

Phalangeal Fracture During Attempted Dupuytren's Release Following Clostridial Collagenase Injection: Case Report

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What to Learn from this Article?

Physicians should consider fracture resulting from release of contracture is possible in elderly persons with osteoporosis.

Abstract

Introduction: Dupuytren's disease can be a challenging condition for both patients and surgeons. Injectable collagen clostridium histolyticum was approved for clinical use by the Food and Drug Administration in 2010. A number of side effects have been described.

In this case report, we present a complication of a proximal phalanx fracture which occurred during attempted release post injection. To the best of our knowledge, this is the first reported case of this particular complication.

Case Report: The patient is an 80-year-old right-hand dominant retired male with bilateral hand contractures and palmar fibromatosis. His medical history is notable for Type II diabetes mellitus and metastatic thyroid cancer. The patient underwent Xiaflex injection of the left small finger and returned 4 days later for planned release. An appreciable release of the contracture was noted; however, there was a concern for plastic deformation of the proximal phalanx as a result of the manipulation. X-rays confirmed the fracture and apex volar angulation at the base of the proximal phalanx. The fracture appeared amenable to non-operative treatment. The patient has been followed closely and has had no pain or tenderness at the fracture site and minimal swelling. X-rays at 1 week and 1 month showed maintained alignment and signs of consolidation at the fracture site. The patient is currently 4-month post-fracture, and no further intervention has been pursued.

Conclusions: This report represents an additional potential complication associated with the use of Xiaflex. We recommend judicious use in elderly patients with severe contractures and/or multiple comorbidities. It is important to appreciate the possibility of this complication at the time of release.

Keywords: Dupuytren's contracture, proximal phalanx fracture, collagen clostridium histolyticum, phalangeal fracture, Dupuytren's release.

Introduction

Dupuytren's can be a challenging condition for both patients and surgeons. It is a disease of the fascia that typically affects persons of northern European ancestry. While it is benign in behavior, recurrence and progression are not uncommon. Treatment options have evolved over the past 200 years since the early days of treatment were first described in the late 17th century [1]. Surgery, primarily limited/complete fasciectomy with contracture release, has been the mainstay of treatment. Unfortunately, recurrence rates with surgery can be substantial and have been reported as high as 66% in some reports [2, 3, 4, 5, 6]. In addition, treatment of recurrence following surgery has proven to be

challenging.

Non-surgical treatments such as needle aponeurotomy and collagenase have gained in popularity over the past few decades. While they are not proven to be superior to surgery in efficacy, they have the advantage of convenience (office based). In addition, they are less expensive and require less formal therapy. Injectable collagen clostridium histolyticum (CCH) was approved for clinical use by the United States Food and Drug Administration (FDA) in 2010. The aim of the treatment is to enzymatically dissolve a portion of the cord, in hopes of effectively weakening it, so that the contracture can be released. Prospective randomized analyses have demonstrated that the medicine is quite effective [7].

Author's Photo Gallery



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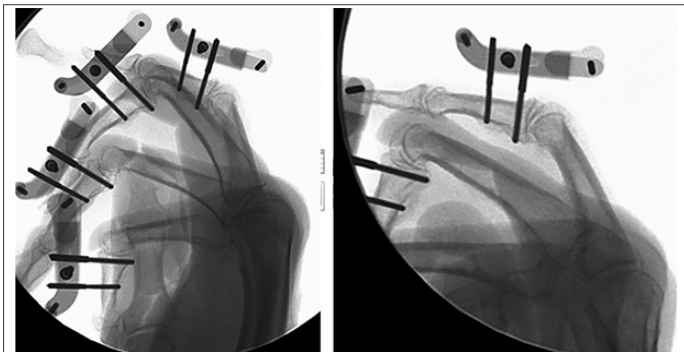


Figure 1: (a and b) Fluoroscopic images obtained at the time of digit widget application show severe metacarpophalangeal and proximal interphalangeal contractures.

A number of side effects have been described, including pain, swelling, bruising, hematoma, lymphadenopathy, pruritus, rash, blood blister, and skin tears [7, 8]. More serious complications have also occurred, including tendon rupture, pulley rupture, allergic reaction, and neurovascular injury. In this report, we present a complication of a proximal phalanx fracture which occurred during attempted release post injection. To the best of our knowledge, this is the first reported case in the literature.

