dear Sir,

A 13 year-old girl presented with pain and a diffusely tender, warm, cystic swelling of the tip of the right little finger of 1-month duration. The skin over the swelling was red and indurated. Plain X-rays showed a lytic lesion in the middle phalanx ([Fig 1](#)). Chest X-ray was normal. Fine needle aspiration cytology was inconclu-



Fig 1 Radiograph showed a permeative pattern of destruction, a soft tissue mass, sclerosis of the tip of distal phalanx and a lytic lesion in the head of middle phalanx and osteopenia. There was no periosteal reaction.

sive, so curettage biopsy was carried out. The histology reported features characteristic of Ewing's sarcoma. Stage AJCC grouping was IA (G1T1N0M0). Amputation at the metacarpophalangeal level was carried out to obtain 2 cm of clearance. Cyclophosphamide, Vincristine, Actinomycin and Doxorubicin were given post-operatively. Two years after surgery, there has been no local or distant recurrence.

Ewing's sarcoma most commonly occurs in the first or second decade of life, with the youngest recorded case occurring in the distal phalanx of a 5 month-old infant, although this tumour rarely occurs in the bones of the hand and feet. Euler et al. (1990) reviewed the literature up to 1990 and found 20 cases of Ewing's sarcoma of the hand. Three of the twenty cases reported were located in the distal phalanx. Later, Jones (1993) reported a case in the distal phalanx of the thumb and reviewed the literature and found a further nine cases involving the phalanges of the hand. Bernstein et al. (2006), reporting on 1426 patients from the European Intergroup Cooperative Ewing Sarcoma Study, recorded only 1% of primary Ewing's sarcomas in the hand.

Pain and swelling of the affected fingers were the most frequent complaints at diagnosis. Osteomyelitis may present a pattern similar to Ewing's sarcoma on plain X-ray. Diaphyseal location suggests a Ewing's sarcoma, as compared with the metaphyseal location more common in osteosarcoma. The European Intergroup Study identified only two histopathology parameters with prognostic significance, viz. topographical pattern and mitotic rate (Bernstein et al., 2006). Our patient had a predominant diffuse pattern, which carries a better prognosis. Management, preferably at a specialist centre by a multi-disciplinary team, has included both local control, by either surgery, radiation or a combination of these, and systemic chemotherapy. Chemotherapy has included cyclical combinations of vincristine, doxorubi-

cin, cyclophosphamide, etoposide, ifosfamide and, occasionally, actinomycin D. Topotecan in combination with cyclophosphamide produced responses in approximately 35% of patients with recurrent Ewing's sarcoma (Bernstein et al., 2006). Modern combinations of treatment are possibly curative due to the tumour's surgical accessibility and apparent restricted involvement of only tubular bone (Euler et al., 1990). With small soft tissue and bone sarcomas in the extremities, local control by surgery has been better than with primary radiotherapy (Euler et al., 1990; Jones, 1993). Therefore, for these distal phalangeal lesions, surgery would seem appropriate and adequate proximal amputation should always achieve wide clearance. Whether only marginal or wide resection is possible, even if the later is preferable, surgery should be performed.

Patients with hand lesions are recorded to have survived for more than 41 months and European Intergroup Study data showed a 68% overall 3-year survival rates in patients with distal extremity lesions. At initial diagnosis, approximately 25% of Ewing's sarcoma patients present with clinically detectable metastases in the lung and/or in bone and/or in bone marrow. Solitary or circumscribed bony metastases should be irradiated to doses of 40 to 50 Gy, in addition to local therapy to the primary site and Ewing's sarcoma-directed chemotherapy. Bilateral pulmonary irradiation has been reported to improve the outcome of patients with pulmonary disease. However, the survival rates of patients with multiple bony metastases are reported to be below 20% (Bernstein et al., 2006).

