



Case report

Total hip arthroplasty in an adult patient with pelvic dysmorphism, unilateral sacroiliac joint autofusion, and developmental hip dysplasia

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ARTICLE INFO

Article history:

Received 19 September 2019

Received in revised form

23 October 2019

Accepted 26 October 2019

Available online 30 November 2019

Keywords:

Hip dysplasia

Pelvic dysmorphism

Total hip arthroplasty

ABSTRACT

This case describes the challenges associated with total hip arthroplasty in a patient with unique anatomy, including developmental dysplasia of the hip, pelvic dysmorphism, and unilateral sacroiliac joint autofusion. A 30-year-old female, with a history of developmental dysplasia of the hip treated with presumed pelvic osteotomy complicated by postoperative infection, presented with hip pain refractory to conservative management. Radiographic studies demonstrated a 10-cm leg length discrepancy, 20° of acetabular retroversion, severe hemipelvic dysmorphism, ipsilateral sacroiliac joint autofusion, and significant femoral head dysplasia. Total hip arthroplasty was performed using a revision acetabular component and modular femoral component, resulting in improvement in the postoperative leg length discrepancy. There were no neurovascular or other perioperative complications, and the patient was ambulating without pain or assistive devices at 1-year follow-up.

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Introduction

Developmental dysplasia of the hip (DDH) encompasses a spectrum of altered hip anatomy that often consists of an underdeveloped acetabulum, uncovered or dislocated femoral head, and acetabular retroversion [1]. Furthermore, additional deformities include a dysplastic femur with increased anteversion, a shorter femoral neck, and an elliptic femoral head [1]. These structures and subsequent altered biomechanics are thought to contribute to accelerated degeneration of the dysplastic hip joint [1,2]. A subset of these patients ultimately requires total hip arthroplasty (THA) due to the long-term sequelae of untreated DDH.

It is also reported that there is an associated but poorly characterized relationship between sacroiliac (SI) arthritis and hip

arthritis [3]. However, there are few reports on THA in the setting of simultaneous DDH and pelvic ring anomalies. In this report, we describe the case of THA in a patient with unique anatomy, including pelvic dysmorphism, DDH with acetabular retroversion, and ipsilateral SI joint autofusion. To our knowledge, this is the first report of THA in a young adult with severe pelvic dysmorphism and concomitant untreated DDH.

Case history

History

A 30-year-old Spanish-speaking female with no significant past medical history presented to the orthopedic clinic with a complaint of several years of left hip pain exacerbated by ambulation and exercise. The patient also endorsed significant difficulty with walking due to her leg length discrepancy. She had failed a total of 6 months of conservative treatments including medications, physical therapy, cortisone injections, pain management, weight loss, and shoe lifts. The patient denied any history of trauma or injury to her hip, but she did endorse a history of left hip

No author associated with this paper has disclosed any potential or pertinent conflicts which may be perceived to have impending conflict with this work. For full disclosure statements refer to <https://doi.org/10.1016/j.artd.2019.10.006>.

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<https://doi.org/10.1016/j.artd.2019.10.006>

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surgery to correct her DDH 20 years ago that was complicated by infection requiring multiple surgical debridements. Although left with chronic hip pain and a leg length discrepancy, she was able to tolerate her symptoms until shortly prior to presentation, when the degree of pain with ambulation had progressively become intolerable.

Physical examination

The left hip joint exhibited flexion that was limited to 80°, but otherwise had full range of motion compared to the contralateral hip with neutral extension and 40° abduction, internal, and external rotation. The left lower extremity measured 5 cm shorter than the contralateral side on examination. A small anterolateral scar was noted from her previous surgery. Pain was elicited with flexion and internal rotation of the left hip, and the patient ambulated with an antalgic gait. She preferred to avoid using shoe lifts or assistive devices.

