Dupuytren's disease of the foot in children: a report of three cases

G. S. RAO and P. K. LUTHRA

Departments of Plastic Surgery, Shotley Bridge General Hospital, Consett, Co. Durham and Withington Hospital, Manchester

Summary—Fibromatosis of the plantar fascia is a much less common occurrence than that of the palmar fascia and it is rarer still in children.

It is essential to differentiate this entity from fibrosarcoma. In spite of its high incidence of local recurrence and the histological presence of mitotic figures, conservative resection is the treatment of choice. Recurrence following dermofasciectomy and skin grafting is less common than simple excision of the nodules in the fascia. Amputation should never be the treatment.

Fibromatosis of the plantar fascia is not as common as that of palmar fascia. It was described by Ledderhose in 1897 as a distinct entity (Aviles et al., 1971). The degree of similarity between the palmar nodules described by Baron Dupuytren and the plantar proliferative masses described by Ledderhose has led to the condition being accepted as “Dupuytren's disease of the plantar fascia” (Cavolo and Sherwood, 1982). It is a benign disorder involving the plantar aponeurosis and eventually invading the overlying skin. The age and sex distribution of plantar fibromatosis is similar to that of Dupuytren's contracture in the hand (Snyder, 1980). Thus it has a preponderance in males over females. A review of 102 cases by Pickren et al. (1951) elicited an age range from 5 to 60 years.

Plantar fibromatosis may present as either a unilateral or a bilateral condition. The lesion usually presents as a nodular swelling in the middle of the medial longitudinal arch of the foot near the base of the first metatarsal. It may sometimes be seen at the bases of the toes or along the metatarsal heads. Pain is not a common feature but it may cause discomfort on walking. As in Dupuytren's contracture of the hand, the thickened plantar fascia involves the overlying skin, thus presenting as a dimpled or puckered skin lesion. Unlike its counterpart in the palmar fascia, contractures of the toes are rare in plantar fibromatosis. According to Lettin (1964), if a contracture is evident it will be a flexion deformity of the second toe.

Case reports

Case 1

An 8-year-old boy first presented to an orthopaedic surgeon with a lump on the instep of his left foot. This lump had been first noticed 4 months prior to admission. On examination he had a firm, discrete and tender lump on the instep of his left foot. An excision biopsy of this nodule was carried out. Histological examination of this tissue showed features of fibromatosis.

About 3 months later he developed a recurrence of the nodule and was referred to the Plastic Surgery Unit. He now presented with a gradually enlarging lump which was painful on walking. On examination he had two firm nodular lumps deep to and attached to the plantar skin of the foot, lying in the instep region and together measuring 2 x 3 cm.

A formal dermofasciectomy of the affected area was carried out and the resulting defect covered by a split skin graft. Histology again showed it to be a benign plantar fibromatosis. At 2 years follow-up the graft remains well healed and there are no signs of recurrence (Fig. 1). Skin grafting has not interfered with walking and weight-bearing.

Case 2

A 5-year-old boy first presented with a painless lump on the sole of his left foot which had gradually enlarged over the last 3 years. There was a strong family history of Dupuytren's contracture on both paternal and maternal sides.

The nodule was excised along with the overlying skin as an ellipse and the skin was closed directly. Histology showed the presence of nodules of proliferative fibrous tissue separated by thick collagen bands, the picture
being consistent with the diagnosis of plantar fibromatosis.

Seven months later he was readmitted with a recurrence of the nodule and on this occasion it was tender. Once again a local excision and direct closure was carried out.

Ten months later he presented with a second recurrence. On this third admission he was referred to the Plastic Surgery Services where a fairly wide dermofasciectomy was carried out and the area resurfaced with a skin graft. Histological examination showed features of the "proliferative" phase of plantar fibromatosis.

At 18 months postoperative follow-up the graft stays well healed and there are no signs of recurrence of the lesion. Skin grafting has not interfered with walking or weight-bearing.

Case 3

An 8-year-old girl first presented with a lump on the sole of her left foot. An excision and direct closure was carried out. At 11 years of age she presented with a recurrence of the nodule, for which further local excision of the lump and overlying skin along with skin grafting was done. She presented for the third time with a recurrence at the age of 16. On this occasion a dermofasciectomy and skin grafting were carried out. Histology confirmed the lesion to be plantar fibromatosis. At 6 months follow-up she stays well healed and there are no signs of recurrence.

Discussion

It is important to differentiate this condition from fibrosarcoma but the histology should be conclusive. Other conditions with which it may be confused are hypertrophic scarring, callosities, benign fibroma, chondroma and exostosis (Pickren et al., 1951).

The gross pathology shows thickening of plantar fascia by lobulated, firm and irregular masses. The cut section shows grey fibrous tissue arranged in fine bands and whorls, or sharply circumscribed zones of dull grey-yellowish tissue (Meyerding and Shellito, 1948; Pickren et al., 1951). The nodular masses may coalesce to form a single mass and become adherent to the overlying skin, but do not involve the other adjacent structures (Allen et al., 1955). The microscopic appearance of this lesion is quite distinct. The nodules consist of cellular islands of proliferating fibroblasts with an acellular collagenous aponeurotic background. There is a characteristic nodular multicentric pattern of growth. The fibroblasts are uniform but occasionally show a variation in shape, size and staining reactions and may show some mitotic figures. A constant finding within the proliferating nodule is the
formation of blood vessels lined with hobnail-like endothelial cells and surrounded by plump fibroblasts (Allen et al., 1955). Pickren et al. (1951) stated that the histological picture observed in some cases of plantar fibromatosis could be interpreted as fibrosarcoma, if it were found in other parts of the body, but that the process in the foot was clinically benign.

Surgical interest in plantar fibromatosis relates to frequent local recurrences following simple excision, and also the not infrequent misinterpretation of the tumour as sarcoma. In spite of the presence of mitotic figures in a recurrent plantar tumour, conservative therapy is still the usual treatment of choice. Plantar fibromatosis has not been found to metastasise to other areas. Three cases of amputation have now been reported as a result of failing to recognise the benign nature of plantar fibromatosis (Aviles et al., 1971).

The same authors, in a review of 22 patients over a period of 20 years, have reported 57% of local recurrence following simple excision alone in contrast to 8% local recurrence following dermofasciectomoy and skin graft. This corresponds to the 53% recurrence following simple excision, as reported by Allen et al. (1955). Since incomplete removal of plantar fascia associated with plantar fibromatosis is followed by a high recurrence rate, wide excision is preferred in the treatment of the disease. Because of the common site of occurrence over the middle of the medial arch of the foot, skin grafting in that region usually does not interfere with walking and weight-bearing.

The surgeon may choose to leave a plantar nodule alone when he is reasonably certain that this is part of Dupuytren's disease. However, such a diagnosis is hard to establish in a child and faced with a recurring plantar nodule in childhood, excision and skin grafting seems the most appropriate course of action.

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References

The Authors
G. Sambasiva Rao, MS, FRCSEd, DNB (Plas. Surg.), Registrar in Plastic Surgery, Shotley Bridge General Hospital, Consett, Co. Durham DH8 0NB; formerly SHO, Newcastle General Hospital, Newcastle upon Tyne.
P. K. Luthra, MS, FRCS, Senior Registrar in Plastic Surgery, Withington Hospital, West Didsbury, Manchester; formerly locum Senior Registrar, Leeds.

Requests for reprints to Mr G. S. Rao.

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