Phalangeal fracture leading to digital amputation, sustained during manipulation for Dupuytren's disease as part of clostridial collagenase treatment

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ABSTRACT

Collagenase injection was approved in Europe for use in the treatment of Dupuytren's disease in 2011. Reported adverse effects include skin tears, swelling, pain and complex regional pain syndrome. Here, we present a case of a rare complication of phalangeal fracture following treatment with collagenase clostridium and the resultant digital amputation.

An 81-year-old man was treated for a 25-year history of progressively disabling Dupuytren's disease of his left middle and ring fingers. Nine days post-manipulation he presented with persistent pain and swelling and was diagnosed with a displaced fracture of the proximal phalanx of his ring finger. Following discussion of surgical options and potential outcome, he elected to undergo amputation of the digit. He has experienced good treatment results in his middle finger and has no significant functional impairment as a result of this complication. Those offering collagenase injection in the management of Dupuytren's disease should be aware of this potential complication and it should be included in the consent process.

KEYWORDS

Dupuytren's disease – *Clostridium histolyticum* collagenase – Amputation

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Introduction

Collagenase clostridium histolyticum is an enzyme produced by the bacterium *Clostridium histolyticum*. It was approved in 2011 by the European Commission's Committee for Medicinal Products for Human Use for the treatment of Dupuytren's disease, which is highly prevalent in Northern Europe and causes significant disability. Contractures were traditionally addressed through surgical release, either minimally invasive or open. While surgery potentially enabled release of all contractures in a single procedure, recurrence rates have remained high. Collagenase treatment has proven highly effective as an alternative option.

Rarely is a treatment without potential adverse effects or complications. As newer treatments become more widely used, it is important that we remain abreast of any new complications and use this to enable us to weigh up individual risk benefit profiles. Here, we describe an unusual case of proximal phalanx fracture as a consequence of collagenase treatment, and resultant digital amputation.

Case history

An 81-year-old right-hand-dominant man presented with a 25-year history of progressive contractures of his left middle and ring fingers. The contractures had shown significant progression in the immediate preceding years, with increasing functional impairment. Significant past medical history included surgery for varicose veins and a previous cholecystectomy. He had been treated three months prior to his referral for fingertip injuries to the left middle and ring fingers, requiring terminalisation at the distal interphalangeal joints. Despite a career as a carpenter, he had sustained no other significant hand injuries and was living independently with his wife.

Examination at the time of referral revealed Dupuytren's disease involving the left middle and ring fingers. The extent of deformity was as follows: left ring finger metacarpophalangeal joint 90-degree flexion, proximal phalangeal joint 110-degree flexion; left middle finger metacarpophalangeal joint 70-degree flexion, proximal interphalangeal joint 100-degree flexion. Discussion of treatment options included no intervention, collagenase injection and

Figure 1 Radiograph demonstrating fracture to base of proximal phalanx of ring finger.

surgical treatment. The patient opted for collagenase injection treatment, which he underwent eight months after initial consultation, followed by manipulation. The collagenase injection used was Xiapex®. The outcomes were reported as significant skin tears, moderate pain and bruising and minimal swelling.

Three days following manipulation the patient attended for routine dressing change and review of skin tears. At this time it was noted there was significant swelling of the ring finger and hand and the patient was referred for review by hand therapy. At nine days post-manipulation, with improvement in swelling, there was noticeable deformity of the fourth metacarpophalangeal joint. Imaging revealed a significantly displaced fracture of the proximal phalanx (Fig 1).

The options for fracture management were discussed with the patient, including the potential for surgical treatment. A decision was reached to amputate the ring finger at the base of the proximal phalanx, in preference to general anaesthesia and challenging fixation (Fig 2). At the time of surgery the bone was noted to be osteopenic. There were no other complicating factors.

Following surgery the patient underwent hand therapy. He achieved good functional outcome. On clinical review six months post-amputation the patient had returned to all normal activities and was satisfied with the resultant outcome (Fig 3).

Discussion

Dupuytren's is a disease most prevalent within Northern Europe, principally among those of Viking descent. As a society, we are therefore well placed not just to strive to be at the forefront of treatment development, but also at recognising those less prevalent and rare complications.

The first surgery for contractures was performed by Dupuytren himself, in 1831.¹ Since this time, a variety of

Figure 2 Postoperative image demonstrating level of amputation.

treatments have been employed, including fasciotomy, fasciectomy, radiation, ultrasound, dimethyl sulfoxide, steroids and interferon. Attempts have also previously been made at dissolving the tissue, including the use of fibrinolysin in 1907 and trypsin with hyaluronidase in 1965. The use of clostridium collagenase was first reported in 2000, by Badalamente and Hurst, who injected the treatment into the hands of 29 patients, to good effect.²

Initial reports of collagenase injection reported both successful outcomes and treatment failures. Current literature seeks to make a comparison between treatment options available and success and complication rates. Pooled complication rates for fasciectomy are reported at 17.4%, for percutaneous needle fasciotomy at 18.9% and for collagenase treatment at 78%.³ As would be expected, different complications are reported for fasciectomy and collagenase treatment, with complex regional pain

Figure 3 Postoperative image demonstrating functional outcome.

syndrome being common to both. In the case of fasciectomy complications included nerve division (2–5%), infection (4–12%), neuropraxia (0.4–52%) and regional pain syndrome (2–13%). The same review of current literature reported that for collagenase injection skin tear predominated, with a complication rate of 9–25%.⁴

A number of other complications have been associated with collagenase treatment. Flexor tendon injury has been reported in 0.27% of cases and is most common in the little finger. Partial tendon injury is also noted. Contusions (51%), injection site haemorrhage (37%), ecchymosis (25%) and blood blister (3.4%) have all been documented.⁵

To date, only a single case of fracture following collagenase treatment for Dupuytren's disease has been reported in the literature. This case report from 2017 documents the case of a patient with contractures of the metacarpophalangeal and proximal interphalangeal joints of 87 and 78 degrees, respectively, who underwent collagenase injection and manipulation. The patient sustained a proximal phalanx fracture of the little finger, which was managed conservatively with good result. The patient also had a number of concomitant comorbidities including oncological treatment. As a consequence, the report highlights the need for caution in elderly patients with comorbidities who may be more susceptible to this complication.⁶ The case we present had worse contractures and no significant comorbidities. We propose that comorbidities will increase the risk of fracture but do not always need to be present and therefore each case needs to be assessed individually.

In combination with the previously reported case, our clinical experience builds a body of evidence for fracture as a complication associated with Xiapex® injection and manipulation for Dupuytren's disease. The two cases in which fractures have occurred involve patients who have contraindications or who are high risk for operative management or have previously relapsed following this. Both patients were probably osteopenic at the time of manipula-

tion. Screening for osteopenia could identify such patients, but it is unlikely this knowledge would impact on management decisions. These are patients who experience significant impact on their daily function because of their limitation and in whom treatment options are limited.

We conclude that fracture should be recognised as a potential complication and consented for prior to treatment. Although the incidence is rare, we have a duty in law to ensure patients are fully aware of all risks associated with the procedure.⁷

Conclusions

We present the second reported case of phalangeal fracture following collagenase treatment for Dupuytren's disease. This complication should be considered in calculating the risks and benefits of this treatment for each individual patient. Those patients undergoing treatment with collagenase injection and manipulation should be informed of this risk.

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