JSES Reviews, Reports, and Techniques 4 (2024) 86–90

Contents lists available at ScienceDirect

JSES Reviews, Reports, and Techniques

journal homepage: www.jsesreviewsreportstech.org

Glenohumeral joint auto-fusion in a morbidly obese patient intubated for severe COVID-19 infection

Viviana M. Serra López, MD, MS[a], Adnan N. Cheema, MD[b], Sarah R. Blumenthal, MD[a], John G. Horneff III, MD[a], G. Russell Huffman, MD, MPH[c,*]

[a]Department of Orthopaedic Surgery, Hospital of the University of Pennsylvania, Philadelphia, PA, USA
[b]Department of Orthopaedic Surgery, Mayo Clinic, Rochester, MN, USA
[c]Advent Health, Rothman Institute, Orlando, FL, USA

ARTICLE INFO

Keywords:
COVID-19
Glenohumeral auto-fusion
Heterotopic ossification
Reverse total shoulder arthroplasty
Revision reverse total shoulder arthroplasty
Shoulder hemiarthroplasty

Heterotopic ossification (HO) is characterized by the formation of bone in extraosseous sites. Its etiology is multifactorial and typically associated with an insult that results in an inflammatory cascade involving prostaglandin release.[12] Heterotopic ossification (HO) formation is most commonly reported following significant brain or spinal cord trauma.[5,21] While some patients are asymptomatic, others can experience significant bone formation resulting in severe pain and dysfunction. Several case reports have documented HO formation following COVID-19 infection, with an incidence of 19% in those with severe illness.[11,20] Heterotopic ossification (HO) of the glenohumeral joint is a rare occurrence and has been described following shoulder arthroscopy and total shoulder arthroplasty.[14] The occurrence of glenohumeral HO resulting from trauma or illness is rare and limited to case reports.[16,17]

Management of HO depends on its clinical manifestations. Prevention is the ideal strategy for high-risk circumstances, such as spinal cord injury and head trauma.[21,22] While prevention measures are not possible, surgical excision may be required if HO is severe enough to affect function. This is typically performed once the HO is deemed mature, or sufficiently constituted for excision.[1] In extreme cases, joint salvage procedures may be indicated to optimize function. However, joint salvage is further complicated by the status of the soft tissues and surrounding neurovascular structures. In the case of reverse total shoulder arthroplasty (rTSA), dislocation has been described as the most common complication and is often multifactorial.[4] It is important to consider the location of the HO during surgical planning and how this can contribute to instability when determining surgical options.

We present the case of a 52-year-old male patient who developed HO resulting in loss of range of motion in his left shoulder following severe COVID-19 infection. The patient required surgical HO excision and rTSA to restore function, followed by a revision to hemiarthroplasty (HA) due to subsequent instability.

Case presentation

A 52-year-old right hand–dominant male presented to the orthopedic clinic for evaluation of a 1-year history of left shoulder pain and stiffness. Medical history was notable for morbid obesity (body mass index 55 kg/m^2), hypertension, and obstructive sleep apnea. One year prior, he had been diagnosed with acute respiratory distress syndrome due to COVID-19, resulting in prolonged immobilization beginning on the day of admission when he was intubated. During the first 5 days of hospitalization, the patient was placed in the prone position multiple times to optimize his ventilation. During the sixth day of his hospitalization, his respiratory status improved to the point that he could tolerate being supine; however, he continued to require intermittent paralytics, high-dose sedation, high levels of positive-end expiratory pressure, and high fractions of inspired oxygen. On day 14 of his hospitalization, he was transitioned to pressure support mode on the ventilator as his mental and respiratory status improved. A tracheostomy was performed on day 20. At that time, the patient complained of severe left shoulder pain and left-sided weakness and swelling. He was placed on a heparin drip for management of a left internal jugular vein thrombosis. Neurology was consulted and a subsequent stroke workup was found to be negative.

Institutional review board approval was not required for this case report.