Imaging and laboratory results

Radiographs and computed tomography scan of the pelvis displayed left hip dysplasia with a high hip center, a superolaterally shifted weight-bearing dome, and a shallow and retroverted acetabulum (Figs. 1-3). The left hemipelvis displayed significant dysmorphism due to both external rotation and extension of the ilium relative to the contralateral hemipelvis. The affected hemipelvis also had an overall atypical morphology, with an autofused left SI joint that descended rapidly into the anterior and posterior columns of the acetabulum. This dysplastic acetabulum then transitioned into a vertically oriented left ischium and pubis. There was severe flattening of the femoral head shortening of the femoral neck. The hip joint demonstrated advanced degenerative changes with near-complete obliteration of the joint space. Some osteopenia and scoliosis of undetermined curvature were also noted. Leg length was measured from a line tangent to the distal border of the inferior pubic rami to a parallel line at the level of each lesser trochanter. The left lower extremity was radiographically noted to be 10 cm shorter than the right lower extremity. Serum white blood cell count (4.6×10^9 cells/L), erythrocyte sedimentation rate (8 mm/h), and C-reactive protein (1.9 mg/L) were obtained and found to be within normal limits. Hence, there was low concern for ongoing infection in the hip joint.



Figure 2. Coronal computed tomography of the left hip demonstrates flattening of the femoral head with a significantly shortened femoral neck. There is near-complete obliteration of the joint space, and cystic and sclerotic changes are noted in both the femur and acetabulum. The left SI joint is seen in this coronal cut and noted to have undergone autofusion, with only a short corridor of bone bridging the posterior ilium to the acetabulum. The axial computed tomography cut demonstrates slight acetabular retroversion, which is augmented by native retroversion of the left hemipelvis.

Preoperative planning

Having failed conservative management, the patient was counseled that the most realistic long-term solution for her



Figure 1. Anteroposterior pelvis and attempted frog-leg lateral of the left hip demonstrate left hemipelvic dysmorphism with a dysplastic left hip joint. The left hip center is situated superolateral relative to the contralateral side, and the acetabulum is shallow with a superolateral shift of the weight-bearing dome. The femoral head has undergone significant flattening and the neck is markedly shortened. Collectively, these findings have resulted in an approximate leg length discrepancy of 10 cm.

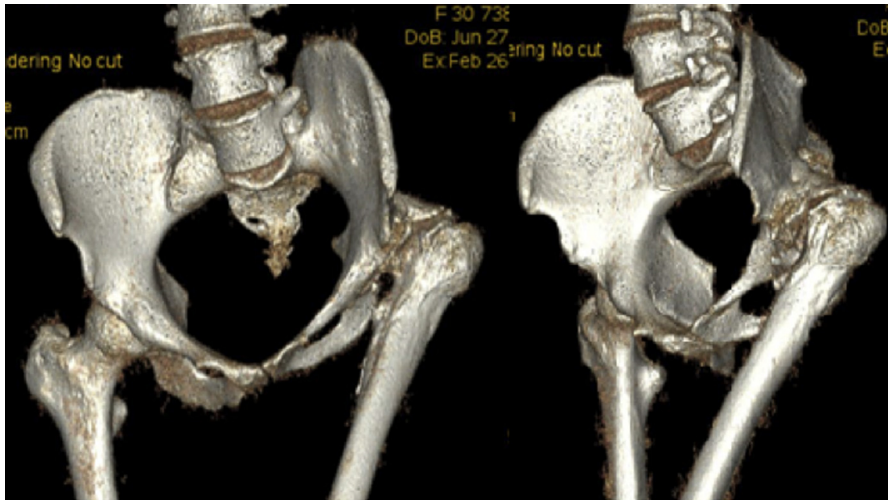


Figure 3. Three-dimensional reconstructions of the pelvic CT scan demonstrate left SI joint autofusion and abnormal external rotation and extension of the left ilium relative to the contralateral hemipelvis.

