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Operatively Treated Pelvic Ring Fractures in Adult Patient with Osteogenesis Imperfecta after Traumatic Injury: A Case Report

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Abstract

Osteogenesis imperfecta (OI) is a rare heritable disorder of type I collagen that predisposes patients to recurrent fractures and skeletal deformities. Although fracture management in children with OI has been widely described, guidance for adult patients is comparatively limited. This is a report of the case of a 35-year-old man with OI who sustained right-sided sacroiliac diastasis and pubic rami fractures following a high-energy bicycle accident. The injury was treated with percutaneous screw fixation, achieving satisfactory reduction and stable fixation without perioperative complications. The patient advanced to full weight bearing at 12 weeks, and at 2-year follow-up demonstrated near-baseline function with Visual Analog Scale and Majeed Pelvic Scores that closely approximated those reported in non-OI populations. Radiographs showed no evidence of malunion or nonunion, though he continued to experience chronic low back pain. This case suggests that standard fixation strategies may be effective for selected adults with mild OI phenotypes, provided that preoperative planning accounts for the condition's distinctive biomechanical and hemostatic challenges.

Acronyms

OI = Osteogenesis imperfecta
VAS = Visual Analog Scale

Introduction

Osteogenesis imperfecta (OI) is a group of genetic disorders involving mutations of type 1 collagen, a substantial component of bone and connective tissues. Deficiencies in type 1 collagen have been associated with fragility fractures, skeletal deformities, and growth disturbances.[1,2] The genetics and phenotypes of OI have been extensively studied, though literature concerning fracture management in adult patients with OI is comparatively sparse.

Much of the current musculoskeletal literature focuses on pediatric presentations and management, even though up to 25% of lifetime fractures will occur when patients are adults.[3] Despite pharmacologic management, many patients will continue to experience sequalae of their conditions, even with advancements in medications and rehabilitative modalities.[4] Some unique challenges implicated in fracture management of adult patients with OI are altered osseous biomechanics, skeletal deformities, sequelae of chronic bisphosphonate use, low bone mineral density, and predisposition for excessive bleeding.[5,6]

While fractures of the pelvis and acetabulum are typically associated with higher energy mechanisms, those with OI may sustain these fractures after low energy mechanisms. Treatment of adult OI pelvic ring injuries include added challenges, as these patients may have a propensity for excessive bleeding, poor bone quality, and preexisting acetabular or femoral head deformities.[7,8] In a case series by Darmanis and Bircher, the authors reported difficulties with standard implant fixation for acetabular fractures in individuals with OI due to acetabular deformities secondary to disease pathology. Their series required surgical modifications to account for acetabular protrusion, thin quadrilateral plate, and poor screw purchase, and highlighted the need for thorough pre-operative planning.[5]

Though the genetics and medical complexities of OI have been well established, the dearth of literature in fracture management, and in particular pelvic ring fractures was the impetus to report this case. As such, the authors present a case report of an adult patient with OI who sustained pelvic ring fractures after a traumatic mechanism requiring operative management, aiming to contribute to the current literature on optimal practice management for this unique patient population.

Case Study

Institutional Review Board committees at the treating hospitals reviewed this case report and appropriate approval was given for commencement of study. The Visual Analog Scale (VAS) and Majeed Pelvic Score questionnaires were utilized. Appropriate patient education regarding each questionnaire was provided. The questionnaires covered the period 1 month prior to injury and then post-operatively at defined intervals: 2 weeks, 6 weeks, 3 months, 6 months, 1 year, 2 years. Of note, the questionnaires were first given at the 6 week post-operative visit, thus the patient had to recall the 1 month pre-injury and 2-week post-op scores from memory.

Patient responses were collected and analyzed on Microsoft Excel 2020 (Microsoft Corporation, Redmond, WA).

