

C A S E R E P O R T

Cementless reverse total shoulder arthroplasty in a patient affected by Osteogenesis Imperfecta: a case report and review of the literature

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Abstract. *Background and aim of the work* Osteogenesis Imperfecta is a rare genetic condition. The use of total shoulder arthroplasty in these patients is very uncommon, with only two cases reported in the literature. This study describes a cementless reverse total shoulder arthroplasty (rTSA) for a multi-fragmented fracture in a patient affected by Osteogenesis Imperfecta (OI) type 1 and aims to review literature results of shoulder replacement in patients affected by this uncommon condition. *Methods:* The case of a woman affected by OI type 1 treated with a cementless rTSA for a multi-fragmented proximal humerus fracture is reported. Focusing on the fixation technique, a literature review regarding the orthopaedic options in patients affected by Osteogenesis Imperfecta was performed and compared to the techniques used in the unaffected population. *Result:* Our patient shows good results in terms of clinical and radiological outcomes at the short term follow up. Few studies treat the orthopaedic manifestation of this rare genetic condition and only two are focused on shoulder arthroplasty. Cement is the preferred method for fixation in both papers. *Conclusion:* Cementless reverse shoulder arthroplasty may be an option in patients affected by OI type 1, although literature seems to support cement as the fixation method of choice. (www.actabiomedica.it)

Key words: Cementless, reverse, total, shoulder, arthroplasty, osteogenesis, imperfecta

Introduction

Reverse total shoulder arthroplasty (rTSA) has been used in Europe for almost 20 years. Originally indicated for patients with rotator cuff arthropathy, in the last few years rTSA has been used to treat various other pathologies including displaced proximal humeral fractures in elderly patients. We present the case of a cementless rTSA in a patient affected by Osteogenesis Imperfecta (OI).

Case report

A 70-years old right hand-dominant woman affected by Osteogenesis Imperfecta type 1 came to our emergency department complaining of severe left shoulder pain, swelling and functional limitation after a domestic fall. Before the trauma, the patient was used to live alone and was able to perform activities of daily living, with some limitation due to her genetic pathology. As a teenager, she was diagnosed with OI type I due to blue sclerae, several fractures (more than 30 reported events) that never required surgery. The

patient has many other comorbidities including blindness secondary to glaucoma, juvenile cataract, retinal detachment, right-convex dorsal scoliosis and osteoporosis treated with annual infusion of zoledronic acid. Clinical examination failed to show any neurovascular deficits or other injuries. Plain radiographs of the shoulder detected a 4-fragments proximal humerus fracture (Fig.1). A CT scan was performed and confirmed the displaced multi-fragmented fracture of the proximal humerus, with subluxation of the humeral head (Fig.2).

A reverse shoulder arthroplasty was preferred over a synthesis because of the fragmentation and the poor quality of the bone stock. Appropriate informed consent was obtained. The patient was placed in beach-chair position and a delto-pectoral approach was performed. The main cephalic fragment was identified and removed without the need of further humeral head osteotomy. A small size metallic glenoid component fixed with two screws and a 40 mm glenosphere were implanted. A preventive bone cerclage on the proximal humeral diaphysis was performed to avoid further rim fracture propagation. A medium size cementless SMR reverse shoulder arthroplasty prosthesis (Lima Corporate, San Daniele del Friuli, Italy) was implanted as

our experience leads to prefer (1). Postoperative plain radiographs were obtained (fig 3).

Postoperative course was uneventful with the patient discharged at home three days after the operation without complications. The involved arm was hold in

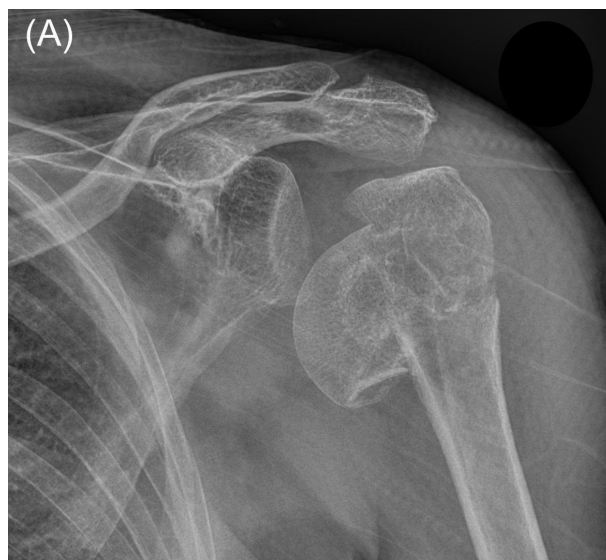


Figure 1. Plain radiographs (antero-posterior view) obtained the day of trauma showing the 4-fragments proximal humerus fracture