symptomatic left hip arthritis was THA. Expectations with leg lengthening were discussed, and the patient was informed that although she would gain some length in her left lower extremity, it would not equal the right side due to her pelvic anatomy. She was informed about the risk of traction injury to the sciatic nerve from limb lengthening or retractor placement. She was also counseled on the possibility of requiring additional osteotomies for exposure and reduction of the hip. The patient was

additionally informed about the risks of instability, dislocation, periprosthetic fracture, loosening, infection, and possible requirement for additional surgery. The DePuy Synthes S-ROM Modular Hip System and GRIPTION TF Acetabular Revision System were selected due to the extent of required reconstruction. As part of preoperative planning, a 3-dimensional printed model of the hip joint was obtained to better characterize the patient's anatomy (Fig. 4).

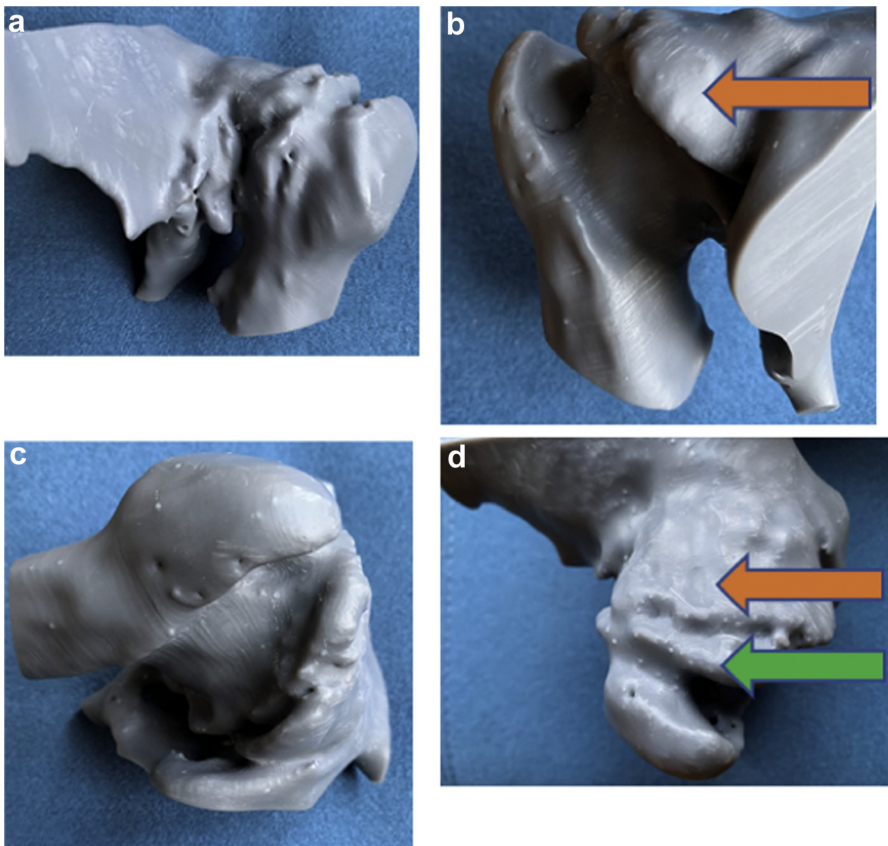


Figure 4. Three-dimensional printed model of the patient's hip joint. (a) Anterior view of the dysplastic hip joint. (b) Posterior view of the dysplastic hip joint (orange arrow indicates shelf). (c) View of the hip joint during the posterolateral approach to the hip. (d) Superior view of the pelvic shelf (orange arrow indicates shelf; green arrow indicates femoral head).

