Case report

Myositis ossificans of the longus coli muscle following cervical spine fracture-dislocation

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Background: Upper cervical spine fractures are traumatic injuries typically associated with high-energy trauma and have a high morbidity and mortality rate. We describe a case of upper cervical spine fracture occurring due to high-energy trauma that resulted in significant myositis ossificans (MO) of the longus coli muscle.

Study Design: Case Report and literature review.

Methods: Retrospective review of medical records.

Results: The patient was treated non-operatively for the neck rotation and MO of his longus coli muscle and had gradual improvement of symptoms.

Conclusions: To our knowledge, this is the first report of symptomatic MO of the longus coli following traumatic cervical spine fracture in an adult. Treatment with supportive measures appears to achieve satisfactory outcome.

Keywords: Cervical, Fracture, Upper cervical spine fracture, Myositis ossificans, Torticollis, Longus coli

Introduction

Myositis ossificans (MO) has been well described in the literature following trauma.¹⁻⁶ MO is a localized and self-limited condition with a proliferation of fibroblasts and heterotopic bone formation in the soft tissues and muscles. It typically occurs several weeks after a traumatic injury to the musculoskeletal system, causing the body to deposit calcium, bone, and occasionally cartilage in the soft tissues and musculature.^{1,7} Symptoms typically resolve over time as partial or full bony resorption occurs. Surgery is rarely indicated.

Case Report

A 30-year-old male presented as a Level-1 trauma following a high-speed motorcycle accident while intoxicated. The patient was diagnosed with a C2 fracture-dislocation with associated C1 ring fracture and occipital condyle avulsion fracture (Fig. 1). The patient was determined to have a C2 incomplete spinal cord injury. Several other orthopaedic injuries and long-bone fractures were present including a right open grade III segmental tibia fracture as well as a traumatic left above-elbow amputation. The patient underwent

multiple orthopaedic surgeries and spine surgery for his injuries during the first 24 hours after presentation.

The patient underwent a C1 to C4 posterior instrumented spinal fusion for his C2 fracture-dislocation. Copious cerebrospinal fluid (CSF) was noted in the beginning of the procedure but no decompression was performed and no identifiable durotomy was noted during surgery. The patient's spine surgery was uneventful, and he was returned to the ICU in stable condition. Six weeks post-operatively, the patient's dense incomplete spinal cord injury had not improved. Radiographs showed acceptable alignment of the spine and appropriate position of the spinal implants (Fig. 2).

The patient was poly-traumatized, critically ill, and remained hospitalized for more than 3 months. At approximately 12 weeks after his initial cervical spine surgery, he began to complain of increasing neck pain and his head appeared rotated and bent to one side, as in torticollis (Fig. 3). A computed tomography (CT) scan demonstrated appropriate positioning of the spinal implants and revealed MO of the right longus coli muscle without any apparent involvement in the strap muscles or sternocleidomastoid muscle. This was believed to be the source of the torticollis (Fig. 4). The patient was treated with passive stretching, nonsteroidal anti-inflammatories (NSAIDs), antispasmodics, and a rigid cervical collar as tolerated. The patient also experienced persistent

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Figure 1 (a) T2 weighted sagittal MRI and (b) coronal CT scan demonstrating C2 fracture-dislocation and right occipital condyle avulsion fracture.

headaches, and a head CT demonstrated chronic subdural hematomas. A neurosurgical consultant suggested that the cause may be persistent CSF leak at the site of the fracture-dislocation. A percutaneous blood patch was performed, and the headache diminished. At 8 months after surgery, the patient noted continued neck pain and

neuropathic pain, however, he stated that the pain was decreased compared to the initial post-operative period.

Literature Review

The free search engine, PubMed, maintained by the United States National Library of Medicine (NML) at

Figure 2 (a) AP and (b) lateral radiographs 4 weeks after surgery demonstrating appropriate alignment and implant positioning.