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## Re: Ossifying Lipoma of the Hand

Dear Sir,

A 54 year-old male labourer with a swelling of the right thenar eminence for more than 10 years presented when the slowly growing mass became progressively more painful during activity over 3 years. There was no history of trauma or surgery to



Fig 1 X-ray (anteroposterior view) showing ossification within the mass.

this part of the hand. Examination revealed a soft mass, measuring  $7 \times 4 \times 3$  cm, on the palmar aspect of the right thenar eminence which was slightly tender to the touch. He had no loss of sensation or limitation of the ranges of motion of the digits. X-rays showed increased soft tissue density over the first metacarpal bone with coarse, internal trabeculation (Fig 1). MRI showed that the tumour was independent of the first and second metacarpals and that the periosteum of these bones was normal. The lesion was hyperintense on T1- and T2-weighted imaging and there was internal low signal intensity content and decreased signal intensity on short TI inversion recovery (STIR) (Fig 2), suggesting a tumour with significant fat content. The pre-operative differential diagnosis favoured a lipoma with tumoral calcinosis or a soft tissue osteochondroma. Although soft tissue osteosarcoma could not be ruled out, the lesion did not contain the characteristic amorphous calcification and ossification. Synovial sarcoma was also a possibility, but unlikely because of the primary lipomatous content. Histopathological analysis of the resected tumour revealed mature adipose tissue with focal ossification, consistent with an ossifying lipoma, and no evidence of any malignancy. Hand use at

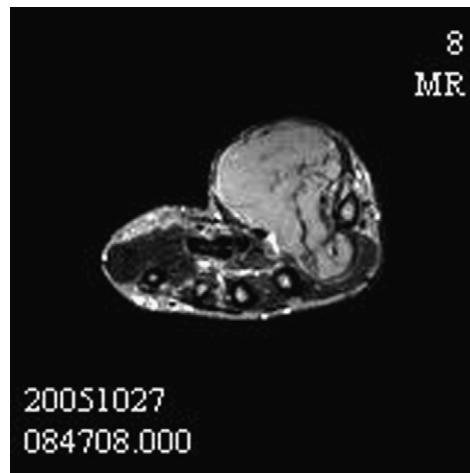


Fig 2 Decreased signal intensity on short TI inversion recovery (STIR) MRI matched adjacent subcutaneous tissue.

final follow-up 8 months after surgery was full and pain-free.

Ossifying lipomas arise independently of bone and are rare. To our knowledge, our patient represents the fourth reported case of an ossifying lipoma of the hand (Teoh et al., 2001). The lipomatous component always predominates, so diagnosis of the primary tissue type is not usually a problem (Obermann et al., 1999). Regardless of site, these tumours have a history strongly suggesting a benign tumor. Plain film X-rays are often the first, and, sometimes, the only, imaging study done pre-operatively, yielding a differential diagnosis of osteochondroma, lipoma with tumoral calcinosis and myositis ossificans, and diagnosis is typically made after histopathological analysis of the resected specimen (Obermann et al., 1999). MRI imaging may identify the lipomatous tissue basis of the tumour, so we recommend this investigation of lesions independent of bone that show internal ossification on plain X-ray as this may make a more definitive pre-operative diagnosis possible.

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## Re: Flexor Pollicis Longus Rupture 25 years after Silicone Lunate Replacement for Stage IV Kienböck's Disease.

Dear Sir,

A 50 year-old right handed manual worker, presented complaining of sudden onset of pain and weakness of the right thumb. Examination revealed a surgical scar on the dorsum of the wrist and absence of flexion of the interphalangeal joint of the thumb. X-rays showed features of Stage IV Kienböck's disease with a degrading silicone lunate replacement. Electrophysiological testing excluded anterior interosseous nerve palsy. Surgical exploration of the carpal tunnel identified attritional rupture of the flexor pollicis longus tendon by a prominent piece of the prosthesis, found at the scapholunate junction. This was excised and the tendon repaired by palmaris longus tendon grafting. Following therapy, a range of motion at the interphalangeal joint of 70° with MRC grade 4/5 power was achieved.

