# Intralesional fenestration and corticosteroid injection for symptomatic Ledderhose disease of the foot: Two case reports

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#### Abstract

The description of corticosteroid injections as a treatment option for Ledderhose disease has received little attention in the literature and often only receives a passing comment in scientific papers. We present a short case series of two patients who underwent corticosteroid injection in combination with fenestration to treat painful Ledderhose disease nodules. Both patients had their lesions injected on two occasions. Significant reduction in pain and lesion volume was seen at 12 months post treatment. Our protocol combines fenestration with the use of triamcinolone acetonide (mixed with local anaesthetic) which we believe conveys further advantage over other steroid preparations or the corticosteroid infiltration alone.

#### **Keywords**

Ledderhose disease, plantar fibromatosis, corticosteroid injection, triamcinolone, fenestration, needling, peppering

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## Introduction

The fibromatoses encompass a broad array of proliferative fibroblastic disorders which can be defined by location (superficial or deep) and biological behaviour (benign, intermediate and aggressive).<sup>1</sup> They share common histological appearances which include spindle-shaped myofibroblasts, significant intercellular collagen fibres, compressed and elongated vessels and varying appearances of extracellular myxoid matrix.<sup>2</sup> Ledderhose disease (LD) belongs to the family of superficial fibroblastic proliferative diseases that includes Dupuytren's disease of the palmar fascia (palmar fibromatosis) and Peyronie's disease (penile fibromatosis). Madelung reported the first isolated case of plantar fibromatosis in 1875,<sup>2</sup> but the condition was described in greater detail by Dr George Ledderhose in 1894.<sup>3</sup> LD is a relatively uncommon benign fibrous proliferation of the plantar fascia (aponeurosis). On immunohistochemistry (IHC), it is characterised by a population of cells that stain for smooth muscle actin, indicating focal myofibroblastic differentiation.<sup>2</sup> Mild perivascular chronic inflammation and deposits of hemosiderin is occasionally seen, focal chondroid or osseous metaplasia possible in chronic lesions.<sup>4</sup>

Many aetiological factors have been described in the literature. Males are at twice the risk of developing LD; in addition, diabetes, nicotine use, medication (phenobarbital and anti-TNF), alcohol misuse and genetic predisposition have all been cited as risk factors.<sup>5</sup> Surgical procedures for LD include open fibrotomy with varying recommendations for the size of margin to be included. A high failure and recurrence rate for the surgical treatment of LD is well documented<sup>6</sup> but despite the poor outcomes associated with the surgical treatment of LD, surgery still accounts for the majority of published evidence available. A wide variety of nonsurgical options exist with variable outcomes, of which injection with a corticosteroid (CSI) receives relatively little attention compared to CSI for plantar fasciopathy.<sup>5,6</sup> Fenestration, also known as needling, is a well-described technique used for many soft tissue pathologies including plantar fasciopathy.<sup>7</sup> For the last 5 years we have utilised a fenestration and CSI combination technique for LD nodules with (anecdotally) excellent results in terms of both symptom relief and reduction in nodule rigidity and size. Here, we report on two cases with a 12-month follow-up. Neither patient had significant pre or post injection conservative care, other than the use of orthoses and occasional

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Figure 1. Case I USS pre-injection (larger, distal lesion).

painkillers, beyond the fenestration technique described below. As far as the authors are aware, this approach to the treatment of LD has not previously been reported.

## Case report I

#### History

A 54-year-old man originally presented to his primary care physician in January 2019 complaining of two painful swellings in the arch of his left foot. An ultrasound scan (USS) examination was requested. Two well-defined hypoechoic lesions arising from the distal portion of the left plantar fascia were described. The distal lesion measured 13.5 mm in length  $\times$  5.2 mm in width (Figure 1) the proximal lesion measured 7.9 mm  $\times$  2.8 mm (Figure 2). No significant intrasubstance vascularity was seen. The remaining plantar fascia was normal. Appearances were in keeping with LD.

The patient had a history of nicotine use (15–20 cigarettes a day), essential hypertension and chronic lumbar region pain. He had stable epilepsy and was recovering from alcohol abuse. His current medication was lisinopril 20 mg od, gabapentin 400 mg tds, codeine/paracetamol 30/500 mg combination prn and sodium valproate 500 mg tds. The patient had concomitant palmar Dupuytren's disease on the left hand with fascial nodules and contracture of the fourth finger. There were no known allergies and no relevant family or surgical history.