Figure 3 (a) AP and (b) lateral radiographs 12 weeks after surgery demonstrating intact implants but with coronal and sagittal malalignment.

the National Institutes of Health was searched in April 2015 and again in January of 2016. The database was searched using the key words “cervical fracture,” “cervical spine injury,” “upper cervical spine,” “myositis

ossificans,” and “myositis ossificans traumatica.” Eight results were found with the above criteria, only 3 of which were related specifically to the cervical spine. Four search results were related to the rare genetic

Figure 4 (a) Coronal and (b and c) axial CT scans demonstrating significant myositis ossificans at several levels in the right longus coli, in particular C5 and C6.

disease Fibrodysplasia Ossificans Progressiva, or FOP. The 3 papers dealing with MO in the cervical spine were reviewed. No papers pertaining to MO in the anterior cervical spine in adult patients following trauma were identified. An additional search was performed, this time substituting “heterotopic ossification” for the “myositis ossificans” keywords. Only one result was identified and this paper was reviewed extensively.⁸ This case report did not discuss heterotopic ossification or myositis ossificans.⁸ To our knowledge, MO occurring in the longus coli muscle in an adult patient following cervical spine fracture has not been reported.

Discussion

MO is a phenomenon typically observed after trauma to skeletal muscle in which there is abnormal calcification of the soft tissues. MO was described as early as 1905 and involvement in the appendicular skeleton has been extensively reported in the literature.^{1–6,9} Most commonly, MO affects the larger muscles of the body including the thigh, upper arm, and forearm. A similar condition and pathologic process that affects muscles around joints of the body is called heterotopic ossification, or HO. This condition commonly affects the elbow, knees, hips, and shoulders. The exact cause and pathophysiology of MO and this similar condition HO is not fully understood. There appears to be a genetic predisposition after muscle trauma, with specific signaling pathways and growth factors including Bone Morphogenetic Protein 4 (BMP-4), BMP-2, BMP-1, Transforming Growth Factor Beta-1, among others.^{10–15}

There have been several definitions and classification schemes for MO dating back to the 1920s, but Kransdorf *et al.* has recently defined it as a “benign, solitary, self-limiting, ossifying soft-tissue mass typically occurring within skeletal muscle.”^{9,16} The classification system by Noble includes 3 types, including Atraumatic Myositis Ossificans Circumscripta, Traumatic Myositis Ossificans Circumscripta (TMOC), and Myositis Ossificans Progressiva, a rare genetic disorder affecting 1 in 2 million people and now referred to as Fibrodysplasia Ossificans Progressiva.^{2,3,9,17–20}

Findlay *et al.* described a case of MO in an 8-year-old child in the posterior C1–C2 spine musculature 4 years after trauma.²¹ The authors repeated a CT scan and found the patient’s lesion to have spontaneously resolved 18 months after it was discovered. The authors concluded that MO is likely underreported and should be suspected in cases of persistent neck pain following a cervical spine injury. Harmon *et al.* described a case of TMOC in a 10-month-old female infant after nonaccidental trauma resulting in a C5

compression fracture.²² The authors described the appropriate work-up and evaluation needed to distinguish MO from other more significant pathologic processes. Lee *et al.* described a case of MO in the posterior musculature of a 26-year-old patient several weeks after receiving acupuncture therapy.²³ Non-operative management was chosen after CT-guided biopsy confirmed the diagnosis.

In the case presented here, the MO formation appeared between 6 to 12 weeks after the patient’s initial injury, typical of when MO has been reported to occur. The authors believe that the MO of the longus coli led to muscle contracture and that this process affected head position. Specifically, the right-sided calcification acted as a tether (Fig. 4). The patient’s incomplete spinal cord injury and lack of muscle strength likely also contributed to his inability to straighten his head and neck, despite positioning efforts of the clinical team. Advanced imaging confirmed appropriate position of the posterior spinal implants and no other cause for the patient’s head position could be determined. The patient experienced partial relief of both pain and the malpositioning, or torticollis, after conservative measures.

Conclusions

MO can occur after any traumatic event and has been commonly reported in many muscle groups. We report a case of MO of the longus coli following severe fracture-dislocation of the cervical spine. MO of the longus coli appeared to contribute to the patient’s torticollis and difficulty with head position. Treatment consisting of conservative management including NSAIDs, stretching, supportive collar wear, and analgesics appeared to provide improved comfort and decreased torticollis positioning by 8 months after the index trauma.

Disclaimer statements

Contributors None.

Funding None.

Conflicts of interest None.

Ethics approval Relevant financial activities outside the submitted work: Fellowship Support by AO Spine.

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