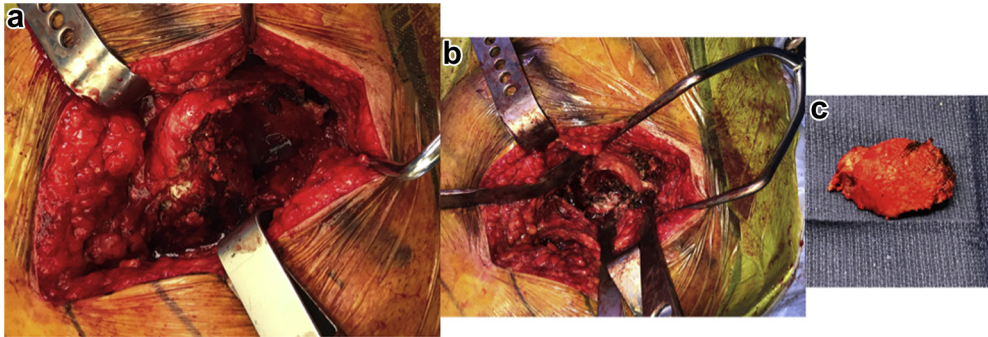


Figure 5. Intraoperative images of total hip arthroplasty. (a) Exposure of the femur after hip dislocation and osteotomy of the femoral neck. (b) Acetabular exposure demonstrating a large superior shelf with retroversion of the acetabulum. (c) Femoral head specimen after osteotomy. Note the severe dysplasia with flattening of the usual spherical contour of the femoral head along with minimal residual femoral neck.

Operative technique

Informed consent was obtained from the patient after extensive discussion of the above risks and benefits of surgery. A posterolateral approach to the left hip was utilized with the patient in the lateral decubitus position. The external rotators and capsule were separately tagged and retracted, and the hip was dislocated posteriorly. The femoral neck was cut at the previously templated level, with the plan to measure the native femoral head size to guide implant sizing. However, due to the severe dysplasia noted on the femoral head, measurements could not be taken to guide implant size. Frozen tissue section was obtained and demonstrated fewer than 5 neutrophils per high-powered field in 5 total fields, indicating the absence of acute inflammation.

A significant anterior capsular release was required in order to properly place the anterior acetabular retractors. In addition, significant portions of the superior and inferior pubic ramus were exposed in order to fully visualize the acetabulum due its atypical position. A drill was used to determine the depth of the acetabulum and locate the optimal position for the acetabular reamer. The acetabulum was noted to be dysplastic with a large superior shelf and 20° of retroversion (Fig. 5). Following reaming, a size 48 multi-hole GRIPTION cup was placed in the acetabulum in approximately 40° of abduction and 25° of anteversion. Three iliac screws and 1 ischial

screw were placed through the cup. The superior shelf was used to stabilize the acetabular component from migrating proximally. A size 32+4 neutral highly cross-linked polyethylene liner was placed.

Attention was then turned toward the femur. Proximal and distal reaming was required for stem and sleeve placement, and ultimately, a 13 × 160 stem (11/13 taper), 18B small cone, 32+0 ceramic head, and size 30 standard offset DePuy S-ROM stem neck in 10° of anteversion were utilized. The hip was found to be stable in 90° of flexion and 60° of internal rotation, as well as the position of sleep (flexion, adduction, and internal rotation). With extension and external rotation, there was no anterior instability. The combined anteversion was approximated to be 35°. The leg length discrepancy was significantly improved but still not symmetric to the contralateral extremity. The capsule and external rotators were sequentially repaired, and the soft tissues were closed in a layered fashion. The patient was awakened and found to be neurovascularly intact in the left lower extremity.