Case Report

An 80-year-old right-hand dominant retired male was referred for ongoing treatment of bilateral hand contractures and palmar fibromatosis. 6 months before presentation to us, he had undergone surgery consisting of subtotal palmar fasciectomy, carpal tunnel release, and full-thickness skin grafting. Unfortunately, he developed worsening recurrent contractures shortly after his surgery, and it progressed in severity to worse than pre-operative levels.

His medical history was notable for Type II diabetes mellitus. The patient was unfortunately diagnosed with metastatic papillary thyroid cancer several months after presentation to us. He subsequently underwent total thyroidectomy with the neck and lymph node dissection, followed by radiation and chemotherapy after an evaluation revealed pulmonary, bone, cervical lymph node, and dermal metastases. His current medications include albuterol, allopurinol, citalopram, lenvatinib, metformin, pantoprazole, budesonide/formoterol, and levothyroxine.

Physical examination revealed flexion contractures of 38, 46, 77, and 87° at the metacarpophalangeal (MCP) joints and 58, 81, 85, and 78° at the proximal interphalangeal (PIP) joints (index-small fingers, respectively). He was essentially able to make a full composite fist. At the time of presentation, the patient was significantly disabled and desirous of intervention. Treatment

options were discussed including revision surgery, needle aponeurotomy with or without the use of digit widget application, and clostridial collagenase.

The patient chose and underwent needle fasciotomy, digit widget application of the left index through small fingers. (Fig. 1) illustrates the degree of contractures at the time of digit widget application. The patient experienced notable improvement of the contractures with the widget. At 4-week post-surgery, the widget was removed. The PIP contractures significantly improved, and though improved, he still had evidence of MCP contractures (Fig. 2). Unfortunately, over the next 2 months, the contractures slowly recurred, with his small finger most significantly involved.

We discussed options at that time and given his current health status and the disappointing outcome from prior surgery; he was understandably reluctant to undergo further surgery. The patient underwent Xiaflex injection of the left small finger. He returned 4 days after the injection for planned release. Physical examination at the time of release revealed an appropriate amount of bruising and swelling. He was able to demonstrate finger flexion. The cord appeared to have responded favorably to the injection as it was quite soft at the injection site. The finger was anesthetized through a digital block before attempted cord release. Despite the lack of an obvious give of the cord, an appreciable release of the contracture was noted. After allowing him to flex and extend the digit, we had concern for the motion of the finger distal to the joint worrisome for iatrogenic fracture of the proximal phalanx as a result of the manipulation. X-rays were obtained at that time, which confirmed the fracture and apex volar angulation at the base of the proximal phalanx (Fig. 2). The fracture appeared amenable to non-operative treatment. No further manipulation was performed, and the patient was placed into a protective splint for immobilization and advised to avoid any strenuous activity.

The patient returned 1 week later. He had no pain and minimal tenderness at the fracture site, with mild swelling. The skin remained intact. There had been no change in the position of the finger. X-rays showed acceptable alignment (Fig. 3). The patient returned 1 month later. X-rays again showed maintained alignment and signs of consolidation at the fracture site (Fig. 4). The patient is currently 4-month post-fracture, and no further intervention has been pursued to date. He is currently undergoing chemotherapy given his metastatic disease.

Discussion

A number of minor side effects and complications of collagenase injections have been described; most of which seem to occur at the time of injection or during the release. Before FDA approval of collagenase, Badalamente and Hurst [9] performed a randomized, double-blind, placebo-controlled trial. Adverse events were generally mild, localized to the injection site (i.e., edema, ecchymosis, skin laceration, and pruritus), and resolved within several weeks. There were no serious adverse events. There were no fractures noted in the original trial, but the