*Corresponding author: G. Russell Huffman, MD, MPH, Advent Health, Rothman Institute Florida, Innovation Tower, Suite 11, 265 E. Rollins St, Orlando, FL 32804, USA.
E-mail address: grussellhuffman@outlook.com (G.R. Huffman).

https://doi.org/10.1016/j.xrrt.2023.09.011

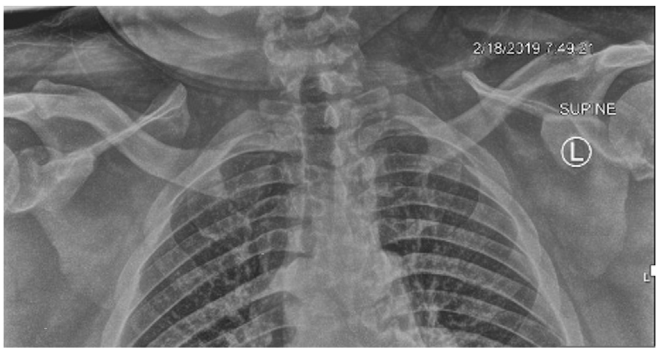

Figure 1 Antero-posterior (AP) of the chest obtained 1 year before hospitalization is presented to evaluate the partially seen left glenohumeral joint. There is no evidence of degenerative changes or ectopic ossification in this limited view.

Physical therapy was initiated during hospitalization to address his overall deconditioning and lack of left shoulder range of motion. Thirty days after admission, the patient was discharged to a long-term acute care hospital where he continued to participate in therapy. However, he had increased stiffness and pain in his left shoulder during the subsequent months. Recovery was also complicated by left foot drop with subsequent electrodiagnostic studies demonstrating a complete left peroneal nerve palsy. He reported no symptoms in his shoulder before his COVID-19 diagnosis. There were no prior dedicated glenohumeral radiographs available for comparison, however chest radiographs from 1 year before hospitalization showed no abnormalities in his left glenohumeral joint (Fig. 1).

At the time of his first examination in our orthopedic clinic, the patient's glenohumeral joint was fixed at 45 degrees of internal rotation with zero degrees of shoulder flexion and zero degrees of abduction. Presumably, this was the position of his arm during prolonged periods of prone intubation. Evaluation of radiographs