Postoperative course

Immediate postoperative radiographs demonstrated appropriately positioned acetabular and femoral components with no evidence of acute fracture or dislocation (Fig. 6). The patient was made weight bearing as tolerated with posterior hip precautions for a

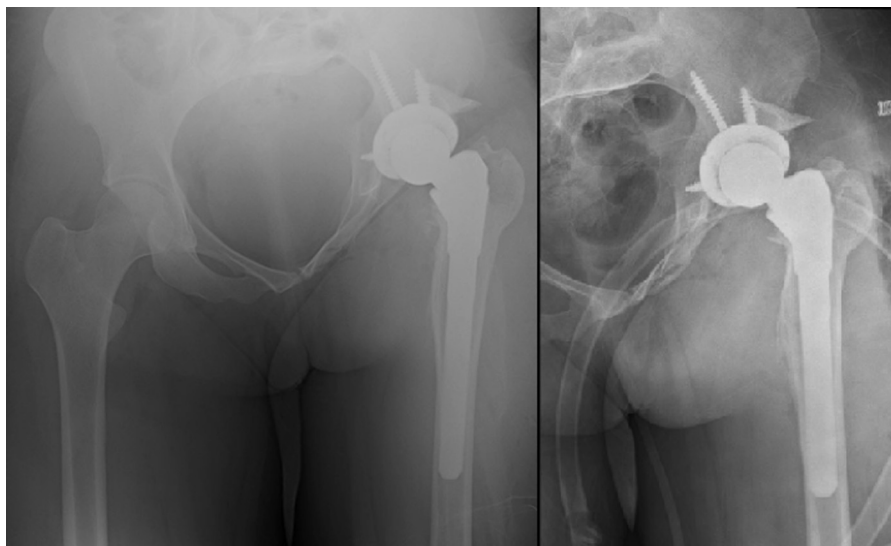


Figure 6. Anteroposterior pelvis and anteroposterior left hip radiographs taken immediately postoperatively demonstrate total hip arthroplasty with an appropriately positioned acetabular component, screws, and femoral component. A residual leg length discrepancy of 5 cm is noted on the anteroposterior pelvis radiograph.



Figure 7. Most recent weight-bearing radiograph at 13 months postoperatively demonstrates stable acetabular and femoral component position with no evidence of complication.

period of 6 weeks postoperatively. On postoperative day 1, she was started on aspirin 325 mg by mouth twice daily for 6 weeks for venous thromboembolism prophylaxis. Her inpatient postoperative course was uncomplicated, and she was discharged home with outpatient physical therapy on postoperative day 3. She was seen in the office 18 days postoperatively and was pleased with her progress at the time. Physical examination in the supine position detected a subtle residual leg length discrepancy of 0.5 cm, but on standing radiographs, this difference was noted to be 5 cm, improved from 10 cm preoperatively. At 6 months of follow-up, the patient was utilizing a cane/walker to ambulate, participating in physical therapy, and no longer requiring narcotic pain medication. Her previously painful and restricted hip flexion had improved to 100° of painless flexion. At 1-year follow-up, she was still participating in physical therapy but was no longer utilizing a cane/walker for ambulation (Fig. 7). The patient is currently being followed at annual office visits.

Discussion

To our knowledge, this is the first reported case of primary THA in an adult patient to treat osteoarthritis of the hip secondary to DDH with concomitant pelvic dysmorphism. Recent literature reports that DDH is a more complex spectrum of anatomic abnormalities than commonly thought. Although local dysplasia of the acetabulum is clinically important, DDH is associated with subtle global abnormalities of the pelvis [2]. In the case of this patient, although her past surgical history was unclear, it is more likely that the severe degree of pelvis dysmorphism is due to congenital abnormalities rather than iatrogenic causes. Other cases of severely dysplastic pelvises in the setting of DDH have been noted in the literature. Soylemez et al. [4] reported on the case of an 18-month-old male with aplasia of the right pubis with associated DDH. Bashyal et al. [5] also reported on a similar case of pelvic dysmorphism in a 44-year-old woman with associated absence of pubic rami, DDH, and genitourinary anomalies. No operative intervention was reported in these cases. Other examples of unique

Table 1
Crowe and Ranawat classification for hip dysplasia in adults.