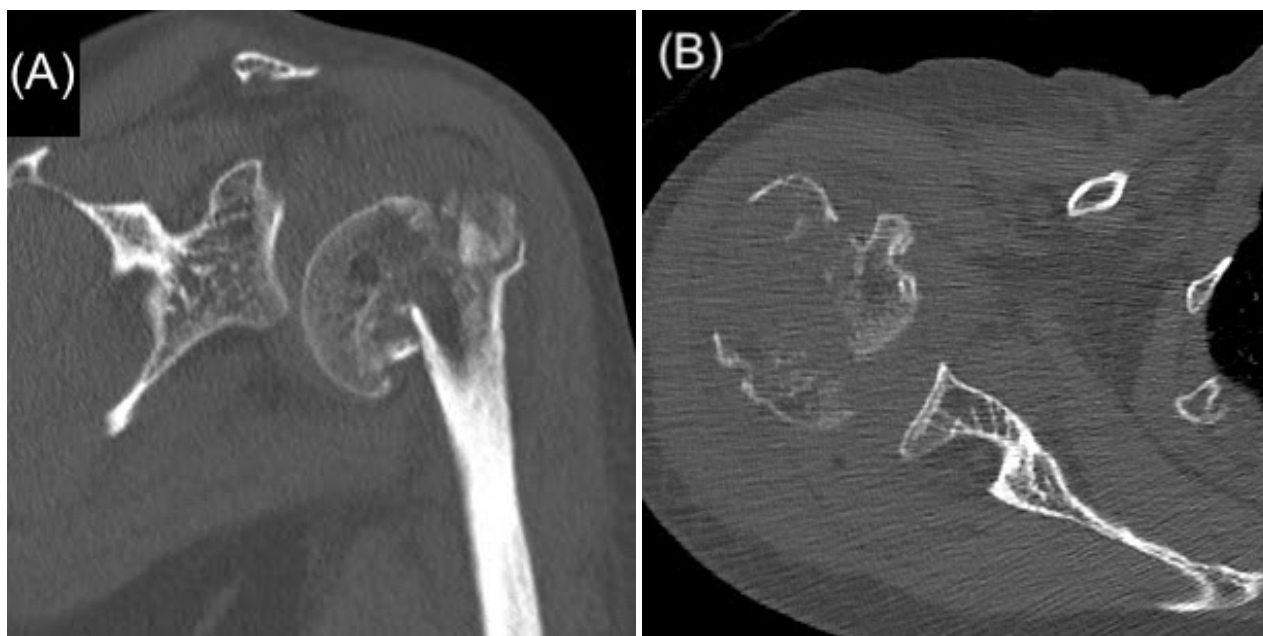


Figure 2. CT scan (coronal and axial view) obtained the day of trauma (A), (B) Left shoulder, multi-fragmented fracture of the proximal humerus, with subluxation of the humeral head