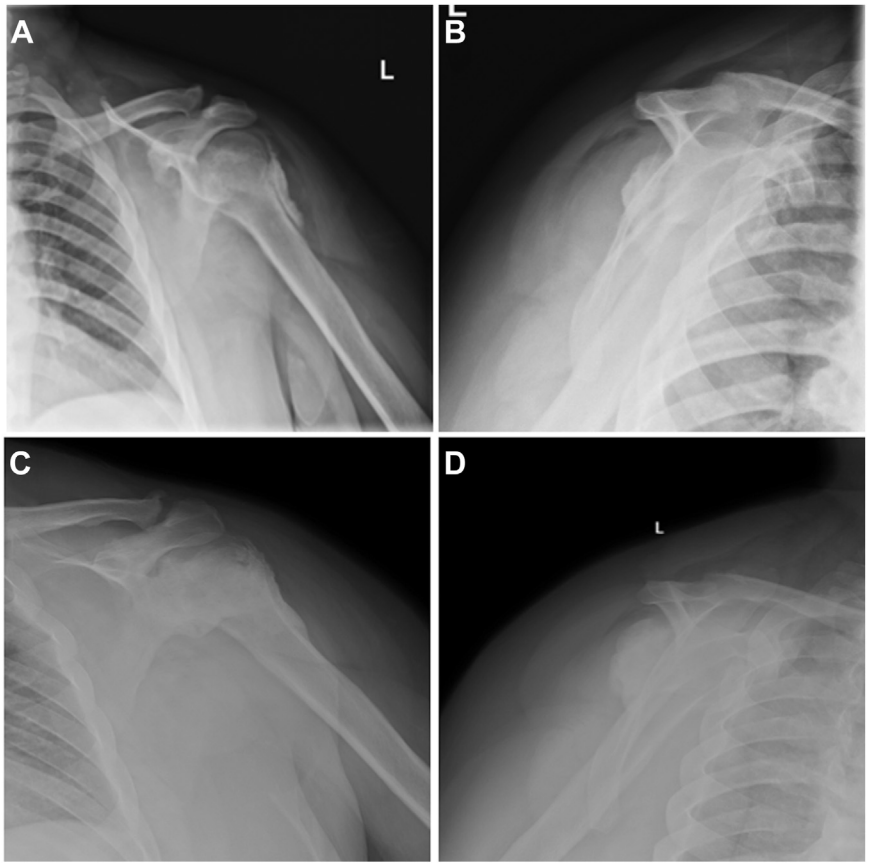

Figure 2 (**A**) AP and (**B**) scapular-Y radiographs of the left glenohumeral joint 4 months after hospitalization. (**C**) AP and (**D**) scapular-Y radiographs of the left glenohumeral joint 1 year after hospitalization. *AP*, antero-posterior.

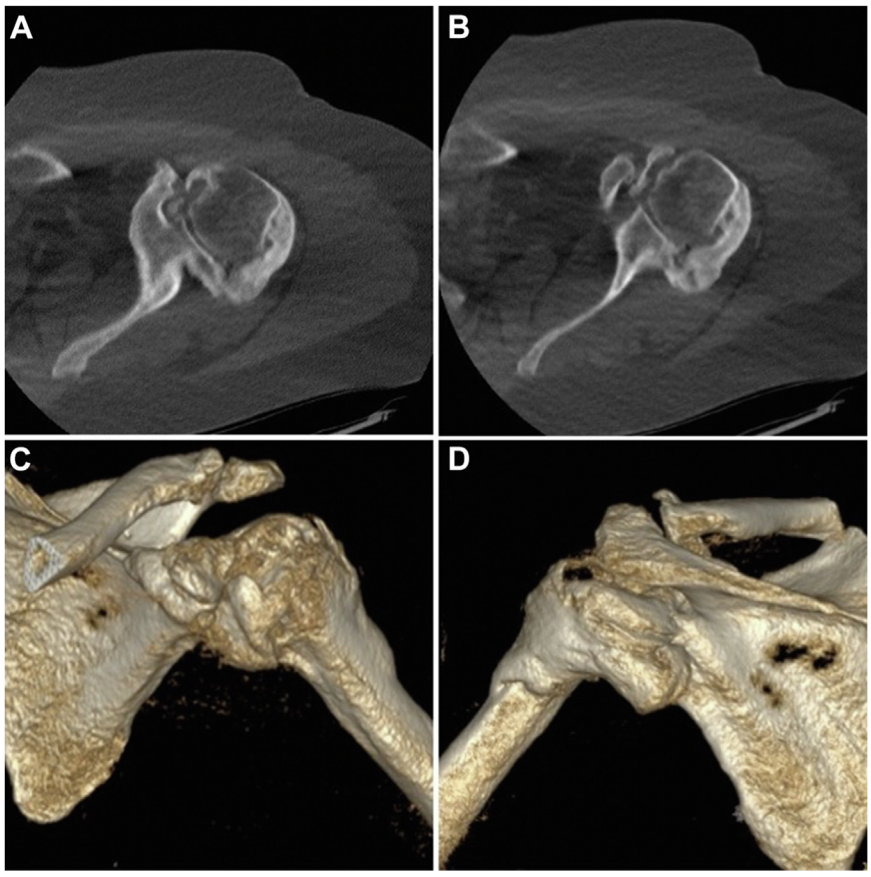

Figure 3 (**A** and **B**) Axial cuts of a left shoulder noncontrast computed tomography scan obtained 12 months after hospitalization showing glenohumeral heterotopic ossification with bridging exostoses from the humeral head to the glenoid resulting in complete absence of glenohumeral motion. Figure A also shows bone formation around the insertion of the subscapularis on the lesser tuberosity. Glenohumeral joint space is noted to be preserved. Given the maturity of the bone on imaging, a bone scan was not pursued. (**C**) 3D reconstruction showing glenohumeral joint fusion anteriorly and (**D**) posteriorly.

