

## A rare case of bilateral plantar fibromatosis (Ledderhose's disease): A case report

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### Abstract

Ledderhose's disease, also known as plantar fibromatosis is a rare, benign hyperproliferative disorder affecting plantar fascia with unknown etiology. Clinical presentation of the disease varies according to the stage of the disease and individual characteristics. Diagnosis of the disease is usually based on clinical findings. Histopathological examination, Ultrasound or MRI can be used to rule out other conditions and for confirmation of the disease. Plantar fibromatosis can mimic the features of plantar fasciitis especially in early stages of the disease, hence it should be considered as a differential diagnosis in patients with pain and nodules in plantar aspect of foot. In this case report, we present a case of 24-year-old male with bilateral plantar fibromatosis, which was managed by surgical excision of the nodules due to unresponsive conservative management.

**Keywords:** Plantar fibromatosis; ledderhose's disease; heel pain; nodular swelling; plantar fasciitis.

### Introduction

Plantar fibromatosis or Ledderhose disease, is a rare benign pathology of the plantar aponeurosis, first described by Dr. George Ledderhose. It is characterised by disordered fibrous tissue proliferation and the subsequent formation of lump or nodules over the plantar aspect of the foot. The Office of Rare Diseases of the National Institutes of Health listed it as a rare disease with frequency about 1–1.75/100,000 [1]. Although etiology of plantar fibromatosis is unknown, it is associated with Dupuytren's disease (palmar fibromatosis), Peyronie's disease (penile fibromatosis) [2]. Increased risk of its occurrence is associated with alcoholism, chronic liver disease, diabetes mellitus, long term anticonvulsive treatment for epilepsy and genetic factors [3]. Males are more commonly affected than females. 25% of cases with plantar fibromatosis present with bilateral disease [4]. Diagnosis of Ledderhose's disease is usually established clinically. Initially the nodule is asymptomatic and it becomes symptomatic as it enlarges in size. Direct pressure on the nodule while walking barefoot, standing for long periods of time and use of restrictive shoes may exacerbate pain and walking disability. Over time, multiple nodules may develop and can cause exacerbation of symptoms, contractures and deformities [5]. Given the benign nature, initial phase of the disease can be managed conservatively and if symptoms persist, definitive management of surgical excision of nodule gives complete relief of symptoms. The nodular swellings affecting the plantar fascia is of greater significance in population with poor socioeconomic status as people prefer

to walk barefoot in developing countries.

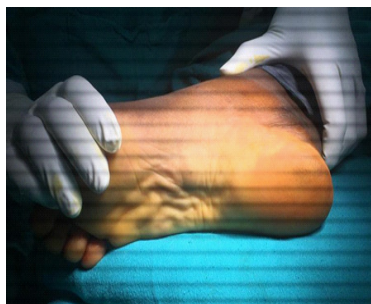
The similarities of plantar fibromatosis to Dupuytren's disease affecting palmar fascia support the theory that, two conditions are different expressions of the same disorder [6]. Even though much has been discussed about Dupuytren's contracture in the literature; only very few literatures are available regarding plantar fibromatosis. In this case report, we present a case of 24-year-old male with bilateral plantar fibromatosis and aims to discuss the clinical presentation and various management options in plantar fibromatosis.

### Case Report

A 24-year-old male presented to our department with dull aching type of pain over the plantar aspect of both feet of 1-year duration. Pain prevented the patient from weight-bearing for long time and walking for small distances. There was no significant familial history of the disease or history of any associated trauma. No associated medical history in the patient. Patient gives history of treatment in another hospital as bilateral plantar fasciitis. Conservative management was given there in the form of analgesics, anti-inflammatory drugs, advice to use footwear with soft insole and gives a history of 3 steroid injections administered 4-6 weeks apart. With persistence of symptoms patient came to our department. On physical examination small, well circumscribed, palpable, firm, nodular, single swelling was present over the medial plantar aspect of his both feet. The swellings measured about 2 x 1.5 cm on the right foot and 1 x 1 cm on the left foot. The skin over the swell-

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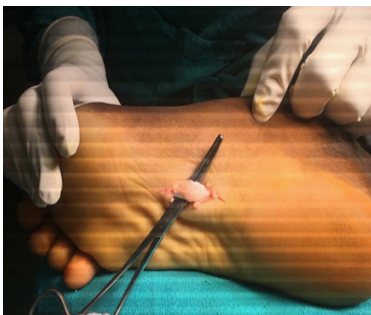
ings appeared normal and there were no neurovascular deficits or deformities. Ankle joint and foot range of movements were within normal range. On further examination, we found a similar swelling of size 0.5 x 0.5 cm on the palmar aspect of right hand with no restriction of movements and clinical signs. FNAC report showed mild to moderately cellular oval to plumb spindle shaped fibroblastic cells with elongated nuclei arranged in clusters and dispersed pattern associated with myxoid matrix. Cytology findings were suggestive of benign fibroblastic lesion. A provisional diagnosis of bilateral plantar fibromatosis was made, based on clinical and cytological findings. Since conservative management was tried earlier and there was persistence of symptoms and limitation in function surgical excision of the nodules was planned. Surgery was performed under spinal anesthesia. Nodules on both sides were palpated and skin over it was marked for surgical incision. The dissection of skin and soft tissue exposed the nodules on both sides, which were greyish white in colour, firm in consistency and attached to plantar fascia (**Figure 3**).