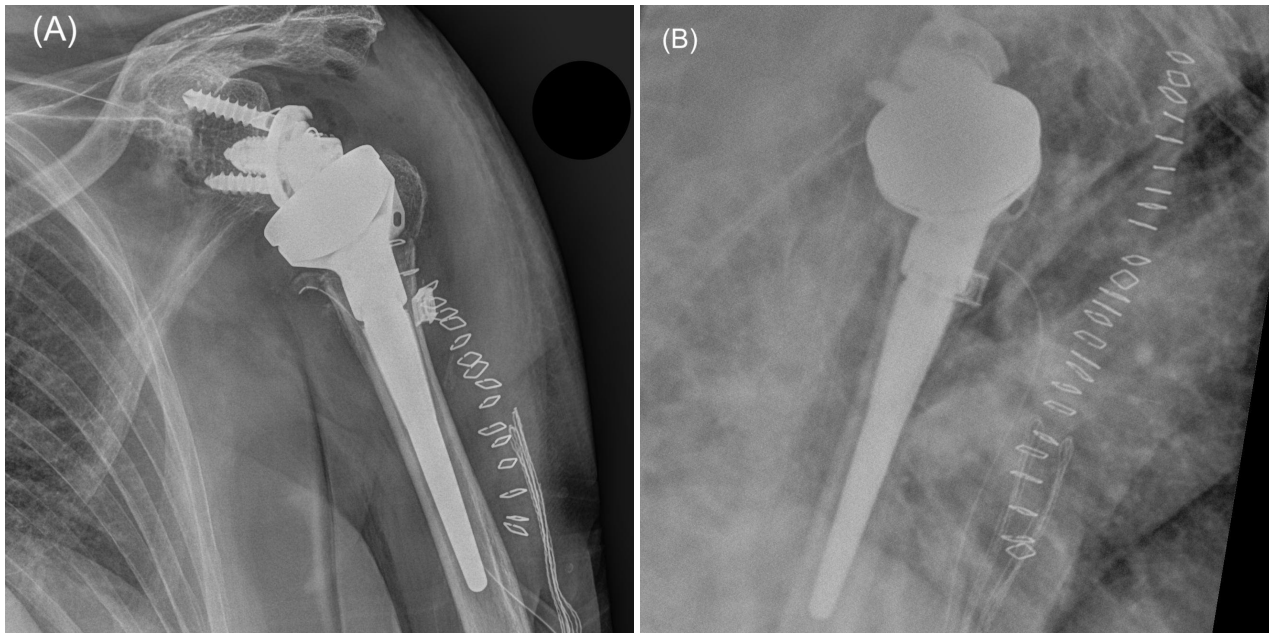


Figure 3. Postoperative plain radiographs. (A), (B) Antero-posterior and axial view of the left shoulder

brace during rest for three weeks with early passive and assisted active mobilization allowed.

After one month, the patient was able to achieve 40° active abduction and flexion, 10° active extra-rotation and internal rotation at the trochanteric region with pain. At 6-months follow-up, the patient im-

proved the range of motion with decreased pain intensity. She was able to perform 75° active abduction and 85° active flexion, 30° active extra-rotation and intra-rotation to lumbar spine. Usual radiographic controls showed good positioning of the implant components without signs of osteolysis or mobilization (Fig. 4).

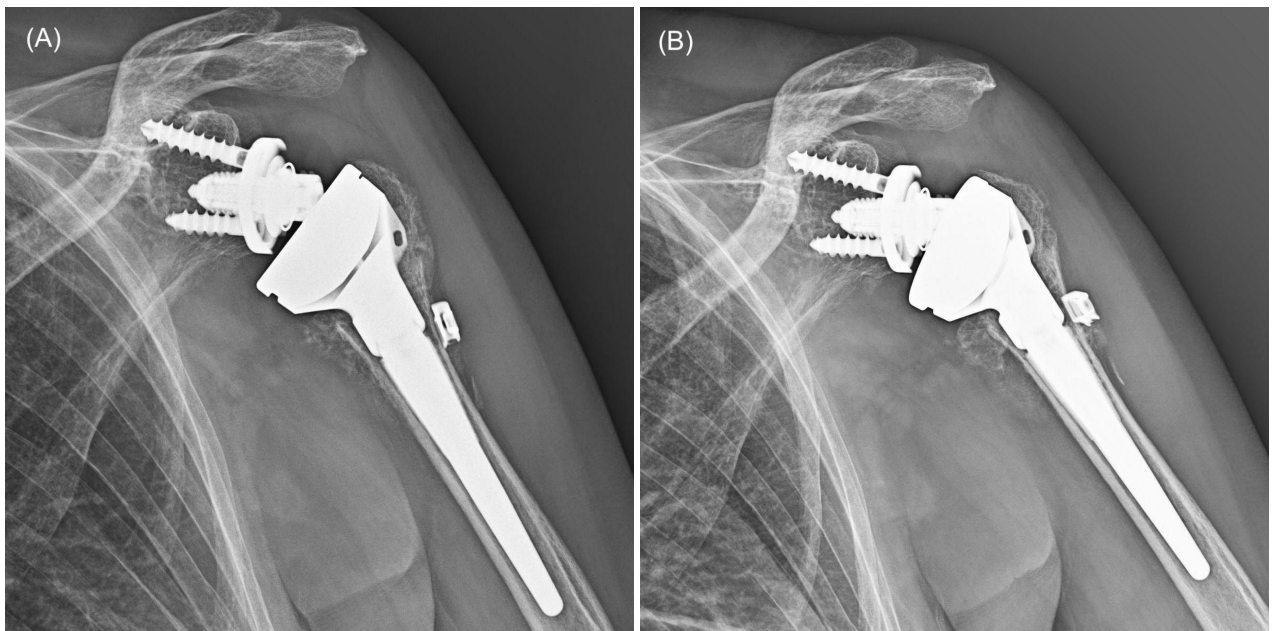


Figure 4. Six months follow up radiographs. (A), (B) Antero-posterior view of the left shoulder

