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Letters to the Editor

Mr. and Mrs. Reader,

Dear Sir,

Plantar fibromatosis is a rare, benign, hyperprolifera-
tive process of the plantar aponeurosis first described as
a distinct disease by Ledderhose in 1897 [1]. It is charac-
terized by local proliferation of abnormal fibrous tissue in
the plantar fascia. The plantar aponeurosis is gradually
form thickened fascia and nodules that range in size from
0.5 to 3 cm. Subsequently, invasion of the skin and flexor
tendon sheath may occur. We report MRI features of
bilateral plantar fibromatosis in the feet with concomitant
hand involvement of palmar fibromatosis and dorsal
knuckle pads.

A 59-year-old woman presented with an 8-year history
of nodules on the plantar surface of both feet. In the pre-
vious 6 months she had experienced painful nodules on
the dorsal surface of the PIP joints of the left fourth and
fifth toes. There was a family history of Dupuytren’s con-
tractures. The patient was otherwise well.

Physical examination revealed marked nodularity of the
plantar fascia of both feet. There were small tender swel-
lings on the dorsum of the PIP joint of the left fourth and
fifth digits. On the palmar surface of the hands there was
fascial cord-like thickening in both hands but no actual
contracture.

MRI of the feet demonstrated multiple heterogeneous
nodular masses along the plantar aponeurosis. Confluent
masses within the distal right plantar fascia measured up
to 4 cm by 1.2 cm in size extending to the fascial insertion
on the head of the first metatarsal. These masses were iso-
tense to muscle on T1-weighted sequences (Fig. 1a) and
heterogeneously high on T2-weighted sequences (Fig. 1b).

MRI of the hand showed subcutaneous nodular thick-
ening of the ulnar aspect of the left palmar aponeurosis
and early traction of the adjacent overlying skin. This
nodularity had low signal on T1- and T2-weighted
sequences (Fig. 1c). There was focal subcutaneous thick-
ening of the dorsal knuckle pads on the third and fourth
PIP joints extending to the underlying extensor tendon
sheaths (Fig. 1d).

Due to the symptomatic nature of the palmar nodules,
the patient was treated with low-dose colchicine and
referred for surgical intervention. She subsequently had
a palmar and digital fasciectomy performed. The plantar
nodules were asymptomatic and therefore not treated.

The plantar aponeurosis is a strong fibrous structure
that extends from the inner tubercle of the calcaneus to
the plantar aspect of the MTP joints and the bases of the
proximal phalanges [2]. The majority of plantar fibromas
are unilateral and solitary; however, as shown in this case,
multiple and bilateral involvements do rarely occur. There
are several reports of an association with Dupuytren’s
contractures of the hand, concomitant disease reported
ranging widely from 10 to 15% [3, 4]. Plantar and palmar
lesions can occur either concomitantly or with one preceding
the other by up to 10 years [5].

The exact aetiology of the disease is still unknown, but it
has been associated with certain risk factors such as ge-
netic predisposition [6] and epilepsy, with a higher reported
incidence in male [1, 3, 7], white [7] and middle-aged
patients [4].

Plantar fibromatosis rarely causes contractures and the
lesions may remain asymptomatic for many years,
preventing as painless firm, fixed nodules on the plantar
aspect of the foot [4]. Symptoms occur because of
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Changes in size of adjacent soft tissues,
local invasion, and mass effect on adjacent muscles and neurovascular bundles [8].

MRI best demonstrates the characteristic lesions of plantar fibromatosis because of the superior soft-tissue contrast resolution. Characteristic features are of soft-tissue nodules on the plantar aponeurosis, with the medial band most commonly affected. The lower margins of the lesions are well circumscribed, whereas an irregular infiltrative upper border occasionally extends into the deep plantar tissues.

The magnetic resonance signal intensity of these nodules is equal to or slightly greater than that of the adjacent muscle on T1 spin echo sequences and heterogeneous in appearance on T2-weighted conventional fast spin echo sequences [10]. Lesions invading deeper structures tend to have a more variable appearance and frequently demonstrate high signal intensity on T2-weighted images [9, 10]. High collagen content has been attributed to be the cause of the occasional low signal intensity appearance in T2-weighted images [10]. Administration of contrast may be useful in identifying small lesions that have been missed on unenhanced scans and in evaluating the extent of infiltration.