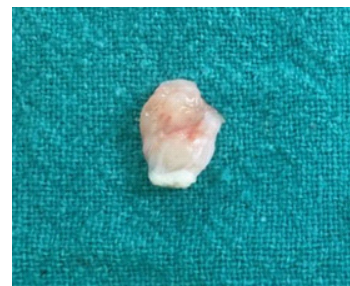
**Figure 1:** Nodular swelling on right foot (dot circle).



**Figure 2:** Dot circle indicating the nodular swelling on right palm.



**Figure 3:** Exposed nodule ( Right foot ) intraoperative image.



**Figure 4:** Excised nodule from right foot (greyish white, measuring 1x0.8x0.2 cm).



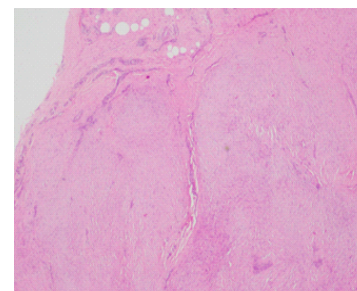
**Figure 5:** Excised nodule from left foot (greyish white to greyish brown, measuring 1.7x1.5x0.4 cm).

Excision of the nodules were done in both feet and primary wound closure was done. The patient was advised for non-weight bearing for 2 weeks and use of soft insole footwear thereafter for 2 weeks. Postoperative period was uneventful and sutures were removed after 2 weeks of surgery.

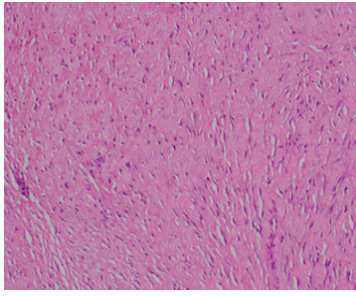


**Figure 6:** Postoperative wound before suture removal.

The histopathological examination of the excised nodules revealed spindle-shaped cells with abundant collagen in a fibrous stroma background and features were consistent with the diagnosis of bilateral plantar fibromatosis. On follow up of 6 months, patient reported complete relief of symptoms and improvement in function.



**Figure 7:** Photomicrograph of HPE slide showing nodular lesion composed of spindle shaped cells in a fibrous stroma background. (H&E staining, x40).



**Figure 8:** Photomicrograph of HPE slide showing spindle shaped cells with abundant collagen in fibrous stroma (H & E staining, x100).

## Discussion

Ledderhose's disease (Plantar fibromatosis) is a fibrous hyperproliferative pathology affecting the plantar fascia characterised by formation of nodules [7, 8]. The diagnosis of Ledderhose's disease is usually established clinically and rarely require further investigations for confirmation [9]. Histopathological analysis and diagnostic imaging helps to differentiate between other lesions that can present with similar symptoms such as plantar fasciitis (The most common disorder of plantar fascia), lipoma, ganglion cyst, leiomyoma, epithelioid sarcoma, rhabdomyosarcoma and liposarcoma [10, 11]. According to the clinical and pathological studies, plantar fibromatosis can be classified into three stages. The first (proliferative) stage of the disease is characterised by cellular proliferation and increased fibroblastic activity. The second stage of the disease which is the active phase is characterised by formation of nodules. It is followed by the third (residual) stage where collagen maturation and tissue contractures occur [11, 12]. Therefore the normal plantar fascia is replaced progressively by abnormal collagen fibres and can present at any stage of the disease with pain, nodule, walking difficulty, contractures or deformities of toes and the treatment is planned accordingly.

Patients presenting in the early stage of the disease with no or mild pain can be conservatively managed with padded shoes with soft insoles or custom offloading to redistribute the weight from the nodules, analgesics, anti-inflammatory drugs and intralesional steroid injections [10,13]. If left untreated, nodules may gradually increase in size and number which in rare cases may result in deformities of the toes due to contractures in later stages. In cases with persistence of symptoms after conservative management, lesions which are progressive, severe limitation of function and in advanced stages of the disease surgical management is considered as the last resort of treatment [14, 15].

The nodular swellings affecting the plantar fascia is of greater significance in developing countries with poor socioeconomic status as people prefer to walk barefoot. For the same reason, early surgical management is indicated for symptomatic cases in the developing countries.

## Conclusions

The diagnosis of Ledderhose's disease can be done clinically alone. Diagnostic imaging such as Ultrasound or MRI and histopathological examination may be used, to exclude other conditions and to rule out malignancies [16]. The treatment of the disorder is planned accordingly. Even though plantar fasciitis is the commonest disorder affecting plantar fascia, plantar fibromatosis should be considered as a differential diagnosis

in patients presenting with pain and nodules in plantar aspect of foot as it can mimic the features of plantar fasciitis. The recommended treatment approach is to start with conservative management in early stages of the disease and perform surgical excision in unresponsive cases and advanced stages. But the best treatment plan is to establish a personalised approach depending on the individual characteristics, type of symptoms, stage of the disease and recurrence.

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