The patient is a 35-year-old male who presented after a bicycle accident. The patient felt immediate pain in his right hip and right hemipelvis and was unable to ambulate, requiring paramedic transport to the local community hospital. Past medical history was pertinent for chronic low

back pain and childhood fractures of his wrist and fingers attributed to a known diagnosis of OI. The patient worked as a farmer and had no functional limitations. The patient could not recall which subtype of OI he had been diagnosed with. After medical stabilization at the community hospital, he was transferred to the Level 1 trauma center in Hawai'i. The patient underwent standard advanced trauma life support and was noted to be hemodynamically stable. Initial imaging revealed pelvic ring fractures, notably diastasis of the right sacroiliac joint and right superior and inferior pubic rami fractures. No additional injuries were identified and the patient was stable. Due to insurance considerations, the patient was transferred to another facility for definitive management.

After evaluation by the accepting orthopaedic team, the patient was diagnosed with right-sided sacroiliac diastasis and right-sided displaced superior and inferior pubic rami fractures. After discussion with the patient concerning treatment options, benefits, risks, and alternatives the patient elected to proceed with surgical management of his pelvic ring injury.

Reduction of the right superior ramus was achieved using a femoral distractor with a Schanz pin placed into the left supra-acetabular corridor and another into the greater trochanter towards the femoral neck. Fixation of the right superior pubic ramus fracture was achieved using the retrograde anterior column screw technique with a 6.5mm x 130mm fully threaded screw. The femoral distractor was removed, and attention was turned to the right sacroiliac diastasis. Reduction was achieved at the sacroiliac joint, with subsequent advancement of a 7.3mm x 110mm iliosacral screw. Appropriate reduction and safe screw placement were confirmed with intra-operative fluoroscopy.

Post-operatively

The patient was discharged on post-operative day 3 and planned for touch down weight bearing on right lower extremity for 3 months post-operatively. The patient was seen at routine follow up with no signs of wound dehiscence throughout the follow-up period. The patient did report right hip pain at his initial 2-week follow-up, with resolution of pain and full active and passive range of motion by his 6-week follow-up. The patient did note acute worsening of his chronic low back pain at his 6-week visit, however the patient attributes this pain to the nature of his work and is well documented prior. The patient was liberated to full weight bearing at approximately 12 weeks post-operatively. At his last follow-up, approximately 2 years post-operatively, the patient reported negligible and occasional right hip pain, as well as his chronic low back pain, though had no functional limitations and had resumed all normal activity.

VAS and Pelvic Majeed Scores

One month prior to injury, the patient felt little to no pain in any portion of his pelvis. On the day of the accident, the patient reported a VAS score of 8 and 9 to his left anterior and posterior hemipelvis respectively, and a score of 10 to both his anterior and posterior right hemipelvis. VAS scores then trended downward at 2 weeks, 6 weeks, 3 months, and 6 months post-injury. At 6 months post-injury, reported VAS began to approximate pre-injury levels, particularly in the anterior and posterior portions of the left pelvis, however scores for the right pelvis remained mild to moderate, reported as a 4 and 3 respectively. By 1 year and 2 years post-injury, the patient's VAS scores were equivalent to that of pre-injury in the left hemipelvis. At 1 year and 2 years post-injury, the right hemipelvis VAS scores remained between 1-3, with the patient reporting scores increasing to 5 while doing physical activities. Summary of VAS scores can be seen in **Table 1**.

Majeed Pelvic Scores were also obtained from patient. The patient reported scores of ⩾ 85 for all portions of the pelvis 1 month prior to injury. Two weeks post-operatively all subscales were poor clinically, with most receiving scores of 0. Thereafter, his scores improved but remained <55 by 3 months post-operatively. At 6 months post-operatively, scores approached pre-injury levels. At 1 year and 2 years post-operatively, scores were comparable to pre-injury scores, namely for the left anterior, left posterior, and right anterior pelvis. The right posterior hemipelvis scores did not show improvement after the 6-months post-operative mark, remaining at a score of 84 until final follow up. Summary of post-operative Majeed Pelvic Scores is presented in **Table 2**.