Following the results of the USS organised in primary care, he was seen by our service in February 2019 complaining of searing pain on the medial longitudinal arch of the left foot of 6 months duration. He described the pain as severe and debilitating and rated his discomfort at 9/10 on a visual analogue scale (VAS). On examination he had two firm, subepidermal swellings in the distal-medial aspect of the arch associated with the central slip of the plantar fascia. Normal neurovascular status was observed. His symptoms had worsened over the previous 3 months, aggravated by activity and weightbearing. Prior treatment included accommodative footwear and orthoses. Intralesional CSI in combination with a fenestration/needling was recommended and informed, written consent was obtained, backed up with a patient information leaflet.<sup>8</sup> Written permission to use images for publication was obtained.



Figure 2. Case I USS pre-injection (smaller, proximal lesion).

#### Procedure

One week following initial consultation, the patient attended for their planned procedure. As the patient presented with significant pain, the procedure was performed under tibial nerve block, performed at the level of the ankle with 3 mL of 0.75% ropivacaine under aseptic technique. The injection sites were prepared with a chlorhexidine gluconate 2%/isopropyl alcohol 70% mix. A sterile cover was placed on the ultrasound probe and sterile coupling gel utilised. The ultrasound probe was placed plantarly and longitudinal to the lesion to allow for a medial injection approach. A mixture of 20 mg (40 mg/mL) triamcinolone acetonide and 1 mL of mepivacaine hydrochloride was deposited with intralesional fenestration (20-30 repeated passes with multiple microdeposition sites from proximal to distal and medial to lateral without removing the needle from the skin) initially to the distal lesion followed by an identical preparation administered to the proximal lesion. The whole of the fibroma is targeted but avoiding being too superficial (skin atrophy), or too deep (reduced effectiveness). A sudden loss of resistance during the injection indicates that the lesion has been penetrated too deeply or adjacent to the lesion.

Both injections were performed under ultrasound guidance with confirmation of intralesional deposition. An extremely small amount of perilesional injectate leakage was noted. Importantly, a 2.5-mL Luer lock syringe with a 23 gauge/25 mm needle was utilised as experience has shown the presence of pressure when injecting a solid mass which can cause de-coupling of the syringe from needle. No significant bleeding was associated with the procedure. A simple self-adhesive dressing was applied to the injection sites. Post procedure advice included rest and foot elevation for the remaining day. The patient could return to daily living activities the following day but was to refrain from impact activity, for example, sports until follow-up at 6 weeks post procedure. As well as the usual post-injection advice, particular reference to the small risk of fascial rupture was made.

## Follow-up

No significant beneficial effect was witnessed at the 6-week review, as such, a second injection was recommended. Eight weeks following the original procedure, the same protocol as



**Figure 3.** Case 1: second injection without USS. We find that a medial approach rather than a plantar approach is more comfortable for the patient.



Figure 4. Case I: USS post second injection (larger, distal lesion).

described above was undertaken. In this instance, palpationguided injection and fenestration was undertaken (Figure 3). Six weeks post the second procedure (total 14weeks post index procedure), a significant reduction in the size and rigidity of both lesions was reported clinically. A repeated VAS was 2/10.

For purposes of audit, the patient was reviewed at 12 months post procedure and a further USS examination was undertaken showing a significant regression of both nodules: the distal nodule to  $6 \text{ mm} \times 1.9 \text{ mm}$  (Figure 4) and the proximal nodule to  $2.4 \text{ mm} \times 1.2 \text{ mm}$  (Figure 5). During this period, the patient had no regression of symptoms. A VAS completed at 12 months was 0/10. Clinically, the proximal nodule was no longer palpable or visible, the distal nodule remained palpable but not visible. No pain was felt on palpation. The patient remarked they were unable to feel any nodule on ambulation. No adverse signs or symptoms were reported.



Figure 5. Case I: USS post second injection (smaller, proximal lesion).

## Case report 2

## History

A 55-year-old woman attended our service in August 2018 complaining of bilateral plantar nodules; at initial presentation only the right foot was symptomatic. The patient's medical history included type 2 diabetes mellitus of 12 years duration (recent HbA1c 52 mmol/mol) and essential hypertension. Current medication was empagliflozin 10 mg od, linagliptin 5 mg od, gliclazide 80 mg bd, ramipril 10 mg od, atenolol 50 mg od and amlodipine 15 mg nocte. There were no known allergies and no relevant family/social or surgical history.