Grade	Proximal displacement	Femoral head subluxation
I	<10% vertical height of pelvis	Proximal migration of head-neck junction from interteardrop line by <50% of femoral head vertical diameter
II	10%-15%	50%-75%
III	15%-20%	75%-100%
IV	>20%	>100%

pelvic dysmorphism reported in the literature include hypoplasia of parts of the pelvic ring, most commonly involving the pubis as with the previously noted case reports [6–8]. No common genetic risk factors were noted in these cases. One theory suggests that a post-zygotic mutation in a segment of mesenchymal cells may be the cause of defects in an isolated anatomic area of the pelvis, similar to Poland Syndrome [5]. Another theory is that disrupted blood supply to a specific anatomic area during embryonic development may also contribute to isolated pelvic agenesis. However, research elucidating the pathogenesis of isolated pelvic and hip congenital defects remains limited. Furthermore, there is no clear literature on the specific type of hemipelvic dysmorphism characterized in this case report.

As noted earlier, the patient in this case had an unclear operation—possibly a pelvic shelf osteotomy—to correct her DDH, which ultimately resulted in a surgical site infection. In the literature, early operative intervention for DDH, such as periacetabular osteotomy, is commonplace and has good long-term functional outcomes, with infection being a relatively uncommon complication [9–11]. Those few patients who had postoperative infection of their hip joint required antibiotic therapy and surgical debridement. However, some patients required subsequent THA 2–3 years afterward due to accelerated degenerative changes secondary to infection. It is likely that our patient's DDH, infection from corrective surgery, and concomitant pelvic dysmorphism collectively accelerated the patient's need for early THA.

In cases of advanced DDH (Crowe-Ranawat grade III or IV; Hartofilakidis type C) for which THA is indicated in the skeletally mature patient, certain modifications to the surgical technique and implant selection can simplify the procedure and reduce the risk of complications (Tables 1 and 2). With respect to acetabular component positioning, early reports noted good long-term results with utilization of a high hip center. However, many subsequent studies have shown that the true acetabulum is the best position for supporting the cup due to better bone stock, less shearing forces on the acetabular component, and improved hip biomechanics [1,11–15]. A smaller acetabular component must often be used for optimal fit, and bone grafting may be beneficial in restoring the true acetabulum. However, the surgeon must be cognizant of the risks of using a small cup (ie, dislocation) and the risks of using bone graft, such as graft resorption and subsidence [16]. Regarding bearing surfaces,

Table 2
Hartofilakidis classification for hip dysplasia in adults.

Dysplasia (type A)	Femoral head subluxated but not dislocated. Segmental deficiency of superior wall. Secondary acetabular shallowness due to fossa-covering osteophytes
Low dislocation (type B)	Femoral head dislocated, creating false acetabulum superior to true acetabulum. Complete absence of the superior wall. Inadequate depth of true acetabulum
High dislocation (type C)	Femoral head is completely uncovered by the true acetabulum and has migrated superiorly and posteriorly. Complete deficiency of entire acetabulum with excessive anteversion

metal-on-polyethylene has the longest follow-up data, but due to the typically young age of patients undergoing THA for DDH, ceramic-on-polyethylene is now most often used due to the improved material wear properties [17]. Dual-mobility cups may improve stability, but recent literature demonstrates that the dislocation rate of THA for DDH is comparable to the dislocation rate of THA for OA [18]. In Wang et al.'s series of 820 hips undergoing THA for DDH, the dislocation rate was reported at 2.93%, and the key factor in dislocation risk was a femoral head size of 28 mm (as opposed to 32 mm), illustrating the importance of using an adequately sized cup-head combination. Furthermore, 69.6% of these dislocations were anterior, and the degree of cup anteversion and combined anteversion was significantly higher in the anteriorly dislocating group, indicating that the surgeon may need to account for the anatomic anomalies intrinsic to DDH when setting version. Furthermore, as new literature emerges on spinopelvic balance, surgeons must be mindful of acetabular component anteversion to account for compensatory changes in the spine. In our case, due to the patient's SI joint autofusion, pelvic dysmorphism, and dysplastic acetabular anatomy, we had to be cautious with regards to placement of the acetabular component. Hence, we opted to use a femoral component that enabled us to achieve version control, effectively allowing for full control of the hip's combined anteversion.