Discussion

At the study's start, the patient decided to identify his genetic variation of OI through commercial genetic testing (Skeletal Disorders Panel, Invitae, San Francisco, CA). His genetic report identified a *COL1A2* gene variation involving a substitution of an arginine with a serine at codon 948. To the authors' knowledge, there is no current literature on this specific variant.

At final follow-up, the patient reported persistent back pain, which was stated to have been present prior to injury but acutely worsened after injury. Low back pain in individuals with OI is common, with McKiernan et al citing 70% of their respondents whom had OI reporting low back pain causing some functional impairment.[3] At last follow-up, no identifiable pathology had been elucidated to explain the patient's low back pain, though the patient was referred to a spine specialist. This patient's reported persistence of back pain is likely a combination of pre-injury chronic pain and aggravation due to the injury and surgery.

Prior literature suggests that manifestations of OI may affect fracture healing. Some studies have suggested high rates of nonunion in those with OI for both fractures and osteotomies, 24% and 52% respectively.[9,10] A recent animal study found callus size and strength were reduced in OI fracture models, suggesting an increased risk of refracture.[11] Additionally, delayed fracture healing in patients with OI has shown to increase the risk of extended periods of immobilization, muscle atrophy, and hindered bone remodeling.[12] However, the current case demonstrates ap-

Table 1. Visual Analog Scores of a Patient with Right-sided Sacroiliac Diastasis and Pubic Rami Fractures from Pre-injury to 2 Years Post-injury

	1 month prior	Day of accident	2 weeks post injury	6 weeks post injury	3 months post injury	6 months post injury	1 year post injury	2 years post injury
Left anterior	0	8	7	5	4	0	0	0
Left posterior	1	9	8	6	4	2	1	1
Right anterior	0	10	8	8	6	4	2	0
Right posterior	1	10	8	7	5	3	3-normal activity; 5-with exertion	2-normal activity; 5-with exertion

propriate chronology of fracture healing. At the 12-week post-operative visit, radiographs demonstrated minimal residual fracture lines, with no clinical or radiographic evidence of delayed fracture healing, nonunion, or malunion at last follow up. His favorable outcomes may have been the result of his genetic variant and mild phenotype, though his young age, lack of co-morbidities, and appropriate rehabilitation and post-operative activity may also have played a role. Additional genetic work could help elucidate which OI variants are susceptible to worse fracture related outcomes.

There is concern for bleeding diathesis in individuals with OI.[7,8] Fortunately, this patient did not experience any such complications in the perioperative period. This may be due to the percutaneous surgical fixation techniques implemented. His genetic variant also may have not had severe effects on normal physiologic vascular and coagulation functions.

This patients pre-injury and post-operative reported measures are comparable to non-OI patients after pelvic ring fractures. In a study by Liu et al, general population patients who sustained posterior pelvic ring injuries treated by percutaneous sacroiliac screws reported pre-operative VAS scores of 7.13 ± 1.00, which was similar to this patient's pre-operative VAS scores.[13] The post-operative scores in the same study were 5.33 ± 0.78 and 1.33 ± 0.66 at 1 week and 6 months, respectively.[13] The 6 month VAS scores in this case were 2-4, which is slightly higher than the scores reported by Liu et al.[13] Close approximation of patient reported scores between the current patient and non-OI patients was also seen in the Majeed Pelvic Score. A retrospective review by Sharma et al, examined complications and functional outcomes of patients with complex fractures of the anterior pelvic ring treated with internal fixation.[14] The mean 6 months post-operative Majeed Pelvic Scores for non-OI patients was reported as 92.67 ± 5.8.[14] This patient, at the same post-operative interval, reported scores of 95, 86, 81, and 84 for the left anterior, left posterior, right anterior, and right posterior portions of the pelvis, respectively. These findings suggest the patient may have had subjective pain levels and functional abilities similar to that of non-OI patients. The comparable scores between the current patient and a non-OI patient group may suggest patients with mild phenotypes of OI may have comparable outcomes after pelvic ring fixation with those who do not have OI.