When assessed the patient described a 12-month history of a symptomatic mass within the medial longitudinal arch of the left foot, a VAS of 8/10. Symptoms were exacerbated by prolonged periods of ambulation and weightbearing. Orthoses and footwear adaptation had not yielded any symptomatic reprieve. Clinically, a solitary mass could be palpated within the medial longitudinal arch of the right foot, and tenderness was apparent on palpation. The mass was rigid, overlying tissue unremarkable. Neurovascular observations were unremarkable. Subsequently, an USS examination was requested (Figure 6). The proximal plantar fascia appeared normal; however, a large solitary well-defined hypoechoic mass was seen within the medial band of the plantar fascia measuring 24.1 mm in length and 7 mm in width. Appearances again were consistent with LD. As conservative treatment options had failed so far, a CSI was suggested. Informed written consent was obtained, backed up with a patient information leaflet.8 Written permission to use images for publication was obtained.

## Procedure

In September 2018, the patient underwent a single injection, without fenestration, of 40 mg (40 mg/mL) methylprednisolone acetonide pre-mixed with lidocaine (10 mg/ mL) under USS guidance. While moderate improvement was seen at 6 weeks post procedure, the patient returned 3 months following the injection noting complete recurrence of symptoms. The patient was subsequently listed for



Figure 6. Case 2: USS pre-injection.

the triamcinolone acetate injection with fenestration under USS guidance. This was performed in March 2019: the injection technique mirrored that described in case study 1. At 6 months post final procedure, the patient reported VAS as 1/10 and return to full activities.

For audit purposes, the patient was contacted for review at 12 months post procedure and USS examination performed in March 2020. The VAS was 0/10; clinically the mass was significantly smaller with only minimal deciphering on manual palpation. The USS demonstrated a small hypoechoic mass sited in the same area as previously demonstrated but with reduced dimensions, as seen in Figure 7 (note: as this coincided with the start of the Covid-19 pandemic, the lesion was scanned by ourselves rather than by the local sonography team who were only taking emergency cases at that time, and therefore no definitive dimensions are offered). No adverse signs or symptoms were noted and no further treatment of any sort had been required.

## Discussion

The use of corticosteroid injection for Dupuytren's disease has been reported in the hand<sup>9</sup> and their use for LD in the foot has been discussed (often only in passing) by various authors.5,6,10-12 Local steroid injections reduce the rate of fibroblast proliferation and increase the rate of apoptosis.<sup>13</sup> Ketchum et al.<sup>14</sup> had previously demonstrated that triamcinolone acetonide softened and flattened hypertrophic scars and keloids and that it degraded the insoluble collagen in hypertrophic scars and keloids to salt-soluble collagen, which was then absorbed and excreted. In a retrospective study,<sup>9</sup> 63 patients with Dupuytren's nodules in the early stages of disease underwent a series of triamcinolone acetonide injections. Each injection contained 60-120 mg of triamcinolone administered directly into nodule(s) of patients with contracture of less than 15 degrees. 97% (62/63) of patients experienced regression of the disease exhibited as softening or flattening of nodules, with an average of 3.2 injections per nodule reported. Some had complete resolution of the nodule



Figure 7. Case 2: USS post injection.

but more (60%–80%) experienced definite but incomplete resolution. Although immediate regression of nodules was generally observed, many experienced recurrences and around half of those required further injections 1–3 years after their initial treatment.

Pentland and Anderson<sup>11</sup> presented a case study of a patient with bilateral multi-nodular plantar fibromas, recurrent on the right foot after excisional surgery 10 years previously. The patient received five intralesional injections of 0.5–1.0 mL of triamcinolone acetonide diluted 3:1 with 1% lidocaine hydrochloride to a final concentration of 30 mg/ mL, at monthly intervals. The reader assumes that each of the lesions had that dose. Considerable softening of the lesions was noted and 4 months after the final injection the patient was able to resume jogging. They discuss the conflicting results of the effect of corticosteroid injections on in vitro fibroblast collagen production but promote corticosteroid for LD.

A 'peppering' technique was first described in 1964<sup>15</sup> and later popularised clinically for lateral epicondylitis.<sup>16</sup> Peppering, needling and fenestration are all terms used in the literature across professional groups and across pathologies, in particular plantar fasciitis. We prefer the term fenestration as we aim to create channels within the lesion. We hypothesise that this has the effect of physically breaking down scar tissue while providing an effective portal of entry for the corticosteroid. As such we believe that fenestration confers greater efficacy than use of corticosteroid injection alone. We routinely use 20–30 passes of a 23-gauge (blue) needle, to patient tolerance, varying depending on the size and turgidity of the lesion and patient discomfort.

