Heterotopic ossification after local steroid injection

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SUMMARY

Pachydermodactyly (PDD) is a rare, benign disease associated with progressive swelling of the periarticular soft tissue of phalangeal hand joints typically treated with local steroid injections. We present a case of a 37-year-old man with PDD treated with local steroid injections. He later developed heterotopic ossification and para-articular calcifications in the injection sites. Heterotopic ossification is not associated with PDD nor is it a recognised complication of local steroid injections. This is the first case in literature of heterotopic ossification occurring after local steroid injection and brings to attention a new potential complication of a widely performed procedure.

BACKGROUND

Pachydermodactyly (PDD) is a rare, benign disease that causes soft tissue swelling resulting in thickening of the skin over the proximal interphalangeal (PIP) joints of fingers II–IV.[1] PDD classically occurs in adolescent men, typically as a consequence of repetitive mechanical trauma from tic-like behaviour.[2] This disease rarely extends to the metacarpophalangeal joints and does not affect the bone or synovium.[3] Diagnosis is based on clinical presentation, laboratory testing and imaging studies. It is typically treated with rehabilitation, which involves cessation of the repetitive movements that caused the lesions and intralesional steroid injections.[3]

There are several recognised complications after steroid injections such as pain at injection site, soft tissue atrophy, tendon and cartilage attrition, and while subcapsular calcification has been recognised as a complication after steroid injection, heterotopic ossification has never been reported. To the best of our knowledge, we describe here the first case of heterotopic ossification following local steroid injection. In addition, we aim to review the management of PDD, a rare but important differential diagnosis for plastic surgeons to consider in a hand patient presenting with joint swelling.

CASE PRESENTATION

We present the case of a 37-year-old right-hand dominant male engineer and concert pianist with no significant medical history who presented with enlargement surrounding the PIP joints of his right middle and ring fingers. The enlargement around his middle finger PIP joint was painless, but its size sometimes interfered with his piano playing. His ring finger PIP joint was less significantly affected and asymptomatic. The patient endorsed a history of cracking his knuckles and gnawing on the sides of his right middle and ring fingers since he was a teenager up until 2 years prior to presentation. The enlargement began in his teenage years and stabilised in his early 20s. He denied any morning stiffness of his joints or reduced joint function and had no medical, surgical or substance use history.

Physical examination of the right hand was significant for localised swelling of the soft tissues surrounding the middle finger PIP joint and to a lesser degree the ring finger PIP joint (figures 1 and 2). The joints and soft tissues were soft, non-erythematous, non-tender and stable to stress. There was no pain on axial loading. The patient had full active range of motion with global hyperextensibility and was neurovascularly intact.

INVESTIGATIONS

Plain films of the right hand (figure 3) were obtained and showed localised soft tissue swelling surrounding the aforementioned joints but no bony abnormalities. Routine laboratory values that the patient had previously obtained including complete blood count (CBC), comprehensive metabolic panel (CMP) and thyroid stimulating hormone (TSH) were unremarkable.

DIFFERENTIAL DIAGNOSIS

A diagnosis of PDD was made given the characteristic findings of painless soft tissue swelling and lack of signs consistent with inflammatory or bony disorders. Plain films of the right hand (figure 3) were obtained and showed localised soft tissue swelling surrounding the aforementioned joints but no bony abnormalities which is consistent with PDD. In addition, routine laboratory values that the patient had previously obtained including CBC, CMP, and TSH were unremarkable which is consistent with PDD.

TREATMENT

While the patient had stopped engaging in repetitive finger gnawing and knuckle cracking for 2 years, he was cautioned on the importance of continued abstinence from these repetitive movements to prevent worsening of his disease. After discussing surgical and non-surgical options, including the risks and benefits of local steroid injection, the patient elected to proceed with local steroid injection. 0.5 cc of Kenalog-40 (triamcinolone acetonide injectable suspension 40 mg/mL) was injected using a 25-gauge needle into the dermal and capsular layers of each side surrounding the right middle finger PIP joints. The patient reported satisfaction and improved mobility after local steroid injection, which was performed three times over a period of 6 months. Follow-up appointments revealed thinning of the injected region and improved range of motion. The patient denied any pain or concerns.

