Painful Proximally Oriented Large Heterotopic Spur Formation in an Active Adult With a Nontraumatic Amputation

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Heterotopic ossification (HO) is excess bone growth in soft tissues, typically juxta-articular and interfascicular, with varying incidence. This excess bone growth has been well-documented in cases of traumatic amputation but less frequently observed in cases of nontraumatic amputation. Symptomatic heterotopic ossification usually includes pain during prosthetic use with management involving prosthetic adjustments for comfort. This atypical case highlights a patient with a nontraumatic amputation and a proximal-oriented large spur formation that was not painful with ambulation but with doffing his prosthesis.

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Heterotopic ossification (HO) is excess bone growth in soft tissues, typically juxta-articular and interfascicular, with varying incidence. Usually, it develops outside the borders of normal periosteum and has a vascular supply.¹ It can be contiguous with the skeleton; however, it does not involve the periosteum, and once matured, it contains cancellous and lamellar bone, bone marrow, and vessels.² While the pathogenesis is not completely understood, it is believed that dormant osteoprogenitor stem cells within soft tissue are triggered to differentiate into osteoblasts, which ultimately leads to bone formation.³ This bone growth may or may not be symptomatic, and symptoms include pain during prosthetic use.⁴ Typically, symptoms can be alleviated through prosthetic adjustments for a more comfortable fit.⁴ Heterotopic ossification can create a pressure-sensitive area that requires multiple modifications of the prosthesis.⁵ Conservatively, nonsteroidal anti-inflammatory drugs and bisphosphonates are used to manage progression, along with therapy to preserve range of motion.³ External beam radiation therapy is also available, but at a more significant cost.³ A minority of patients may need surgical excision of the ectopic bone to relieve discomfort; however, this requires mature bone.³ Maturation of bone occurs approximately 12 to 18 months after injury.⁴ During amputation, the surgeon should consider resecting the bone such that periosteum covers the distal end.⁵ If the bone is not well covered with periosteum, a spikelike HO might develop in the soft tissue and cause pain and irritation.⁵ The following case highlights an atypical presentation of heterotopic spur formation that physicians should be mindful of when evaluating pain in the residual limb.

Report of Case

A 58-year-old man taking warfarin with a history of atrial fibrillation and peripheral vascular disease presented to an emergency department in Italy in June 2015 with shortness

of breath. The patient was in acute respiratory failure and required intubation, mechanical ventilation, and prolonged vasopressor support for septic shock. The patient needed 15 days of extracorporeal membrane oxygenation for support, as his respiratory function was improved with antibiotics. After vasopressor therapy was discontinued, hypoperfusion was found in both lower extremities and required a right transtibial amputation and a left transfemoral amputation. The left lower extremity required an additional debridement for surgical site infection and a prolonged antibiotics course. He returned to the United States in September 2015.

The patient received inpatient preprosthetic training in September 2015 and inpatient prosthetic training in February 2016. He demonstrated proficiency and independence in ambulation and activities of daily living with his prostheses. At that time, he did not have any atypical pain, nor was there any evidence of HO.

The patient was highly active with his tight-fitting prostheses. However, he began to complain of severe pain in the left residual limb, not with weight-bearing, but with the removal of the prosthetic socket from his residual limb. Upon follow-up with a physiatrist, a radiograph revealed a 3.9×2.3 -cm calcified lesion at the medial aspect of the femoral shaft of his residual left femur. This heterotopic spur was oriented proximally. He was referred to a plastic surgeon, who removed the spur and noted that the spur had a significant blood supply and a separate bursa around the spur. The pathology report noted “hemorrhagic soft tissue” and bone fragments measuring $8.0 \times 5.0 \times 3.5$ cm in aggregate. Postoperatively, the patient was pain-free after his surgical wound healed and continued to be active and pain-free with his prostheses.

Discussion

The functional changes that accompany HO are not very well described in the literature, but are thought to result from the transformation of dormant osteoprogenitor stem cells into osteoblasts, which leads to bone

formation.³ Various humoral, neural, and local factors must come together to create the necessary environment for HO to occur.⁷ When the periosteum covering bone that is retained is stripped, ectopic bone formation can occur and results in a simple bone spur.⁸ However, what makes this case unique is the extension of the bony growth into the adjacent soft tissue, as well as its irregular shape.

An extensive literature review revealed a single case report of HO in the residual lower limb in an adult non-traumatic amputee.⁹ To our knowledge, HO in an adult nontraumatic amputee is a rare occurrence. In the current case report, the patient’s presentation of pain with doffing the prosthesis, along with the proximal orientation of his heterotopic spur, appears to be unique. Dudek et al⁵ described 2 separate case reports of patients with HO that caused significant residual limb pain after amputation. The first case described a 39-year-old woman who had a left transfemoral amputation after a traumatic event. Radiologic examination at 8 months after amputation revealed a bone growth along the femoral shaft extending from the amputation site to adjacent soft tissues. Release of the sciatic nerve as well as adjustments to her prosthetic socket did not help relieve the magnitude of her limb pain. Surgical excision was performed after amputation, providing significant relief of her pain along with increased functional use of the prosthesis. The second case described a 59-year-old man who also underwent a left transfemoral amputation after a traumatic injury. His radiograph revealed bony spurring at the distal end of the femur as well as a bony formation in adjacent soft tissues. The patient was limited in movement, as weight-bearing on the amputated limb caused him pain. His pain resolved after a new prosthetic socket provided pressure relief at the distal lateral femur.

Kömürçü et al¹⁰ described a 35-year-old man who also had a transfemoral amputation following a traumatic injury. Sonographic as well as radiographic imaging of the right knee revealed a bony spur on the tibia. There was no mention of the treatment of this patient’s condition because the report only focused on

Figure.

Radiograph of the patient's residual limb in June 2017, 2 years after initial hospitalization for acute respiratory failure.

diagnosis of the bony spur. Melcer et al⁶ reported occurrences of HO in a small series of veterans from the Afghanistan and Iraq wars who had an amputation. Roughly one-third of patients with at least moderate HO showed no adverse symptoms. This finding highlights that not all HO cases are generators of pain and can be asymptomatic. Other cases showed that HO can affect pressure-sensitive areas of the amputated limb, with adverse symptoms prompting management with prosthetic adjustment or even surgical excision. Furthermore, a patient care issue they explored was the potential beneficial use of HO in prosthetic fitting in patients with transfemoral amputations. When the bony outgrowth expands to surround the distal portion of the amputated bone, it becomes useful as an anatomical support structure that the prosthetist can use to improve the fitting of the residual limb into the prosthetic socket.

We hypothesize that our patient's large spur formation with characteristics of HO was related to his high activity in a tight-fitting prosthesis while taking

warfarin, which created local trauma that perhaps led to hematoma and calcium deposition. Although bony overgrowth can occur in a skeletally mature patient, our radiologic findings point to HO as the cause, since the bony spike was not contiguous with the periosteum of the residual limb.

Conclusion

Heterotopic ossification can be a pain generator in adult patients with nontraumatic amputations. It remains to be determined whether the pathogenesis differs between populations with traumatic and nontraumatic amputations.

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