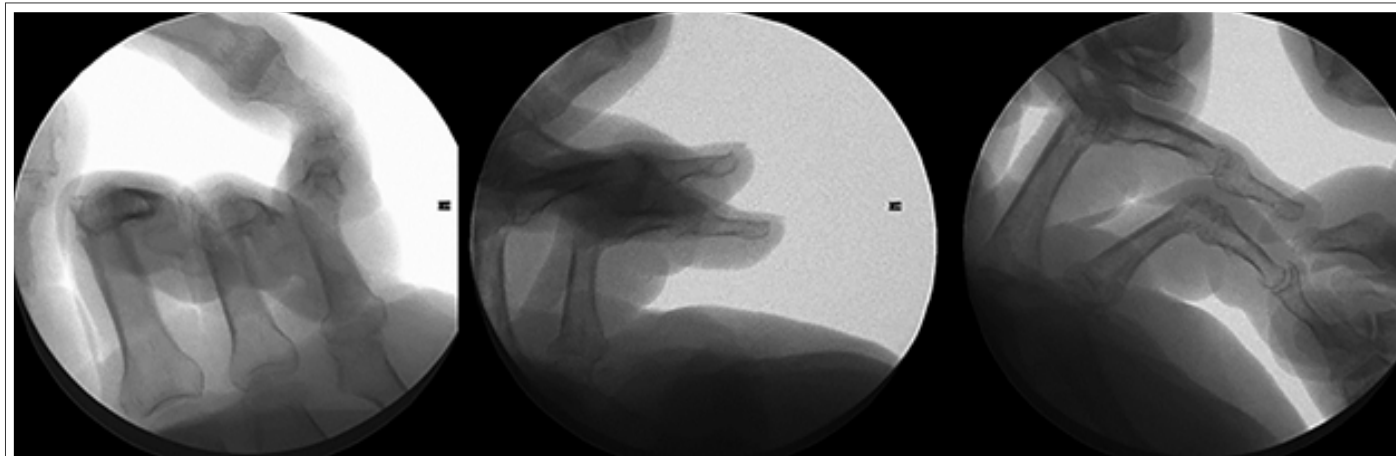


Figure 2: (a, b and c) Fluoroscopic images obtained at the time of digit widget application show severe metacarpophalangeal and proximal interphalangeal contractures.

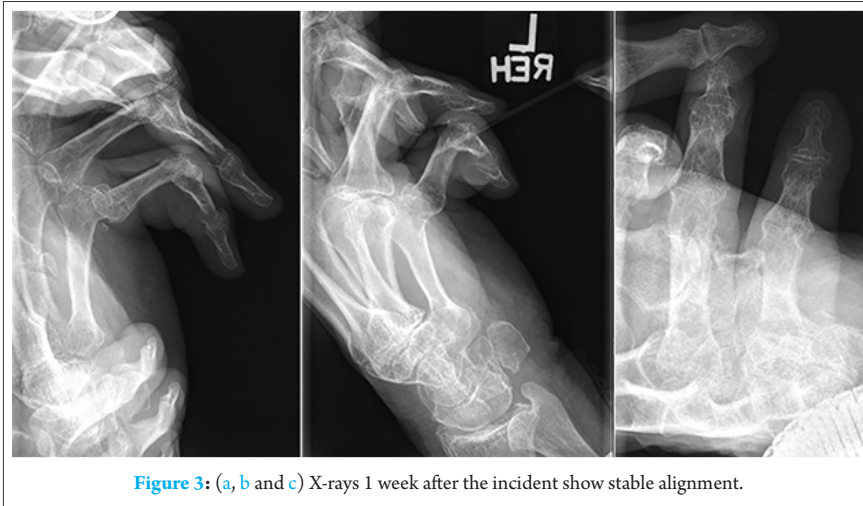


Figure 3: (a, b and c) X-rays 1 week after the incident show stable alignment.

releases on post-injection day #2 were attempted without anesthesia.

Further investigation by these authors and others [7] included 308 patients in a multicenter trial. A total of 741 injections (444 collagenase and 297 placebo) were performed. 96.6% of patients who received collagenase reported at least one treatment-related adverse event (most common were bruising, pain, or swelling) compared to 21.2% of those who received placebo ($P < 0.02$). Most adverse events were considered mild or moderate and resolved spontaneously within 10 days on average. There were two tendon ruptures and one case of complex regional pain syndrome in this group. There were no nerve injuries. No fractures were reported.