Figure 1

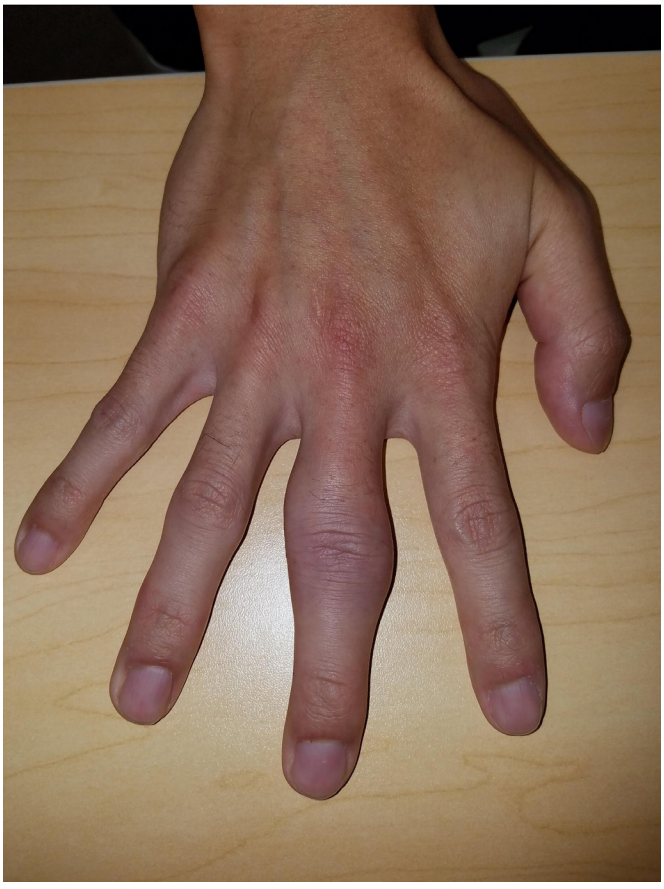

Figure 1 Dorsal view of right hand. This image shows the localised swelling of the soft tissue around the proximal interphalangeal joint of the middle finger.

Figure 2

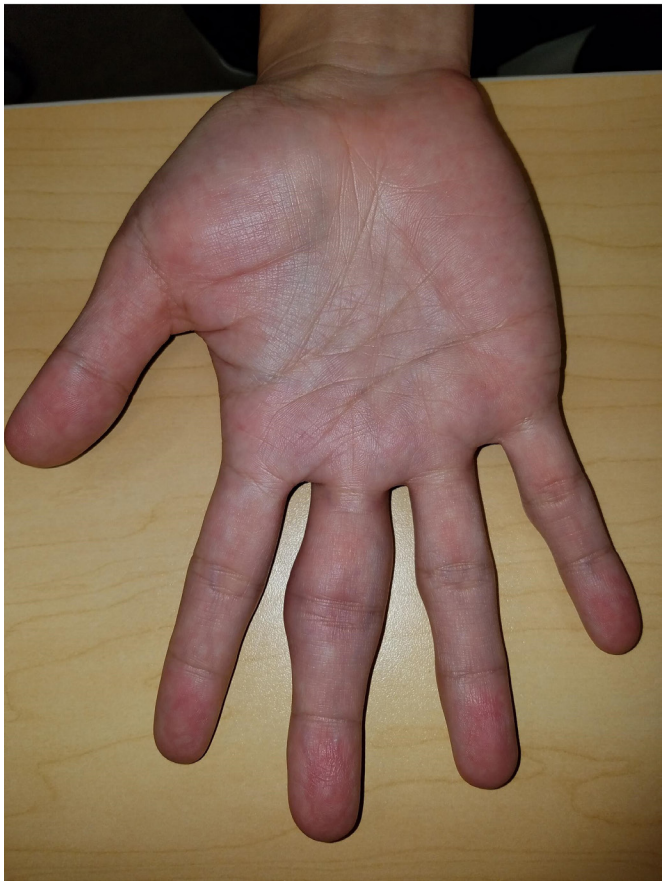

Figure 2 Palmar view of right hand. This image shows the localised swelling of the soft tissue around the proximal interphalangeal joint of the middle finger.

OUTCOME AND FOLLOW-UP

However, 2 years after his last injection, the patient returned to clinic complaining of decreased range of motion and pain of the right middle finger PIP joint. He reported feeling a hard lump proximal to his ulnar middle finger PIP joint 6 months after his last steroid injection, which gradually enlarged and led to pain that limited his ability to flex his finger. He denied any trauma to his right hand that preceded these symptoms. Examination revealed increased soft tissue enlargement around his right middle finger PIP joint and stable soft tissue enlargement around the right ring finger PIP joint. He was tender to palpation around the middle finger PIP joint and flexion was limited by pain.

Figure 3

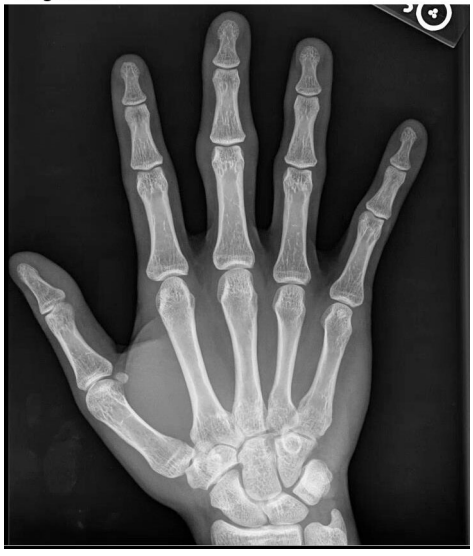

Figure 3 Initial X-ray of right hand. X-ray showed localised soft tissue swelling surrounding the aforementioned joints but an absence of any bony abnormalities.