Intratendinous rupture of the flexor pollicis longus tendon is rare and most commonly seen in the setting of chronic rheumatoid disease, often as a result of scaphoid exostoses (O'Dwyer and Jefferiss, 1994). Zangger and Simmen (1993) reported a case secondary to extreme DISI deformity of the lunate in a rheumatoid wrist. Cases also occur in association with other distinct pathologies causing bone abnormalities, e.g. congenital carpal bone anomalies, Colles' fractures, Bennett's fractures, carpal fractures, carpal dislocations, scaphoid non-union and Kienböck's disease (Imai et al., 2004; McLain and Steyers, 1990). Yang et al. (1995) reported a case due to migration of a retained foreign body. Spontaneous rupture due to axial tendon loading alone has been reported in men aged 30–60 years during manual labour. Imai et al. (2004) described a case of spontaneous rupture which occurred in an elderly woman who was not involved in an occupational task but had spent a lifetime using the involved hand following a contralateral obstetric brachial plexus injury.

Long-term follow-ups of lunate silicone replacement for the treatment of Kienböck's disease have not previously identified this problem as a risk of surgery (Alexander et al., 1990; Egloff et al., 1993) although degradation of silicone lunate replacements is well recognised and has led to their use being discontinued.

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## Posterior Interosseous Nerve Palsy Caused by a Myxoma

Dear Sir,

A 67 year-old woman presented with inability to extend the metacarpophalangeal joints of all of the fingers of the right hand actively and a mass in the anterior aspect of her right elbow. She could extend her wrist actively and the full passive range of motion of the fingers was preserved. The elbow lesion was a soft tissue mass of approximately 3 cm in diameter which was well defined, firm and slightly tender. There was no sensory loss in the hand. Transverse T1-weighted MRI revealed a lesion with iso-signal intensity with normal muscle adjacent to the anterior surface of the radius and with an incomplete rim of fat (Fig 1A). Transverse T2-weighted MRI demonstrated a lesion with homogeneous high signal intensity, in which there were low signal striae (Fig 1B). Electrodiagnostic studies showed denervation of the extensor digitorum communis, identifying a posterior interosseous nerve palsy secondary to tumour. At surgery, the radial nerve was identified at the elbow and traced distally through an anterior approach. The posterior interosseous nerve was entrapped and compressed between the arcade of Frohse and the tumour, which was located deep to the nerve (Fig 1C). The posterior interosseous nerve was dissected off the mass under direct vision. The tumour, which was enclosed by a white capsule and attached to the neck of the radius