In restoring the anatomic center of hip rotation in cases of THA for severe DDH, the limb may need to be lengthened greater than 4 cm, placing neurovascular structures at risk of traction injury and increasing the risk of aseptic loosening of components due to excessive joint reactive forces from over-tensioned abductors [19]. In cases where this degree of lengthening is required, subtrochanteric shortening osteotomy of the femur can be performed, which allows for rotational correction of the often over-anteverted femoral neck, in addition to facilitating safe reduction of the hip without compromising neurovascular structures [13,19,20]. If an osteotomy is used, a modular stem, such as the one used in this case, may be beneficial to correct for the often over-anteverted femoral neck seen in DDH, provide good fit in the often hypoplastic femoral canal, obtain torsional stability at the osteotomy site, and allow for easy control of femoral offset and leg lengths [21].

For cases in which there is question of neurologic integrity after restoration of the anatomic hip center, intraoperative neurologic monitoring or the wake-up test can be utilized [22,23]. Sutter et al. reported on a series of 69 patients who underwent multimodal intraoperative monitoring during complex hip surgery and found that in 35% of cases, the surgeon was alerted about a possible lesion to the femoral or sciatic nerve, most commonly when performing periacetabular work. Although there was 1 case of clinical neurologic injury in this series, the authors concluded that intraoperative neurologic monitoring is an effective means to reduce the risk of neurologic injury during complex hip surgery [23]. Chen et al. reviewed 22 THAs performed for Crowe type IV DDH that underwent an intraoperative wake-up test to check for nerve function. One patient was found to have lost ankle dorsiflexion during the wake-up test, and the surgeon responded by reducing limb length by an additional centimeter. The authors concluded that the wakeup test is a simple, safe, and reliable alternative to intraoperative neurologic monitoring and may be beneficial when correcting leg lengths in cases of severe DDH [22].

In addition to the patient's DDH, infection history, and pelvic dysmorphism, the patient's unique anatomy also consisted of an autofused ipsilateral SI joint. SI joint dysfunction is thought to be closely linked to hip arthritis [3]. In addition, pathology of the SI joint has recently been implicated in altering global pelvic anatomy. Patients with SI joint dysfunction and arthritis often have abnormal rotation and tilt of the pelvis secondary to asymmetry of the right and left innominate bones [24]. Cibulka [24] propose that SI joint

disease may be responsible for improper acetabular coverage more so through alteration of global pelvic anatomy rather than by causing local abnormalities in acetabular morphology. Morgan et al. reported that up to 21% of patients with SI joint disease had acetabular retroversion on hip radiographs, which may be a contributing factor to accelerated wear of the hip joint [25]. Thus, it is plausible that ipsilateral SI joint autofusion played a role in our patient's abnormal hemipelvic tilt and acetabular coverage, ultimately compounding distortions already present from the patient's anomalous pelvic and acetabular morphology. Since we did not modify the SI joint morphology, nor did we correct the hemipelvic tilt, we may notice faster wear of the bearing components. Given the rarity of such cases, it is difficult to estimate a life span of total hip replacements in these patients.

Summary

Overall, this case report adds to the growing body of literature that demonstrates an association between gross pelvic dysmorphism and DDH [2,26]. To our knowledge, this is the first report in the literature demonstrating positive outcomes of THA in a patient with pelvic dysmorphism, DDH, and ipsilateral SI joint autofusion. Further research is needed to determine the long-term outcomes of THA in patients with DDH and significant pelvic dysmorphism, but this case report provides preliminary evidence that THA is a viable and effective treatment option for patients with debilitating hip arthritis secondary to congenital and developmental abnormalities of the pelvis and hip joint.

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