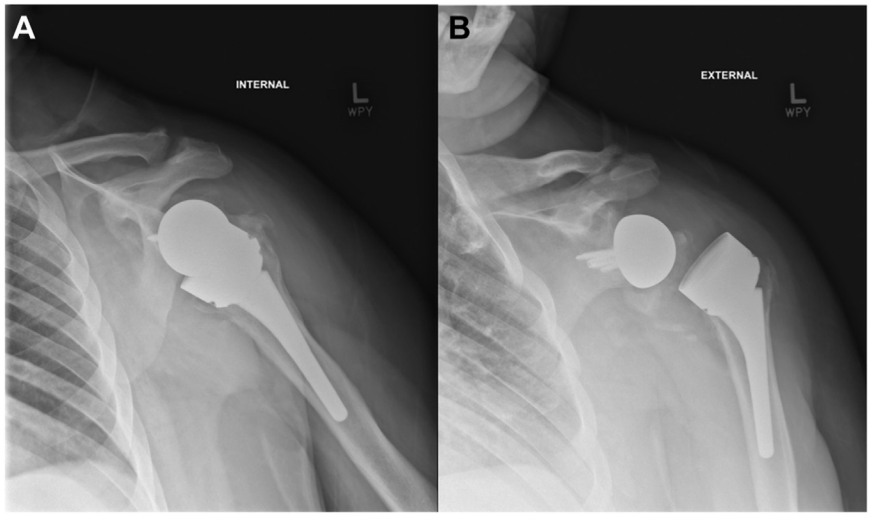

Figure 4 (**A**) Internal rotation and (**B**) external rotation AP views of the glenohumeral joint which show widening of the distance between the glenosphere and the humeral tray. *AP*, antero-posterior.

obtained 16 months after his hospitalization showed progressive autofusion of his left glenohumeral joint (Fig. 2). Additional workup included electromyography (EMG) and nerve conduction studies (NCS), which demonstrated intact axillary nerve function. A computed tomography scan was performed to further evaluate the heterotopic bone, which showed a cortical outline with cancellous bone in the center, consistent with mature HO[3] (Fig. 3).

Following his prolonged hospitalization, the patient also developed left foot drop with EMG and NCS demonstrating a complete left peroneal nerve palsy. The patient was hygienically unable to wash

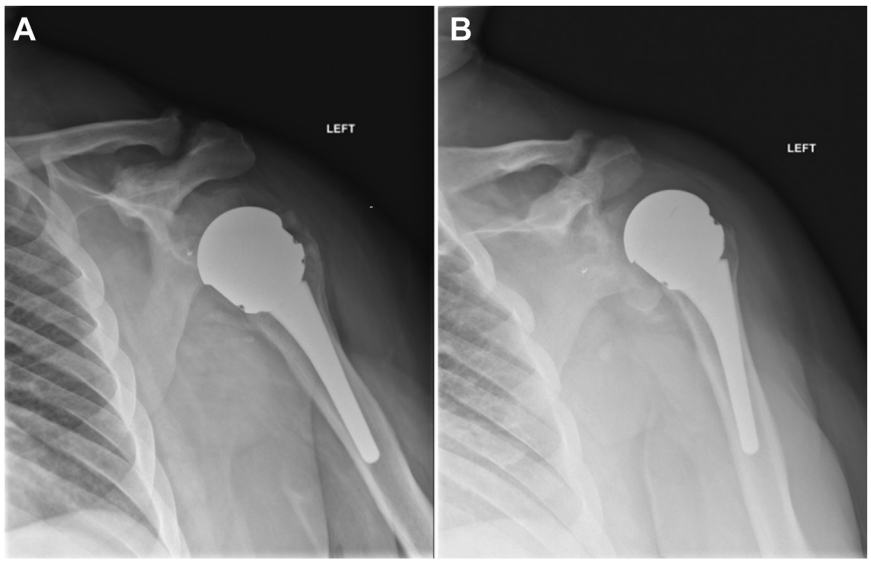

Figure 5 (**A**) Internal rotation and (**B**) AP views of the glenohumeral joint showing a stable hemiarthroplasty 6 months postoperatively. *AP*, antero-posterior.

under his arm or reach his face with his hand. A discussion of treatments ensued, with the patient expressing a desire to have at a minimum passive range of motion for hygiene and the ability to position his arm to allow him to reach his face. Although his preference was for active motion, he understood his risk of implant instability given his body habitus and increased fall risk with his foot drop.