While it has been stated that a number of classification systems exist,<sup>5</sup> the only noted staging system is that developed by Sammarco and Mangone.<sup>17</sup> They produced a fourstage system incorporating the focal nature of the lesion(s), the extent of fascial involvement, the presence of skin adherence and the depth of tumour extension. The remaining classification systems used for LD are derived from the work of Luck<sup>18</sup> in staging Dupuytren's disease. The proliferative stage is characterised by increased fibroblastic activity and reduced collagen network, followed by the active or involutional stage showing fibroblast maturation, myofibroblast differentiation and increased collagen synthesis. A final residual stage displays both reduced fibroblast and collagen maturation. While it has been opined that corticosteroid injection would specifically help with collagen breakdown at the residual (end) phase of a chronic LD nodule,19 corticosteroid injection has been shown to suppress VLA-4 a common integrin integral to cell adhesion in early inflammation.<sup>20</sup>

The authors have treated over 25 patients with the procedure described in the text, while outcome data for those patients is anecdotal and/or preserved within medical notes; the authors believe that the majority of patients show significant improvement with the particular combination of fenestrated triamcinolone acetate with a small amount of local anaesthetic. It is recognised that the variability of local anaesthetic as part of the injectate may influence outcomes, as local anaesthetic itself has been shown to cause apoptosis.<sup>21</sup> Case report 2 has previously trialled a single injection of methylprednisolone acetate and lidocaine, and this was followed by a quick and complete return of symptoms: this is a trend seen anecdotally within our service with the use of methylprednisolone acetate. We have abandoned its use for the treatment of LD.

While a tibial anaesthetic block allowed for procedural anaesthesia in case report 1, it should be noted that the majority of patients tolerate local infiltration and fenestration with local anaesthetic added to the injectate only. The use – or not – of concurrent ultrasound guidance is a further matter that is open for debate. Typically, the lesions are sub-dermal and easy to identify with the needle tip though Sofka and Adler suggest that ultrasound guidance is useful to help prevent inadvertent injection of corticosteroid to non-target tissues.<sup>22</sup> With our small case series, we are not in a position to state whether the use of ultrasound is an advantage or not. Certainly in those lesions not readily discernible by palpation, the use of ultrasound guidance is helpful.

We have not, to our knowledge, seen a case of fascial rupture or significant tissue atrophy post corticosteroid injection/fenestration but of course this remains a concern. However, given that the surgical option is excision (narrow or wide margin) or sub-total fasciectomy, the occurrence of a partial tear could be considered to be of minimal concern. The authors do however note the mounting evidence to support the use of ultrasound-guided injection, particularly in this anatomical region.<sup>23</sup> While described as an uncommon condition,<sup>24</sup> Bakotic and Borkowski<sup>25</sup> found LD to be the most prevalent of all the plantar lesions identified by histopathology in a series of 401 cases.

## Conclusion

Throughout this short review and – a common theme in the literature cited – was the need for further high-level investigation into the effect of corticosteroid on LD nodules. While the conclusion from this case series is limited, we feel that the addition of fenestration to triamcinolone acetonide intralesional injection warrants further investigation. A study of sufficient size and power might suggest at what stage this intervention achieves the best outcome, and if the concurrent use of ultrasound guidance conveys further advantage.

#### Authors' note

The authors confirm that they adhered to virtue-based and principle-based ethics when producing this work.

#### **Author contributions**

I.N.R. conceived and wrote the first draft of this document. All authors approval the final version.

#### **Declaration of conflicting interests**

The author(s) declared the following potential conflicts of interest with respect to the research, authorship, and/or publication of this article: The senior author (G.F.) is a consultant for the British Dupuytren's Society.

#### Ethical approval

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#### **Informed consent**

Written informed consent was obtained from the patient(s) for their anonymised information and images to be published in this article, a copy of which is retained in their medical records.

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#### References

- 1. Walker EA, Petscavage JM, Brian PL, et al. Imaging features of superficial and deep fibromatoses in the adult population. *Sarcoma* 2012; 2012: 215810.
- 2. Goldblum JR, Weiss SW and Folpe AL. Benign fibroblastic/myofibroblastic proliferations, including superficial

fibromatoses in Enzinger and Weiss's soft tissue tumors. 7th ed. Amsterdam: Elsevier Health Sciences, 2019, pp. 260–263.