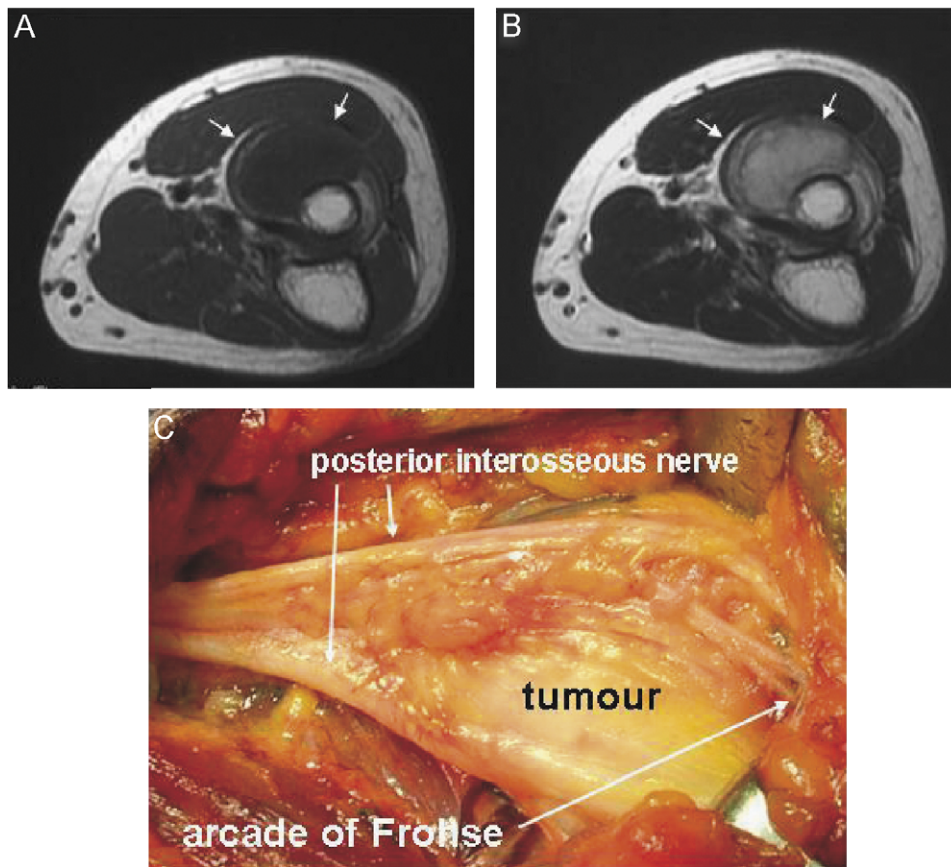


Fig 1 MRI findings (A) and (B) arrows indicate the tumour and (C) intraoperative finding. (A) Transverse T1-weighted MRI showing a lesion with an iso-signal intensity and normal muscle attached to the anterior surface of the radius. (B) Transverse T2-weighted MRI demonstrating the lesion as a homogeneous high signal intensity area in which there are low signal striae. (C) The posterior interosseous nerve is entrapped and compressed between the arcade of Frohse and the tumour, which is located deep to the nerve.

and the supinator muscle, was then excised. Histological examination reported a myxoma. Two years after surgery, the patient was asymptomatic and there was no recurrence of the lesion.

Non-traumatic posterior interosseous nerve palsy can be caused by various space occupying lesions around the arcade of Frohse, including ganglion, lipoma, and bicipital bursa (Hashizume et al., 1996). Myxoma has been reported as a rare cause (Valer et al., 1993). The MRI findings of myxoma are similar to those of a cystic lesion, such as a ganglion or bursa, except that T2-weighted MRI demonstrates a lesion with homogeneous high signal intensity with low signal striae, suggesting septa in a tumour (Murphey et al., 2002).

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#### Osteonecrosis and Pathological Fracture of the Metacarpal Shaft caused by Silk Sutures

Dear Sir,

A 32 year-old woman presented with right palmar pain following a jolt when she was riding a bicycle. Four



Fig. 1 X-ray 6 weeks after the first operation showing synchronous bone healing and absorption.



Fig. 2 X-ray after the second injury showed bone damage and a pathological fracture.

months previously, she had undergone open reduction and fixation of a closed oblique fracture of the ring metacarpal with silk sutures elsewhere. On confirmation of bone healing on X-ray (Fig. 1) 6 weeks after surgery, she had resumed her normal daily activities without problems. X-ray on this occasion showed bone damage and a pathological fracture of the same bone (Fig. 2). At surgery, a mass of silk sutures, liquefaction and granulation was found around the fracture site. The sclerotic fracture ends were excised and the defect internally fixed with a titanium miniplate over iliac bone autograft. Histology showed widened and disordered bone trabeculae in the bone ends and chronic inflammatory changes around the silk sutures. The miniplate was removed 8 months after surgery, and there was no residual pain or dysfunction at 2-year follow-up.

We do not think the osteonecrosis in this patient was initiated by an inadequate blood supply as bone healing at 6 weeks indicated that this was adequate and, theoretically, the blood supply would have improved, not deteriorated, with time if no other event had

supervened. Therefore, a delayed foreign body reaction to the silk sutures probably played the key role in the pathogenesis of the subsequent osteonecrosis. Sutures, acting as foreign bodies, probably produce a foreign body reaction in every surgical patient, but they only rarely cause trouble (Warne, 2004). In this case, an excessive foreign body reaction, resulting from the use of multiple sutures, only came to light following the second, minor trauma.

The use of sutures of any kind for fracture fixation is not common because they cannot provide reliable stabilisation, as witness the need for a large number of silk sutures in this case. We have only found two reports in the recent literature about silk sutures being used for bone fixation (Jordon and Burch, 1976; Kabardin, 1979), although they may have been used more commonly in earlier days. However, sutures, now mostly absorbable, are sometimes used as an adjunct to fixation in children to avoid use of wires etc and a need for secondary removal, necessitating further general anaesthesia.