Several limitations apply to this study. First, the genetics of OI are heterogenous, which limits the generalizability to other OI patients. Additionally, this patient may have a milder phenotype given his occupational and functional abilities. Likewise, this patient's injuries were sustained because of a higher energy mechanism, which too may hinder generalizability to similar injuries sustained at lower energy mechanisms. This patient also had to provide some intervals of VAS and Majeed Pelvic Scores based on memory, which may introduce recall bias. And as mentioned, the use of a commercial genetic testing company for evaluation of genetic mutations may have incomplete or limited clinical utility.

While limitations in any case study must be recognized, there remains benefit in this study. There are likely many patients either with no or incomplete knowledge of their particular mutation or mutations. OI overall is not a common condition, and patients with OI sustaining pelvic ring injuries is even more rare. This case study contributes to knowledge of a patient population with a dearth of literature on pelvic ring fracture management. In this study, an adult with an apparent mild phenotype of OI presented with traumatic pelvic ring injuries after a high energy mechanism. He was treated with currently accepted, standard of care surgical techniques and fixation, and had short, intermediate, and long-term post-operative follow-ups without complications. His VAS and Majeed Pelvic Scores were similar to those of patients who did not have OI at most post-operative follow-up intervals. At final follow-up, and there was no radiographic evidence of malunion or nonunion, with the patient reporting continued low back pain, which may have been in part due to pre-existing chronic back pain. This patient had no complications at final follow-up and was back to performing recreational and professional activities at near pre-operative levels, suggesting standard pelvic fixation methods may be safe and effective in adult patients with mild OI phenotypes.

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Table 2. Majeed Pelvic Scores by Region of a Patient with Right-sided Sacroiliac Diastasis and Pubic Rami Fractures from Pre-injury to 2 Years Post-injury

Left Anterior							
	1 Month Prior	**2 Weeks Post**	**6 Weeks Post**	**3 Months Post**	**6 Months Post**	**1 Year Post**	**2 Years Post**
Pain	30	0	4	10	25	30	30
Work	20	0	2	6	20	20	20
Sitting	10	3	6	8	10	10	10
Sexual Intercourse	4	0	0	2	4	4	4
Standing (A)	12	6	6	6	12	12	12
Standing (B)	12	0	0	4	12	12	12
Standing (C)	12	2	4	6	12	12	12
Total	100	11	22	42	95	100	100
Left Posterior							
	1 Month Prior	**2 Weeks Post**	**6 Weeks Post**	**3 Months Post**	**Six Months Post**	**1 Year Post**	**2 Years Post**
Pain	25	0	3	7	20	25	25
Work	20	0	2	6	20	20	20
Sitting	10	1	5	6	9	10	10
Sexual Intercourse	4	0	0	2	4	4	4
Standing (A)	12	6	6	6	12	12	12
Standing (B)	12	0	0	2	12	12	12
Standing (C)	12	2	4	6	9	10	10
Total	95	9	20	35	86	93	93
Right Anterior							
	1 Month Prior	**2 Weeks Post**	**6 Weeks Post**	**3 Months Post**	**6 Months Post**	**1 Year Post**	**2 Years Post**
Pain	30	0	2	7	18	25	30
Work	20	0	2	4	20	20	20
Sitting	10	1	6	8	8	10	10
Sexual Intercourse	4	0	0	1	4	4	4
Standing (A)	12	6	6	6	12	12	12
Standing (B)	12	0	0	2	10	12	12
Standing (C)	12	2	4	6	9	12	12
Total	100	9	20	34	81	95	100
Right Posterior							
	1 Month Prior	**2 Weeks Post**	**6 Weeks Post**	**3 Months Post**	**6 Months Post**	**1 Year Post**	**2 Years Post**
Pain	25	0	2	6	20	20	20
Work	20	0	2	6	20	20	20
Sitting	10	1	5	6	9	8	8
Sexual Intercourse	4	0	0	2	4	4	4
Standing (A)	12	6	6	6	12	12	12
Standing (B)	12	0	0	2	10	10	10
Standing (C)	12	2	4	6	9	10	10
Total	95	9	19	34	84	84	84

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