- DeBrule MB, Mott RC, Funk C, et al. Osseous metaplasia in plantar fibromatosis: a case report. *J Foot Ankle Surg* 2004; 43(6): 430–432.
- 4. Ledderhose G. Über zerreisungen der plantarfascie. *Arch Klin Chir* 1894; 48: 853–856.
- Espert M, Anderson MR and Baumhauer JF. Current concepts review: plantar fibromatosis. *Foot Ankle Int* 2018; 39(6): 751–757.
- 6. Veith NT, Tschernig T, Histing T, et al. Plantar fibromatosis: topical review. *Foot Ankle Int* 2013; 34(12): 1742–1746.
- Kiter E, Celikbas E, Akkaya S, et al. Comparison of injection modalities in the treatment of plantar heel pain: a randomized controlled trial. *J Am Podiatr Med Assoc* 2006; 96(4): 293–296.
- Department of Podiatric Surgery. Local steroid injections: pre injection. Northamptonshire Healthcare Foundation NHS Trust, 2019, https://www.nhft.nhs.uk/download.cfm?doc= docm93jijm4n13065 (accessed 25 December 2020).
- Ketchum LD and Donahue TK. The injection of nodules of Dupuytren's disease with triamcinolone acetonide. *J Hand* Surg Am 2000; 25(6): 1157–1162.
- Aluisio FV, Mair SD and Hall RL. Plantar fibromatosis: treatment of primary and recurrent lesions and factors associated with recurrence. *Foot Ankle Int* 1996; 17(11): 672–678.
- Pentland AP and Anderson TF. Plantar fibromatosis responds to intralesional steroids. *J Am Acad Dermatol* 1985; 12(1 Pt. 2): 212–214.
- Neagu TP, ŢigliÅŸ M, Popescu A, et al. Clinical, histological and therapeutic modern approach of Ledderhose disease. *Rom J Morphol Embryol* 2018; 59(3): 691–697.
- Meek DRM, McLellan S, Reilly J, et al. The effect of steroids on Dupuytren's disease: role of programmed cell death. J Hand Surg Br 2002; 27(3): 270–273.

- Ketchum LD, Smith J, Robinson DW, et al. The treatment of hypertrophic scar, keloid and scar contracture by triamcinolone acetonide. *Plast Reconstr Surg* 1966; 38(3): 209–218.
- 15. Pruce AM, Miller JA and Jr Berger IR. Anatomic landmarks in joint paracentesis. *Clinical Symposia* 1958; 10(1): 3.
- Altay T, Günal I and Öztürk H. Local injection treatment for lateral epicondylitis. *Clin Orthop Relat Res* 2002; 398: 127–130.
- Sammarco GJ and Mangone PG. Classification and treatment of plantar fibromatosis. *Foot Ankle Int* 2000; 21(7): 563–569.
- 18. Luck JV. Dupuytren's contracture: a new concept of the pathogenesis correlated with surgical management. *J Bone Joint Surg Am* 1959; 41-A(4): 635–664.
- Downey MS. Point-Counterpoint: is conservative care the best approach for plantar fibromatosis? *Podiatry Today* 2013; 26(2): 36–42.
- Meek RMD, McLellan S and Crossan JF. Dupuytren's disease: a model for the mechanism of fibrosis and its modulation by steroids. *J Bone Joint Surg Br* 1999; 81(4): 732–738.
- Perez-Castro R, Patel S, Garavito-Aguilar ZV, et al. Cytotoxicity of local anesthetics in human neuronal cells. *Anesth Analg* 2009; 108(3): 997–1007.
- 22. Sofka CM and Adler RS. Ultrasound-guided interventions in the foot and ankle. *Sem Muscul Radiol* 2002; 6(2): 163–168.
- Li Z, Xia C, Yu A, et al. Ultrasound-versus palpation-guided injection of corticosteroid for plantar fasciitis: a meta-analysis. *PLoS ONE* 2014; 9(3): e92671.
- National Center for Advancing Translational Sciences. Ledderhose disease: genetic and rare diseases information center, 2018, https://rarediseases.info.nih.gov/diseases/6873/ ledderhose-disease (accessed 25 December 2020).
- Bakotic BW and Borkowski P. Primary soft-tissue neoplasms of the foot: the clinicopathologic features of 401 cases. *J Foot Ankle Surg* 2001; 40(1): 28–35.