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## A Case of Brachial Amyotrophy Mimicking Rupture of the Flexor Digitorum Profundus Tendon

Dear Sir,

A 58 year-old lady with no medical history of note presented with a 3-week history of spontaneous inability to actively flex the distal interphalangeal joint of her right index finger, associated with occasional intermittent aching in her forearm. Neurological examination was otherwise normal. An ultrasound scan suggested avulsion of the insertion of the flexor digitorum profundus tendon. Exploration of her index finger, however, revealed an intact, although rather lax, flexor digitorum profundus tendon. Carpal tunnel decompression revealed oedematous tissue only but more proximal exploration revealed an atrophic looking muscle belly. Despite intensive hand physiotherapy, she regained no active movement at the index finger distal interphalangeal joint. Sensory loss within the median nerve territory (thumb and index finger) subsequently developed. No neurological deficits of the radial and ulnar nerves became evident. Histology of the thickened flexor digitorum profundus tenosynovium and carpal tunnel synovium showed non-specific reactive changes only. An isolated anterior interosseous nerve palsy, only affecting flexor digitorum profundus to the index finger, was suspected. However, neurophysiological studies indicated a diagnosis of brachial amyotrophy with reduced median nerve sensory potentials both in the hand and forearm (mixed nerve), absent sensory potentials in the lateral cutaneous nerve of the forearm, as well as patchy changes of chronic denervation of the flexor digitorum profundus and flexor carpi radialis muscles.

Brachial amyotrophy is a rare, but under diagnosed and potentially debilitating, neurological syndrome, also known as Parsonage–Turner syndrome, neuralgic amyotrophy, brachial plexitis, shoulder girdle neuritis and idiopathic brachial plexopathy. The estimated incidence

is 1 to 2 per 100,000, with a male to female ratio of 2 to 3:1, and with a usual age of onset in the third to sixth decades (Rubin, 2001). Although the exact aetiology and pathophysiology are unclear, there is often an antecedent prodromal illness or insult (Favero et al., 1987; Tsairis et al., 1972). A hereditary form of this disorder – hereditary neuralgic amyotrophy – is also recognised (Dunn et al., 1978). The classical presentation is with sudden onset of (severe) pain, followed by gradual weakness, patchy, and often multi-focal, sensory loss and atrophy (Rubin, 2001; Tsairis et al., 1972). Symptoms most commonly occur in the proximal upper limb musculature, but may involve any distribution of the brachial plexus, or present as a mononeuropathy or mononeuritis multiplex type picture – including isolated involvement of the anterior interosseous or median nerves (Rubin, 2001). Some patients, as in our case, present with minimal, or absent, pain and relatively sudden onset of weakness, or, even, complete paralysis (Rubin, 2001). Neurophysiological assessment is key in identifying brachial amyotrophy and distinguishing it from other conditions. It is characterised by absent, or reduced sensory responses ± abnormal motor potentials (reduced compound muscle action potentials, fibrillation potentials and motor unit potential abnormalities) (Rubin, 2001; Seror, 2004). Radiological studies and laboratory tests are often helpful only in excluding other pathologies (Rubin, 2001). Treatment is supportive, including steroids, analgesia and physiotherapy (Rubin, 2001). The prognosis is good with most (approximately 90% of cases) making a near full recovery over 3 to 4 years (Tsairis et al., 1972).

Favero et al. (1987) found that 64% of patients with brachial amyotrophy had an orthopaedic consultation during their acute illness. As it affects the shoulder girdle most commonly, its' more common differential diagnoses include rotator cuff injury, myopathies, acute calcific tendonitis and adhesive capsulitis. However, it may also mimic pathology in the hand or forearm, as in this case. Our recommendation is that, when faced with an uncharacteristic history of symptoms or a triad of upper limb pain, muscular weakness and atrophy, or sensory loss, neurophysiological tests are performed to avoid missing a rare diagnosis which is best treated expectantly.

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### An Alternative 'Viewing Box' for Hand X-rays

Dear Sir,

Accurate interpretation of hard-copy X-rays depends on several factors, including the consistency of spatial illumination brightness of viewing boxes (Alter et al., 1982). To maximise visual acuity, it is important that the retinal cones receive an incident luminance of  $100 \text{ cd m}^{-2}$  (Quality Criteria, 1990). X-ray viewing boxes of adequate quality are not always available (McCarthy and Brennan, 2003).

