Pachydermodactyly: A case report
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Pachydermodactyly is a rare benign fibrous condition of the hand related to repeated mechanical trauma to the skin. The recommended management is nonsurgical. Most of the literature regarding the topic appears in the nonsurgical literature and, hence, the condition is not familiar to hand surgeons. In the authors’ hand centre, one patient with pachydermodactyly was misdiagnosed with a fibrous tumour and underwent surgical excision. The correct diagnosis was made retrospectively from the histology and on asking the patient about habitual manipulation of the finger. The authors present the case and review the literature to increase the awareness of hand surgeons to this condition.

Key Words: Fibromatosis; Fibrous tumour; Hand; Pachydermodactyly

In our hand centre, one patient with pachydermodactyly was misdiagnosed with a fibrous tumour and underwent surgical excision. The correct diagnosis was made retrospectively from the histology and on specifically asking the patient about habitual manipulation of the finger. We present our case and review the literature to increase the awareness of hand surgeons to this condition.

CASE PRESENTATION

A 20-year old man presented with a diffuse fibrous mass of the dorsal aspect of the proximal phalanx and proximal interphalangeal joint of the left ring finger. There was no pain or tenderness, and the range of motion was full. The differential diagnosis included juvenile rheumatoid arthritis, Dupuytren knuckle pads, idiopathic swelling around the proximal interphalangeal joint, and a fibrous tumour. Hence, history, clinical assessment and investigations were focused toward these diagnoses. There was no history of joint trauma. There were no symptoms or signs suggestive of juvenile arthritis or Dupuytren disease. Family history was negative for the latter two conditions. X-ray of the hand, complete blood count, erythrocyte sedimentation rate and C-reactive protein levels were all normal. The autoimmune screen (including rheumatoid factor and antinuclear antibody factor) was negative.

Under digital block, a longitudinal dorsal incision was made. A diffuse fibrous mass was found to involve the deep dermis and subcutaneous tissue, and it was adherent to the extensor mechanism. Complete excision was not attempted; instead, the fibrous mass was marginally excised making sure to maintain sufficient thickness of the raised skin flaps (to ensure skin viability) and to preserve the extensor mechanism (Figure 1A). Histopathological examination showed fibroblast proliferation infiltrating in between the eccrine glands (Figure 2). Immune staining was negative for a smooth muscle actin, desmin, CD34,
cytokeratin and S-100 protein. The histopathological diagnosis was benign fibromatosis, suggestive of pachydermodactyly. On asking the patient again, there was a positive history of habitual ‘chewing’ of the same area; the patient was advised to discontinue such behaviour. The patient was last seen one year later and had recurrence at radial side of the proximal interphalangeal joint (Figure 1B). The patient admitted that he occasionally chewed the finger at the same area of recurrence. The patient was offered local triamcinolone injections but he refused and did return for further follow-up.

**DISCUSSION**

The term ‘pachydermodactyly’ was coined by Verbov (6) in 1975 (‘pachy’ is thick, ‘dermo’ is skin and ‘dactyly’ is finger in Greek). The most accepted theory of pathogenesis is repeated mechanical injury of the skin. Hence, the condition is usually seen in patients with compulsive habits of hand manipulation and anxiety disorders (2). However, pachydermodactyly is also known to occur in certain workers with repetitive trauma of the fingers such as poultry processing workers (7). Our patient had a history of ‘chewing’ on the finger; this form of pachydermodactyly is also known as ‘chewing pads’ in the dermatology literature (8).

The most common presentation of pachydermodactyly is bilateral hand swelling around the proximal interphalangeal joints in multiple fingers; this is known as the ‘classic’ type (1-5). Other types have been described, including ‘monopachydermodactyly’ (affecting one finger, as seen in our patient) and the ‘transgradients’ type, which affects the skin around the metacarpophalangeal joint (9).

The classic type, with multiple joint involvement, is usually confused with autoimmune arthritis (4). However, plain x-rays are normal and clinical examination does not reveal any evidence of autoimmune disease. Furthermore, the autoimmune blood screen is negative. The monodactyly type may be confused with fibrous tumours/conditions of the hand including Dupuytren knuckle pads.

Benign fibrous swellings of the hand includes a long list of conditions and tumours (10,11). All of these fibrous swellings have a common histological feature of spindle cell proliferation. Hence, the histopathological diagnosis is based on characteristic histological features and positivity to immune stains. Table 1 summarizes the specific features of several benign fibrotic conditions/tumours of the hand including pachydermodactyly. Spindle cells may be seen with anaplastic epithelial tumours as well as in tumours of neural origin. Hence, cytokeratin and S-100 protein immune stains are routinely performed in spindle cell tumours to rule out tumours of epithelial and neural origin, respectively. Positivity to smooth muscle actin indicates the presence of myofibroblasts; this is a feature of several tumours/conditions, as shown in Table 1. Positivity to desmin is characteristic of desmoid tumours, while positivity to CD34 is characteristic of dermatofibrosarcoma protuberance and superficial acral fibromyxoma. Negativity to above-mentioned immune stains is observed in giant cell tumours of tendon sheath, calcifying aponeurotic fibroma, fibrous hamartoma of infancy and pachydermodactyly. Soo and Sung (1) reviewed the literature and found two characteristic histological features in pachydermodactyly: the epidermis has acanthosis and hyperkeratosis (a sign of mechanical trauma), and the deep dermis/subcutaneous tissues show entrapment of the eccrine sweat glands by the fibroblast proliferation (Figure 2).

Nonsurgical management is initially recommended for pachydermodactyly. The most important part of management is to stop repeated mechanical trauma to the skin (1-4). Intralesional injection of triamcinolone (which enhances collagen degradation) and systemic (given orally) collagen-synthesis inhibitors have also been tried with moderate success (5,12). Surgical intervention is only indicated for patients who do not respond to removal of the irritating behaviour and are seeking a cosmetic improvement.

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REFERENCES