Differential diagnosis of a mass on the sole of the foot includes leiomyoma, rhabdomyosarcoma, neurofibroma and liposarcoma [11]. Correlation of clinical, radiological and histological findings helps in confirming a diagnosis.

Treatment depends on the extent of the symptoms and the degree of invasion of these lesions. MRI is the best imaging modality to delineate the margins and depth of soft-tissue invasion of these lesions. Conservative management with padded shoes or moulded insoles may alleviate symptoms. Painful or deeply infiltrative lesions may be treated surgically with wide local excision and occasionally with radiation therapy or MTX chemotherapy.

Plantar aponeurotic and palmar fibromatosis display key characteristic features on MRI. Thus MRI has an important role in both the diagnosis and assessment of the severity of the lesions, thereby guiding appropriate clinical management.

**Rheumatology key message**

- MRI displays characteristic features and demonstrates the severity and depth of fibromatosis, thus guiding clinical management.

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**Collette English**, Robert Coughlan, John Carey and Diane Bergin

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Fetal death in primary SS associated with chronic intervillositis

Sr., Chronic intervillositis is a rare and poorly understood disease characterized by intervillos infiltrates of maternal origin. The intervillos infiltrates originate from mononuclear cells (monocytes, histiocytes) correlating with placental insufficiency, and result in intrauterine growth restriction, intrauterine fetal death and spontaneous miscarriage [1]. Chronic intervillositis aetiology is not well established. Recent physiopathological studies have argued in favour of an immune response resulting from an abnormal maternal reaction to the placental tissue, which could preferentially implicate T-lymphocytes. Thus chronic intervillositis could be associated with autoimmune diseases, such as antiphospholipid syndrome (APS), but its association with other autoimmune disorders has not yet been determined [2]. We describe the case of a pregnant woman with underlying primary SS who presented intrauterine fetal death in relation with chronic intervillositis of the placenta.

A 38-year-old woman, gravida 2 para 1, with no significant past medical history had isolated intrauterine growth restriction during the previous pregnancy. In the 12th week of pregnancy she complained of having had a dry mouth, dry eyes and swelling of the salivary glands, as well as RP for the previous 2 months. There was no argument for any other associated autoimmune disease, in particular SLE or APS. She had positive ANAs (1/1280e), with anti-Ro/SSA and anti-La/SSB. Antibodies against dsDNA, Sm, anti-phospholipid, thyroglobulin and transglutaminase were all negative. The screening of thrombophilia (ATIII, protein C, S, homocysteinaemia, factor V and II mutations) was normal. The serum dosage of vitamin D (250HD3) was 28 ng/ml. The diagnosis of primary SS was confirmed according to the American European Consensus Criteria. HCQ (400 mg/day) with aspirin (100 mg/day) was started at 16 weeks of pregnancy. Regular weekly fetal cardiac echocardiography did not detect any fetal cardiac abnormalities. An US examination at 26 weeks of pregnancy showed a normal biparietal diameter (50th percentile), head (50th percentile), abdominal circumferences (70th percentile) and the absence of oligohydramnios. At 30 weeks of pregnancy sudden fetal death occurred. A fetal autopsy did not reveal any abnormality. In particular, a fetal cardiac examination did not reveal cardiac fibrosis or inflammation. The placenta showed intense mononuclear cells and inflammatory cell infiltrates involving >75% of the intervillos space without villitis (Fig. 1). Immunological staining was positive with anti-CD68 antibodies (Fig. 1). She was pregnant again 6 months later, and the pregnancy resulted in a live-bom baby without any complications at the 38th week of gestation with HCQ (400 mg/day), aspirin (100 mg/day) and prednisone (10 mg/day), started just before the pregnancy.

Reports on pregnancy outcomes beyond neonatal lupus and congenital auriculoventricular (AV) block are rare in primary SS, in contrast to the situation in SLE. Beyond intrauterine AV block and neonatal lupus syndrome, it is usually anticipated that SS is not associated with unfavourable pregnancy outcome [3]. Even fetal cardiac echocardiography did not reveal abnormalities, and AV block can be sudden. AV block cannot be excluded in our case, as no intrauterine growth restriction was noted during pregnancy, but cardiac histological examination was normal. The use of steroids is not routinely recommended in pregnant patients with SS. The chronic intervillositis associated with primary SS has not yet been reported.

The risk of recurrence of chronic intervillositis seems to be high, but no treatment guidelines are currently available [1]. The use of steroids in association with aspirin could improve the obstetrical outcome, but is based mainly on a few case studies [4].