X-ray reviewed new para-articular calcifications and heterotopic ossification of the ulnar proximal phalanx soft tissues with persistent soft tissue swelling and skin thickening of the middle finger (figure 4). The diagnosis of heterotopic ossification was made radiographically as the bone in this patient's soft tissue is not amorphous calcification as seen in calcium pyrophosphate dihydrate crystal deposition disease or tumorous calcinosis. Rather, radiographically, the calcific density in the soft tissue appears like mature bone and is corticated with central cancellous appearance. Management of heterotopic ossification was discussed with the patient and treatment with indomethacin was begun.

The patient returned after 8 weeks of treatment with indomethacin and noted decreased pain and stiffness though he was still unable to make a fist. He also noted interval increase in size of the ulnar proximal phalanx mass. Notable changes in the examination were a decrease in range of motion of the middle finger PIP joint 0–90 degrees (0–135 degrees other digits). X-ray

Figure 4

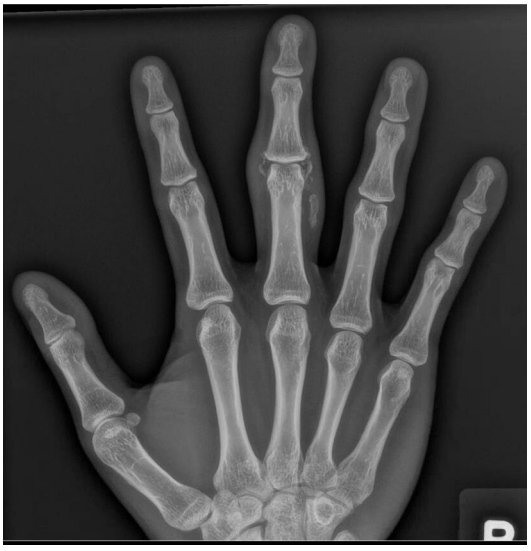

Figure 4 Post-injection X-ray of right hand. X-ray showed new para-articular calcifications and heterotopic ossification with characteristic central lucency of the ulnar proximal phalanx. Soft tissues showed persistent soft tissue swelling and skin thickening of the middle finger.

Figure 5

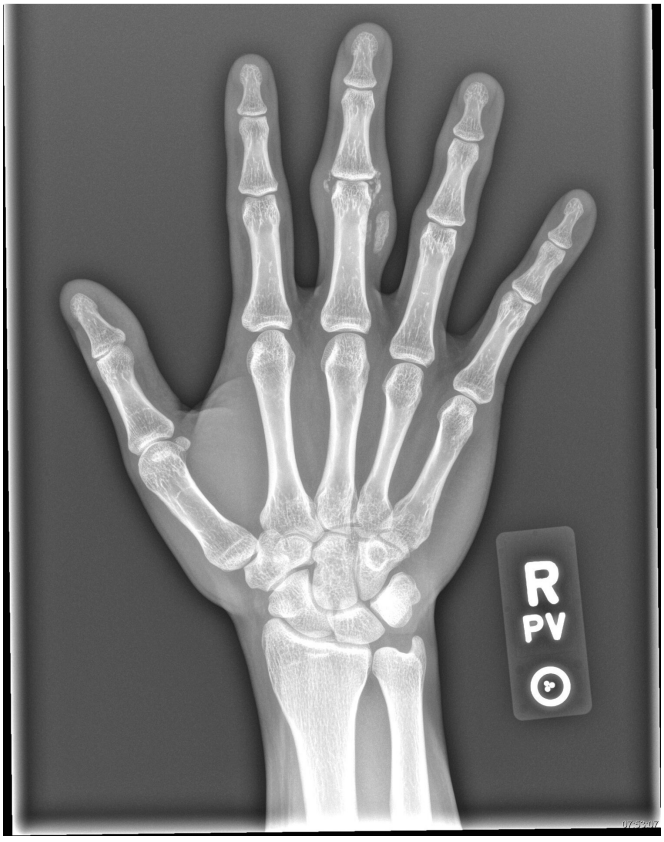

Figure 5 X-ray of right hand after 8 weeks of treatment with indomethacin. Redemonstration of long finger soft tissue prominence centred at the proximal phalanx and proximal interphalangeal joint, suggestive of pachydermodactyly, with capsular calcifications at the proximal interphalangeal and heterotopic ossification in the soft tissues ulnar to the proximal phalanx. There is increase in size of the heterotopic ossification along the ulnar aspect of the proximal phalanx in comparison with the prior radiographs.

revealed increased size of heterotopic ossification and persistent soft tissue swelling (figure 5). Excision of the heterotopic ossification was offered to the patient; however, the patient declined surgery and opted for continued indomethacin treatment.