The patient subsequently underwent HO excision and rTSA. At the time of surgery, significant ossification of the rotator cuff including the subscapularis, supraspinatus and infraspinatus tendons was noted. Intraoperatively, a constrained liner was placed to obtain stability, which was satisfactory during trialing of the components. After excision of HO, insufficient tendinous insertions of the rotator cuff remained to repair around the implant. Postoperatively, the shoulder was noted to be dislocated. Given the severity of soft tissue loss, the decision was made to treat this by implant revision. The patient returned to the operating room the next day, where the component sizes were increased, and a pectoralis major tendon transfer to the remnant lesser tuberosity was performed to place an internal rotation and compressive moment to the implant.

Over the next 4 months, the patient regained upper extremity range of motion with therapy and was able to use his arm for activities of daily living such as washing his contralateral side. Approximately 6 months after his revision surgery, the patient started experiencing instability in the shoulder. New imaging showed gapping between the glenosphere and humeral tray (Fig. 4). The decision was made to revise the rTSA. Intraoperatively, the shoulder was noted to be unstable with minimal manipulation. Heterotopic ossification (HO) was also noted on the posteroinferior aspect of the glenoid, but it was not resected due to its proximity to neurovascular structures. The patient already had the largest available components for the rTSA system in place; therefore, he was converted to a HA. A metal augment was placed onto the humeral stem and the glenoid was packed with bone graft. Intraoperative range of motion showed a stable prosthesis, except with extremes of extension. At the 6-month follow-up, imaging showed stable components and the patient had painless active forward elevation of 30° and abduction to 30° (Fig. 5).

Discussion

Heterotopic ossification (HO) develops in several stages. In the early phase, symptoms are characterized by localized pain and swelling, and radiographs may be negative. In this patient's case, the diagnosis of left internal jugular vein thrombosis was thought to explain his left shoulder pain and symptoms, and therefore additional workup was not pursued at that time. In the later stages, as HO matures, the swelling becomes more localized, and as the joint becomes affected limitations in range of motion become evident. This explains the delayed presentation to clinic as the patient experienced increasing stiffness, and the inability to use his left upper extremity for activities of daily living. Various treatment options were discussed with the patient including repositioning of the glenohumeral fusion into a more functional position, resection arthroplasty, HA, and total shoulder arthroplasty. Once electrodiagnostic studies confirmed intact axillary nerve function, it was decided that total shoulder arthroplasty would provide the greatest range of motion out of these options.

Given the extensive involvement of the patient's rotator cuff in the HO, restoring stability during surgery was challenging. During the initial revision, component sizes were increased, and a pectoralis major transfer was performed given a deficient subscapularis tendon. Six months later, during the second revision, the humeral component was noted to be dislocated anteriorly and the glenosphere anteverted. The humeral stem was well fixed and therefore the prosthesis was converted to a HA. Conversion from rTSA to HA has been described in cases where there is insufficient glenoid bone stock or stability from soft tissues. While patients report improved pain scores, range of motion is typically unchanged.[10,19] In this patient's case, the HA allows him limited but painless range of motion and increased stability of his glenohumeral joint.

Treatment for glenohumeral HO initially consists of conservative management in the form of anti-inflammatory medications and physical therapy. Once a patient is symptomatic and does not respond to conservative treatment, they require excision of the bony growth.[8] In a case series of patients who developed glenohumeral HO after a traumatic brain injury, 90% of patients improved their functional status after surgical HO resection.[6] Polfer *et al* also reported a successful result with HO excision in a patient with glenohumeral HO after a traumatic event.[17] In severe cases, fusion procedures can be performed to provide pain relief and a functional limb.[2] This highlights that while surgical solutions may offer some improvement in range of motion, restoring the premorbid state of the glenohumeral joint is unlikely.