The author would like to suggest an alternative light projecting media for viewing X-rays, namely, the personnel computer plasma screen monitor. These are readily available and have a uniform brightness level of  $200 \text{ cd m}^{-2}$  across the screen. A computer screen is adequate to display small films such as hand X-rays (Fig 1).

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Fig 1 A photograph of a fifth metacarpal neck fracture X-ray placed against a monitor screen (opened against a white background of a blank Word Document page).

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### Re: Intra-neural Perineurioma of the Median Nerve. A Rare Cause of Carpal Tunnel Syndrome

Dear Sir,

A 42 year-old right handed woman was seen because of progressive pain in the right wrist of 2 years duration, including pain at night that routinely interrupted her sleep. She also suffered from numbness in the distribution of the right median nerve and wasting of the thenar muscles. She had no known history of trauma. On examination, there was a  $2 \times 2 \text{ cm}$ , elliptical, firm, subcutaneous mass on the flexor aspect of the wrist. Sensory examination identified paraesthesiae in the distribution of the median nerve with a positive Phalen's test and Tinel's sign. MRI of the right wrist showed a

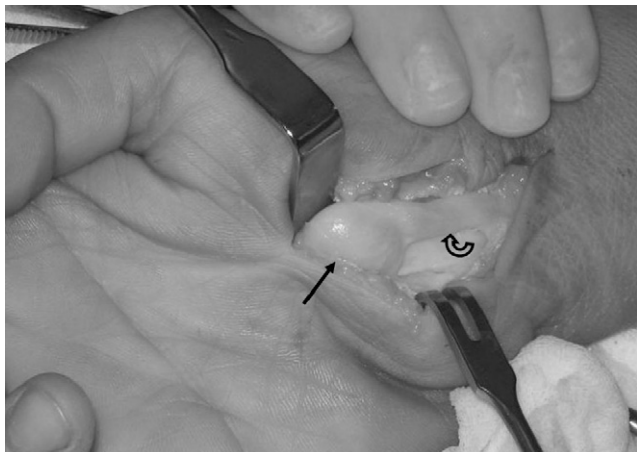


Fig 1 Intraoperative photograph of the tumour (straight arrow) in continuity with the normal median nerve (curved arrow).

heterogeneous mass which appeared well circumscribed. The patient underwent release of the carpal tunnel and incisional biopsy of the mass. The mass extending for approximately 2 cm within the median nerve just proximal to the take-off of the motor branch (Fig 1). There were no postoperative complications and there has been sustained regression of the symptoms of carpal tunnel syndrome over the 16 months since surgery. During this period, there has been no clinical or radiological evidence of increase in size of the tumour.

Perineuriomas present as focal, intraneural, peripheral nerve, or isolated, soft tissue lesions. Intraneural perineuriomas, generally, present as a slowly progressive, painless, focal motor neuropathy. They are characterised histologically by onion-bulb-shaped whorls (Imaginarijda et al., 1964). The aetiology of these lesions is controversial, with some authors asserting that this is a reactive process to trauma and others proposing that this is a true neoplasm (Emory et al., 1995). In the recent literature, the lesion continues to be referred to interchangeably as a perineurioma and as a localised hypertrophic mononeuropathy (Gruen et al., 1998).

According to Jazayeri et al. (2000), there is no consensus as to the correct management of these lesions. However, these authors advocated excision of the lesion and interpositional nerve grafting, although return of sensory nerve function was not seen in their case. In our patient, we confirmed the diagnosis by incision biopsy and adopted a policy of conservative management. At this point in time, there appears to be no evidence to support either of these two treatment options over the other from an oncological point of view.

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## Dupuytren's Disease in an 8 Year-Old

Dear Sir,

An 8 year-old boy was referred by his family doctor with a progressive contracture of the right little finger over a 12-month period. On examination, there was a flexion contracture of 40° at the proximal interphalangeal joint with hyperextension of 20° at the distal interphalangeal joint (Fig 1). A fasciectomy was performed with a full correction of the contracture. At



Fig 1 Contracture of the little finger due to an abductor band in an 8 year-old.

the time of surgery, it was noted that the cord arose from the abductor digiti minimi and inserted into the flexor sheath and the ulnar side of the proximal interphalangeal joint. The cord was excised and sent for histological examination, which demonstrated hypercellular fibromatosis, typical of Dupuytren's disease. The patient was subsequently treated by mobilisation and an extension night splint. At 1 year, there was no recurrence or extension of disease and full finger function.