Discussion

Osteogenesis Imperfecta, also known as “brittle bone disease”, is a heritable group of collagen-related disorders that may cause many orthopaedic manifestations. It involves a heterogeneous group of genetic conditions characterized by increased bone fragility, low bone mass, and susceptibility to bone fractures which are prone to nonunion and malunion. (2) The classification of Silence corresponds better with the genetics basis of OI. (3) This disease is known mainly for its skeletal manifestations in pediatric age, as it is frequently reported in literature. Actually, few studies deal with the consequences on adult individuals, even if more than 25% of lifetime fractures are reported to occur during adulthood (4,5). The literature is very limited and focuses mainly on joint replacement of the lower limbs.

Papagelopoulos and Morrey retrospectively analysed the Mayo Clinic registry from 1969 to 1990 by reviewing the outcomes of cemented total hip arthroplasty (THA) and total knee arthroplasty (TKA) in patients with OI. At the end of the follow-up, 7 and 10 years, they found no complications or radiographic signs of osteolysis or mobilization. (3) In a case series, Krishnan et al retrospectively reviewed 4 patients and 6 uncemented THA. The authors described a higher complication and revision rate, with four intraoperative femoral fractures, one acetabulum fracture and four postoperative acetabular protrusions. (6) In line with these experiences, Roberts et al recently recommend the use of cemented implants to respect the increased bone fragility of those patients. (5) Therefore, limited cohorts recommend caution during the femoral canal preparation and stem implant to prevent iatrogenic fractures that are very common and can affect final outcomes. (6,7) According to these indications, in our case, a preventive bone cerclage to avoid any iatrogenic damage was performed.

Lower limbs have higher functional demands because they involve joints which work under weight bearing. This physiological situation combined with bone fragility due to OI, leads the surgeons to prefer the use of cemented implants.

Moreover, few publications deal with shoulder replacement in patients suffering from OI. Mnif et al reported the case of bilateral posterior fracture-

dislocation of the shoulders in a man with OI treated with a bilateral cemented total shoulder arthroplasty with successful outcomes after 3 years of follow-up. (8) McLaughlin et al described the case of a patient with OI type I complicated by a proximal humeral enchondroma and a rotator cuff tear treated with a cemented reverse shoulder arthroplasty with good clinical and radiographic outcomes at 5 months of follow-up. (9)

Therefore, this suggests that upper extremity arthroplasty may be an effective treatment option for post-traumatic injuries in patients with OI and rTSA could be an effective technique using a cemented fixation, if a rotator cuff tear is present. However, there is no evidence against the choice of a cementless fixation technique for rTSA.

The challenge was to use a cementless technique also in a patient affected by OI using the rational concept that the upper limb has fewer functional demands, being not heavily stressed as the lower limbs.

Several studies discuss the difference between cemented or cementless rTSA in patients with healthy bone. Wright et al suggested that cementless rTSA offers good clinical outcomes and stable fixation with a low rate of complications in treatment of 3- and 4-parts proximal humeral fractures advising that cement may be not necessary. (10) Youn et al, in a radiological focused study, reported that the cementless nature of the humeral component did not result in early loosening or failure of the implant (11) In a retrospective cohort study, Wiater et al demonstrated that uncemented stems have at least equivalent clinical and radiographic outcomes compared with cemented stems but a better complication profile. (12) Phadnis et al, reviewing 41 clinical studies, demonstrated that the cementless stems have significantly lower incidence of postoperative acromion fractures but a higher incidence of non-progressive radiolucent lines at two years. (13) Concerning this, Castagna et al reported that these lines, commonly seen in uncemented stems, do not appear to compromise the fixation or the clinical outcomes in a mid-term follow-up. (14)

According to these evidences, cementless rTSA is becoming the preferred therapeutic approach at our institution for comminuted proximal humeral fractures and cuff tear arthropathy in otherwise healthy patients. (15) This because the cementless technique

reduces the operating time, avoids the morbidity associated with the use of cement and allows the realignment or exchange of the humeral component during surgery. Moreover, in our experience the use of cement could significantly complicate a future revision surgery that may be frequent in these patients. (6)

Conclusion

Shoulder replacement and fixation could be challenging and technically demanding for the orthopaedic surgeon, especially in those patients with extreme bone fragility. Literature agrees on the indication to cement lower limb joints replacements while little has been written about shoulders. Our experience, considering the good outcome though after a short follow-up, highlights that the cementless rTSA may be an option also in patients with a poor bone quality as it usually happens in OI. Longer follow up is obviously required to draw further conclusions while controlled trials may be difficult to conduct due to the low prevalence of OI.