DISCUSSION

PDD is a rare disease associated with digital fibromatosis resulting in deposition of abnormal collagen in the periarticular dermis. The pathogenesis in our patient most likely involved his repetitive gnawing of his fingers and knuckle cracking throughout his adolescence which is typical of this diagnosis. The leading clinical sign of PDD is progressive, painless swelling of PIP joints. The affected skin is thick and firm but not irritated or erythematous. In addition, the joints are non-tender, there is no effusion nor limitation of function. Laboratory studies reveal normal values for all reported cases in the literature including inflammatory markers, thyroid function and autoantibodies. Imaging with plain films and ultrasound reveal soft tissue hypertrophy but no joint disease. Calcifications and bony abnormalities are not associated with PDD. Given the rarity of the condition, there are limited data to guide therapy for PDD. Treatment begins with cessation of the mechanical stimulation the patient experiences, which may involve pharmacological management of any underlying psychiatric conditions. In select cases, surgical excision is performed to remove the lesions, but most commonly, the treatment involves local steroid injections.[1–3]

Local corticosteroid injections are used for a variety of conditions, including rheumatoid arthritis, gout and bursitis. Side effects of steroid injections include pain, tissue atrophy, acne and skin hypopigmentation.[4 5] Rare cases of Cushing's syndrome and abscess formation after corticosteroid injections have been reported in the literature.[6] While recognised complications of local steroid injections include pain at injection site, soft tissue atrophy, tendon and cartilage attrition, and pericapsular calcification, heterotopic ossification has not been previously described.

Heterotopic ossification is treated with physical therapy, Nonsteroidal anti-inflammatory drugs (NSAIDs) or radiation therapy. The risk of complications after local steroid injection can be mitigated by combining injections with other treatments or using lower doses of up to 40 mg per site. This case brings to attention a new potential complication of a widely performed procedure. Heterotopic ossification can occur after surgery,

PATIENT'S PERSPECTIVE

I first noticed the development of this swelling in my fingers in my teenage years and it stabilised in my 20s. I did some research online and found it consistent with pachydermodactyly. I had a habit of cracking and gnawing those affected fingers which I have since stopped. The swelling wasn't painful and my range of motion was not limited but it sometimes affected my piano playing. I therefore sought treatment with steroid injections which I had read about. At first the swelling improved with the steroid treatments and things were going well. I then noticed some hardened swelling which was not there before associated with some pain. The doctors offered me medical and surgical treatment options but as my symptoms were controllable with the NSAIDs I opted not to undergo surgery. I have some regret now undergoing the steroid shots as it has lead to this unforeseen problem and my symptoms worsened. However, it is manageable and does not affect my daily functions including performing piano much.

Learning points

- Pachydermodactyly (PDD) is a rare disease associated with digital fibromatosis resulting in deposition of abnormal collagen in the periarticular dermis.
- The leading clinical sign of PDD is progressive, painless swelling of proximal interphalangeal joints. The affected skin is thick and firm but not irritated or erythematous. In addition, the joints are non-tender, there is no effusion nor limitation of function.
- Treatment begins with cessation of the mechanical stimulation the patient experiences, which may involve pharmacological management of any underlying psychiatric conditions. In select cases, surgical excision is performed to remove the lesions, but most commonly, the treatment involves local steroid injections.
- This is the first case in literature of heterotopic ossification occurring after local steroid injection and brings to attention a new potential complication of a widely performed procedure.

trauma, spinal cord injury, hypoxia or thermal injury. The pathogenesis involves the conversion of primitive mesenchymal cells into osteogenic precursor cells, resulting in local ossification of non-skeletal tissues.[7] This results in decreased range of motion, erythema, soft tissue swelling, fever and pain that is often challenging to distinguish from cellulitis or osteomyelitis. The typical radiological appearance is circumferential ossification with a lucent centre.[8] Some preventative measures have been developed to decrease the chances of heterotopic ossification. Coumadin may be prescribed because it inhibits the activity of vitamin K, which is involved in bone development. NSAIDs may also be used by blocking the prostaglandin E_2 activity essential for ossification.[7]

Treatment of heterotopic ossification involves passive range-of-motion exercises, local radiation therapy and NSAIDs.[9] Surgical resection is often required to restore range of motion. However, if surgery is delayed after maturation of the lesion, failure and recurrence rates are high.[8]

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