Dupuytren's disease in children is extremely rare. One study identified 108 cases of fibromatoses in children in a 25-year period, from which there were only three cases of Dupuytren's disease (Cheryl et al., 1991). Several studies have, however, identified certain features in respect of the appearance of this disease in childhood (Bebbington and Savage, 2005; Mandalia and Lowdon, 2003; Rhomberg et al., 2002; Urban et al., 1996). The literature includes 13 cases of suspected Dupuytren's disease under the age of 13 without histological confirmation. Since 1832, there have only been seven published cases of histologically proven Dupuytren's disease in children under 10 years of age. All reported cases have been treated surgically. Four underwent fasciectomy. The surgical technique was not disclosed in the other three cases. In one case, there was recurrence of the disease, which subsequently required dermofasciectomy.

Dupuytren's disease should be considered in the differential diagnosis of a child with a finger contracture. The natural history of Dupuytren's disease in children is unknown and, therefore, the possibility of recurrence and extension of disease should be explained to the family.

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## Closed Rupture of a Flexor Tendon of Unknown Aetiology

Dear Sir,

Closed ruptures of flexor tendons have been described in association with several distinct pathologies (Boyce et al., 1960; Papp et al., 2006). Here, we report a case of closed rupture of a flexor digitorum profundus (FDP) tendon with the intact flexor digitorum sublimus (FDS) tendon having a distinct, and visible, pathology, which was impossible to classify into the established pathologies normally associated with this condition.

A 61-year-old, right hand dominant housewife was referred to our clinic with inability to flex the distal interphalangeal (DIP) joint of her left index finger. Four weeks prior to presentation, she tried to lift a plastic supermarket bag of approximately 3Kg in weight. After hearing a painless snap, the patient lost the ability to flex the DIP joint of her left index finger. The patient denied any previous history of trauma or pain. No local heat or tenderness was noted. X-rays confirmed absence of bony erosions or osteophytes.

In contrast to the expected intra-operative findings of a ruptured FDP tendon alone, the intact FDS tendon displayed striking macroscopic and microscopic findings. On this tendon were intra-tendinous cysts with three surface enlargements, the proximal completely covered and the distal two partially covered by proliferating synovium (Fig. 1). Incision of a cyst exposed fragmented tendon and yellowish mucinous fluid. Histologically, the cyst wall was invested by synovial lining cells but lympho-plasmocytic infiltration was not found, as in active rheumatoid arthritis (Fig. 2). Fibrinoid necrosis affected the whole width of several collagen bundles, resulting in disruption of the tendon. The disrupted collagen fragments were encircled by histocytes, including multinucleated giant cells, composing the central component of the granulomas. Granulomas were also formed around incomplete



Fig. 1 Three cystic enlargements covered by hypertrophied synovium. Through the opened transparent wall of a cyst, 'chopped noodle-like' white pieces are visible (arrow).



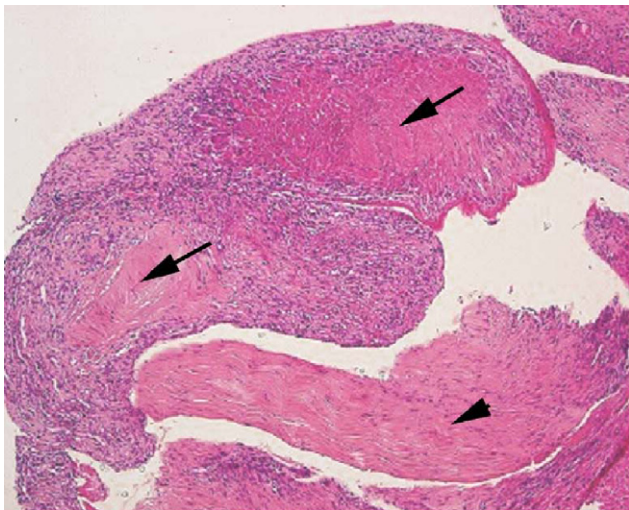


Fig. 2 Tendinous collagen fibers undergoing necrotic changes. Two focal lesions are seen (arrows). Undisrupted, normal collagen bundles are also seen adjacent to the damaged bundles (arrowhead). (Haematoxylin and Eosin,  $\times 80$  magnification).

necrosis, i.e., necrobiosis, of the tendon. Alcian blue staining detected abundant mucoid matrix. Collectively, the histopathological characteristics of the present lesion were summarised as multiple necrobiotic granulomas with mucoid degeneration and cystic changes.