Warwick et al. [8] reported on a large series of patients treated with collagenase. These authors found a relatively high complication rate overall, involving 46% of patients. The most common complications were skin tears and blood blisters. No serious complications were identified.

Gilpin et al. [10] conducted a prospective, randomized controlled trial comparing collagenase injection in 45 patients to placebo in 21 patients. In this series, two serious adverse events were identified: One flexor pulley rupture requiring PIP arthrodesis and tenotomy and one cord proliferation and sensory abnormality requiring routine fasciectomy. There were no neurovascular injuries, tendon ruptures, or systemic allergic reactions.

A recent randomized controlled trial by Scherman et al. [11] compared 1 year outcomes following treatment by needle fasciotomy versus collagenase injection. There were no tendon ruptures or neurovascular complications in this series. Skin rupture was identified in 8% of the fasciotomy patients and 20% of the collagenase patients ($P = 0.13$).

Recent work by Peimer et al. reported long-term outcomes following collagenase injection [12, 13]. Approximately 49,000 injections were performed in the first 3 years following approval of CCH in the US. There were 26 tendon ruptures (0.05%), 1 pulley injury, and 1 ligament injury. Edema and contusion were the most common adverse events (12% and 11%, respectively). Most adverse events were mild to moderate, localized to the injection site, and transient. During the first 3 years, 193 of 643 patients experienced 370 adverse events (osteoarthritis, hypertension, and cataract). 31 patients (4.8%) experienced severe adverse events (atrial fibrillation, small-bowel obstruction, cerebrovascular accident, nephrolithiasis, and death), none of which were considered to be related to treatment.

To the best of our knowledge, this is the first reported case of a proximal phalanx fracture to occur at the time of release. In retrospect, this patient was a higher risk for this complication. He was diabetic and undergoing cancer treatment. Another possibility to consider is pathologic fracture, given his known history of metastatic thyroid cancer. He also had osteoporotic bone (in part due to disuse following two recent procedures) and was susceptible to fracture, especially given the severity of contracture. The senior author has done over 300 releases to date, and the amount of force with this release was no

greater than others. However, the point of leverage for the release was somewhat more distal than we wanted due to severity of the contracture. Ideally, the release of the MCP contracture is performed with a dorsally directed force at the middle of the proximal phalanx. However, since the MCP contracture was more severe in this patient, the point of dorsally directed force was at the PIP joint. This increased the moment arm and load at the eventual fracture site. Thankfully, the complication was appreciated at the time of incident. Had it not been, we fear that, if the patient had undergone our usual protocol of dynamic daytime splinting and nighttime extension splinting, we would have invited the risk of further pain and fracture displacement.

While fractures are a known concern in the setting of manipulation under anesthesia, the fracture sustained in the setting of attempted cord release in collagenase is notably different. When manipulating following needle aponeurotomy or collagenase injection, there is an anticipation of a cord release and less on joint contracture release. This shifts the focus on the release rather than amount of finger extension achieved.

The use of anesthesia to facilitate release was, in our opinion, a contributing factor. The senior author has performed many needle aponeurotomy procedures and has not experienced fractures in this setting, due in large part to the fact that they do not undergo anesthesia before release.

As we further reflect on this case, it illustrates the limitations of collagenase. In retrospect, clostridial collagenase should not have been considered for this patient. His age, comorbidities, bone quality, and severity of contracture may be considered too much to overcome with collagenase alone. However, having failed prior surgery times two, combined with his significant functional limitations, we were understandably desperate for something that may provide some improvement of function. Although we could have done two injections that day, we opted to first see how much improvement occurred with one and use that to determine whether further injections on other digits was warranted. Of course, no further injections have been pursued.



Figure 4: (a, b and c) X-rays 1 month after the incident again show stable alignment, with signs of early healing.

Conclusion

We hope that this case is helpful to the reader and can be used as a cautionary tale illustrating an additional potential complication associated with the use of clostridial collagenase. We recommend judicious use in elderly patients with severe contractures. It is important to appreciate the possibility of this complication at the time of cord release.

Clinical Message

Dupuytren's can be a challenging condition for both patients and surgeons. Needle aponeurotomy and collagenase injections are a reliable option in some patients, though the limitations and potential complications should be recognized.

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