Conflict of interest: Each author declares that he or she has no commercial associations (e.g., consultancies, stock ownership, equity interest, patent/licensing arrangement etc.) that might pose a conflict of interest in connection with the submitted article.

References

1. Beltrame A, Di Benedetto P, Cicuto C, Cainero V, Chisoni R, Causero A. Onlay versus Inlay humeral stem in Reverse Shoulder Arthroplasty (RSA): clinical and biomechanical study. *Acta Bio-Medica Atenei Parm.* 2019;90(12-S):54-63. doi:10.23750/abm.v90i12-S.8983
2. Marini JC, Forlino A, Bächinger HP, et al. Osteogenesis imperfecta. *Nat Rev Dis Primer.* 2017;3:17052. doi:10.1038/nrdp.2017.52
3. Papagelopoulos PJ, Morrey BF. Hip and knee replacement in osteogenesis imperfecta. *J Bone Joint Surg Am.* 1993;75(4):572-580. doi:10.2106/00004623-199304000-00011
4. Gil JA, DeFroda SF, Sindhu K, Cruz AI, Daniels AH. Challenges of Fracture Management for Adults With Osteogenesis Imperfecta. *Orthopedics.* 2017;40(1):e17-e22. doi:10.3928/01477447-20161006-04
5. Roberts TT, Cepela DJ, Uhl RL, Lozman J. Orthopaedic Considerations for the Adult With Osteogenesis Imperfecta. *J Am Acad Orthop Surg.* 2016;24(5):298-308. doi:10.5435/JAAOS-D-15-00275
6. Krishnan H, Patel NK, Skinner JA, et al. Primary and revision total hip arthroplasty in osteogenesis imperfecta. *Hip Int J Clin Exp Res Hip Pathol Ther.* 2013;23(3):303-309. doi:10.5301/hipint.5000014
7. Wagner R, Luedke C. Total knee arthroplasty with concurrent femoral and tibial osteotomies in osteogenesis imperfecta. *Am J Orthop Belle Mead NJ.* 2014;43(1):37-42.
8. Mnif H, Koubaa M, Zrig M, et al. Bilateral posterior fracture dislocation of the shoulder. *Chir Main.* 2010;29(2):132-134. doi:10.1016/j.main.2009.11.001
9. McLaughlin RJ, Watts CD, Rock MG, Sperling JW. Reverse total shoulder arthroplasty in a patient with osteogenesis imperfecta type I complicated by a proximal humeral enchondroma: a case report and review of the literature. *JSES Open Access.* 2017;1(2):119-123. doi:10.1016/j.jses.2017.03.012
10. Wright JO, Ho A, Kalma J, et al. Uncemented Reverse Total Shoulder Arthroplasty as Initial Treatment for Comminuted Proximal Humerus Fractures. *J Orthop Trauma.* 2019;33(7):e263-e269. doi:10.1097/BOT.0000000000001465
11. Youn S-M, Deo S, Poon PC. Functional and radiologic outcomes of uncemented reverse shoulder arthroplasty in proximal humeral fractures: cementing the humeral component is not necessary. *J Shoulder Elbow Surg.* 2016;25(4):e83-e89. doi:10.1016/j.jse.2015.09.007
12. Wiater JM, Moravek JE, Budge MD, Koueiter DM, Marcantonio D, Wiater BP. Clinical and radiographic results of cementless reverse total shoulder arthroplasty: a comparative study with 2 to 5 years of follow-up. *J Shoulder Elbow Surg.* 2014;23(8):1208-1214. doi:10.1016/j.jse.2013.11.032
13. Phadnis J, Huang T, Watts A, Krishnan J, Bain GI. Cemented or cementless humeral fixation in reverse total shoulder arthroplasty? a systematic review. *Bone Jt J.* 2016;98-B(1):65-74. doi:10.1302/0301-620X.98B1.36336
14. Castagna A, Delle Rose G, De Giorgi S, Gumina S, Garofalo R, Borroni M. Do radiolucent lines and stress shielding of the humeral shaft really matter in shoulder arthroplasty? *J Biol Regul Homeost Agents.* 2020;34(4 Suppl. 3):309-314. Congress of the Italian Orthopaedic Research Society.
15. Beltrame A, Di Benedetto P, Salviato D, et al. The SMR reverse shoulder arthroplasty in rotator cuff arthropathy management. *Acta Bio-Medica Atenei Parm.* 2017;88(4S):81-89. doi:10.23750/abm.v88i4 -S.6798:

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