Given the complications associated with HO excision, prophylaxis can be administered to patients who are deemed high risk. This can be in the form of nonsteroidal anti-inflammatory medications or radiation. The specific choice of nonsteroidal anti-inflammatory medication varies in the orthopedic literature.[23] While radiation offers the advantage of a one-time treatment, risks associated include wound complications and it is not feasible in patients with systemic insults.

Several factors could have contributed to tissue injury in this patient and subsequent HO formation. Extended ventilator times can lead to inflammatory states and are associated with musculoskeletal challenges such as sarcopenia and osteoporosis.[9,15] In addition, this patient underwent several episodes of prone positioning, which has been shown to improve oxygenation and survival in severe acute respiratory distress syndrome.[7,18] Risks of prone positioning include peripheral nerve injuries or brachial plexopathies[13] due to traction on the upper extremity or inadequate padding or support of extremities, which was also considered in this case and evaluated with EMG/NCS. In fact, this patient did experience an ipsilateral common peroneal nerve palsy. Certainly, his height and weight contributed to the challenges of safe ventilation and safety with prone positioning which were superseded by life-saving measures.

Conclusion

Similar to this patient, reports of HO formation after COVID-19 infection are limited to case series in which patients required prolonged mechanical ventilation and prone positioning.[11,20] While further studies are needed to define the pathophysiology of HO and the long-term outcomes in patients with associated COVID-19 infection, this case highlights the importance of considering HO formation in the differential diagnosis of a patient presenting with severe musculoskeletal pain and associated stiffness after a prolonged illness, as well as the challenges associated with re-establishing a functional joint once soft tissue stabilizers of the glenohumeral joint are no longer competent.

Disclaimers:

Funding: No funding was disclosed by the authors.
Conflicts of interest: The authors, their immediate family, or any research foundation with which they are affiliated did not receive any financial payments or other benefits from any commercial entity related to the subject of this article.
Patient consent: Obtained.