An interposition tendon grafting using palmaris longus tendon was performed and the patient recovered full ROM of the index finger after two months of hand therapy. The patient was followed up for 3 years to rule out involvement by rheumatoid arthritis because the post-operative blood chemistry detected slightly elevated rheumatoid factor. Positive CRP value, anti-nucleic acid antibodies and anti-DNA antibodies were not detected throughout the follow up period. During the 3-year follow up the patient has not developed RA, nor has she suffered from other collagen diseases or tuberculosis.

Neither the histological nor the clinical features fulfilled the diagnostic criteria of rheumatoid arthritis. The wide-spread multifocal granulomas with giant cells and histiocytes, suggesting involvement of a foreign body reaction or mycobacterial infection, were not confirmed by detection of fungi, tuberculous organisms or other infectious organisms postoperatively. The pathology occurring to this flexor tendon remains unclear.

## APPENDIX

The authors will be happy to provide further slides to anyone who wishes to look at them.

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## Spontaneous Subperineural Haemorrhage Causing Acute Carpal Tunnel Syndrome

Dear Sir,

A right hand dominant, 63 year old housewife presented to us with intractable right wrist pain of two days duration. The pain was of sudden onset, throbbing in nature, exacerbated by wrist movement and associated with numbness along the right thumb and middle finger. There was no weakness of the hand or fingers. She had sustained a right distal radial shaft fracture, which had been treated with open plating using a volar approach, 12 weeks previously. The surgical exposure did not extend to the flexor retinaculum. She had been attending physiotherapy for wrist mobilisation. On presentation, there was tenderness over the volar aspect of her right wrist with a markedly positive Phalen's test and Tinel's sign. She also had sensory loss in the median nerve distribution but no motor deficit. Laboratory

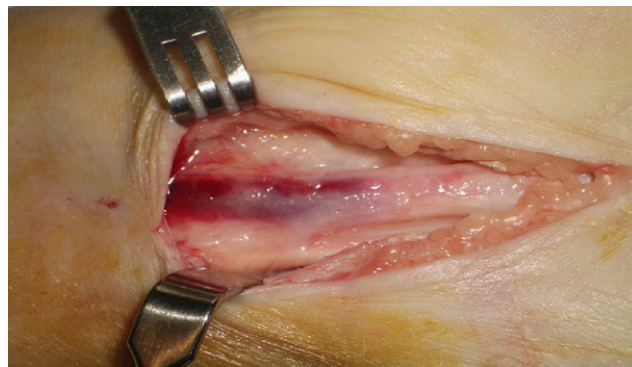


Fig. 1 Subperineural haemorrhage of the right median nerve found intraoperatively.



investigations, including coagulation profile, were within normal limits. A diagnosis of acute carpal tunnel syndrome was made and exploration carried out. Subperineural haemorrhage involving the median nerve throughout its course in the carpal tunnel was found (Fig. 1) and decompression of the carpal tunnel carried out. The investing layers of the nerve were not incised. There was immediate postoperative improvement of symptoms, which had almost completely disappeared 4 weeks later.

There are four previous reports of haemorrhage causing acute carpal tunnel syndrome in patients with a normal bleeding profile. [Watson-Jones \(1949\)](#) and [Faithfull and Wallace \(1987\)](#) each reported a case of haemorrhagic carpal tunnel syndrome following trauma. Both [Hayden \(1946\)](#) and [Mandal et al. \(2004\)](#) described spontaneous intraneural haemorrhage in the median nerve leading to acute carpal tunnel syndrome, with no immediate history of trauma. Whether the distal radius fracture had any bearing in our case is impossible to say, although a time difference of 12 weeks makes this less likely.

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