References

1. Almangour W, Schnitzler A, Salga M, Debaud C, Denormandie P, Genêt F. Recurrence of heterotopic ossification after removal in patients with traumatic brain injury: A systematic review. Ann Phys Rehabil Med 2016;59:263-9. https://doi.org/10.1016/j.rehab.2016.03.009.
2. Boynton MD, Enders TJ. Severe heterotopic ossification after arthroscopic acromioplasty: A case report. J Shoulder Elbow Surg 1999;8:495-7.
3. Bressler EL, Marn CS, Gore RM, Hendrix RW. Evaluation of ectopic bone by CT. Am J Roentgenol 1987;148:931-6.
4. Chae J, Siljander M, Michael Wiater J. Instability in reverse total shoulder arthroplasty. J Am Acad Orthop Surg 2018;26:587-96. https://doi.org/10.5435/JAAOS-D-16-00408.
5. Cipriano CA, Pill SG, Keenan MA. Heterotopic ossification following traumatic brain injury and spinal cord injury. J Am Acad Orthop Surg 2009;17:689-97. https://doi.org/10.5435/00124635-200911000-00003.
6. Fuller DA, Mani US, Keenan MAE. Heterotopic ossification of the shoulder in patients with traumatic brain injury. J Shoulder Elbow Surg 2013;22:52-6. https://doi.org/10.1016/j.jse.2012.01.028.
7. Guérin C, Reignier J, Richard J-C, Beuret P, Gacouin A, Boulain T, et al. Prone positioning in severe acute respiratory distress syndrome. N Engl J Med 2013;23:2159-68. https://doi.org/10.1056/NEJMoa1214103.
8. Hallock J, Field LD. Surgical management of shoulder heterotopic ossification. Arthrosc Tech 2019;8:e1057-62. https://doi.org/10.1016/j.eats.2019.05.019.
9. Kizilarslanoglu MC, Kuyumcu ME, Yesil Y, Halil M. Sarcopenia in critically ill patients. J Anesth 2016;30:884-90. https://doi.org/10.1007/s00540-016-2211-4.
10. Kriechling P, Andronic O, Wieser K. Hemiarthroplasty as a salvage treatment for failed reverse total shoulder arthroplasty. JSES Int 2021;5:1055-61. https://doi.org/10.1016/j.jseint.2021.07.003.
11. Meyer C, Haustrate MA, Nisolle JF, Deltombe T. Heterotopic ossification in COVID-19: A series of 4 cases. Ann Phys Rehabil Med 2020;63:565-7. https://doi.org/10.1016/j.rehab.2020.09.010.
12. Meyers C, Lisiecki J, Miller S, Levin A, Fayad L, Ding C, et al. Heterotopic ossification: A comprehensive review. JBMR Plus 2019;3:e10172. https://doi.org/10.1002/jbm4.10172.
13. Miller C, O'Sullivan J, Jeffrey J, Power D. Brachial plexus neuropathies during the COVID-19 pandemic: a retrospective case series of 15 patients in critical care. Phys Ther 2021;101:1-8. https://doi.org/10.1093/ptj/pzaa191.
14. Olsen E, Endrizzi D, Stephenson M, Farraher S. Characteristic heterotopic ossification of the proximal humerus after total shoulder arthroplasty. Skeletal Radiol 2021;50:973-9. https://doi.org/10.1007/s00256-020-03653-z/.
15. Orford NR, Pasco JA, Kotowicz MA. Osteoporosis and the critically ill patient. Crit Care Clin 2019;35:301-13. https://doi.org/10.1016/j.ccc.2018.11.006.
16. Patel P, Brkljac M, Sonar U, Kumar S. Heterotopic ossification following anterior shoulder dislocation. BMJ Case Rep 2018;2018:bcr2018226968. https://doi.org/10.1136/bcr-2018-226968.
17. Polfer EM, Nappo KE, Giuliani JR, Nesti LJ. Global excision of severe heterotopic ossification of the shoulder: A case report. JBJS Case Connect 2020;10:e0080. https://doi.org/10.2106/JBJS.CC.18.00080.
18. Shelhamer MC, Wesson PD, Solari IL, Jensen DL, Steele WA, Dimitrov VG, et al. Prone positioning in moderate to severe acute respiratory distress syndrome due to COVID-19: A cohort study and analysis of physiology. J Intensive Care Med 2021;36:241-52. https://doi.org/10.1177/0885066620980399.
19. Song IS, Jung D, Jeong U, An CH. Conversion of failed reverse total shoulder arthroplasty to hemiarthroplasty: Three cases of instability and three cases of glenoid loosening. Clin Orthop Surg 2019;11:436-44. https://doi.org/10.4055/cios.2019.11.4.436.
20. Stoira E, Elzi L, Puligheddu C, Garibaldi R, Voinea C, Chiesa AF, et al. High prevalence of heterotopic ossification in critically ill patients with severe COVID-19. Clin Microbiol Infect 2021;27:1049-50. https://doi.org/10.1016/j.cmi.2020.12.037.
21. Sullivan MP, Torres SJ, Mehta S, Ahn J. Heterotopic ossification after central nervous system trauma: A current review. Bone Joint Res 2013;2:51-7. https://doi.org/10.1302/2046-3758.23.2000152.
22. Teasell RW, Mehta S, Aubut JL, Ashe MC, Sequeira K, MacAluso S, et al. A systematic review of the therapeutic interventions for heterotopic ossification after spinal cord injury. Spinal Cord 2010;48:512-21. https://doi.org/10.1038/sc.2009.175.
23. Winkler S, Wagner F, Weber M, Matussek J, Craiovan B, Heers G, et al. Current therapeutic strategies of heterotopic ossification - a survey amongst orthopaedic and trauma departments in Germany. BMC Musculoskelet Disord 2015;16:313. https://doi.org/10.1186/s12891-015-0764-2.