



Arthroplasty in patients with rare conditions

Bilateral Total Hip Arthroplasty in the Setting of Developmental Dysplasia of the Hip and Extreme Hip Flexion Requirements due to Phocomelia

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ARTICLE INFO

Article history:

Received 15 October 2020

Received in revised form

7 January 2021

Accepted 21 January 2021

Available online 23 February 2021

Keywords:

Total hip arthroplasty

Hip dysplasia

Phocomelia

Limb defect

Contracture

Computer navigation

ABSTRACT

Phocomelia is a rare congenital birth defect marked by hypoplastic or markedly absent limbs. Developmental dysplasia of the hip (DDH) is a congenital disorder with a failure of the native acetabulum to provide complete coverage over the femoral head. The secondary osteoarthritis that develops from DDH is technically challenging for orthopedic surgeons because of distorted anatomy. The present case describes the diagnosis of Crowe 3 DDH in a phocomelia patient with hyperflexion requirements who successfully underwent staged bilateral total hip arthroplasty via a direct anterior approach. It highlights the utility of preoperative computerized tomography and intraoperative computer navigation to assist in implant placement. Recognizing difficult arthroplasty cases in advance is imperative as these cases may require great expertise and more extensive surgical planning.

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Introduction

Phocomelia is a congenital birth defect with an estimated incidence of 4.2 per 1 million births [1]. It is marked by prominent changes to musculoskeletal anatomy including hypoplastic or markedly absent limbs in both upper and lower extremities [2]. Thalidomide exposure during pregnancy and the development of phocomelia in an embryo is a well-documented association [3,4]. Based on the absent portion of limb relative to the trunk, phocomelia can be classified into 3 categories [5]. Type 1 or complete phocomelia describes the complete absence of proximal and distal limbs of an extremity in which the hands or digits are directly attached to the trunk. Type 2 or proximal phocomelia describes the absence of the proximal limb in which the humerus is missing, and the forearm with hands and digits are attached to the trunk. Finally, type 3 or distal phocomelia describes the absence of the distal limb in which the forearm is absent, and the hand or digits are directly attached to the humerus [6]. In patients with phocomelia who have

multiple congenital anomalies, congenital malformation of the hips has been reported to be present in 11.5% of nonsyndromic cases [7].

Developmental dysplasia of the hip (DDH) is a congenital disorder resulting from abnormal acetabular development with a failure of the native acetabulum to provide complete coverage over the femoral head [8]. The estimated incidence of DDH is 1 to 5 per 1000 live births [9,10]. With high interobserver ($\kappa = 0.92$) and intraobserver ($\kappa = 0.95$) reliability [11], the Crowe classification system was created to quantify the severity of femoral head subluxation relative to the native acetabulum [12]. It categorized dysplastic hips based on the degree of proximal migration of the medial head-neck junction above the inferior margin of the acetabulum or the amount of proximal displacement of the femoral head relative to the pelvic height [13].

Given that symptomatic osteoarthritis forms secondary to DDH, surgical options such as hip arthroscopy, periacetabular osteotomy, salvage pelvic osteotomy, hip resurfacing, and total hip arthroplasty (THA) may be considered in select patients based on age and condition severity [14,15]. The purpose of this article is 2-fold: (1) to present the clinical course of a patient with phocomelia who had severe osteoarthritis secondary to Crowe 3 DDH with hyperflexion requirements undergoing staged bilateral THA and (2) to discuss

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the clinical and surgical considerations for managing these patients.

At the time of submission, the patient had consented for her unique case to be published in medical literature and elected to provide information about her medical condition.

Case history

Case 1

A 44-year-old female with multiple previous bilateral hand surgeries and a previous right knee surgery during childhood presented with severe bilateral hip pain. She was born with idiopathic phocomelia of her bilateral upper extremities, in which her bilateral forearms including her radius and ulna did not form, and her hands were directly attached to her distal humerus bilaterally. Her medical history included thrombocytopenia and hypothyroidism for which she was taking levothyroxine. Social history was negative for alcohol or tobacco, and she worked in an office setting at a desk job. She routinely performed activities requiring excessive bilateral hip flexion because of accommodations made during development with foreshortened limbs for her activities of daily living (ADLs) including opening doors, picking up objects from the floor, and other tasks normally assigned to arm usage.

On physical examination, she was 5'0" weighing 112lbs. and had a waddling gait with a 4-point walker because of leg length inequality and flexion contracture of the right knee (Image 1). She walked in a flexed forward position of the lumbar spine with prominent hyperlordosis, causing concern for an increased risk of dislocation postoperatively (Image 2). Her left hip was shortened compared with her right hip by 2 cm, and she had increasing bilateral hip pain during range of motion testing. Left hip flexion was up to 115° with 4/5 strength, with a range of motion of 0° internal rotation and 30° of external rotation with extension limited because of positive impingement. Her left knee range of motion lacked 5° of full extension and was only able to achieve active and passive flexion of 115°. Right hip flexion was up to 115° with 4/5 strength, with a range of motion of 5° internal rotation and 40° external rotation. Her right knee had a flexion contracture of 20° and flexed to only 100° actively/passively. Preoperative AP films of the pelvis, lateral right hip, and lateral left hip can be seen in Figure 1. There was moderate femoral head subluxation of the left hip and prominent osteophytes consistent with a Crowe 3 classification. Her right hip demonstrated a loss of joint space and eburnation of the articular surface consistent with Crowe 1 DDH. After failure of all nonsurgical measures of pain control, all treatment options were discussed, and the patient elected to proceed with a staged bilateral THA.

She was taken to the operating room for a left THA by S.W.K., who is a large-volume hip reconstruction surgeon highly experienced in direct anterior hip arthroplasty. Intraoperatively, the patient was given general and spinal anesthesia. A direct anterior approach was performed with great difficulty per surgeon record, and an OrthoMap hip navigation system (Stryker Ltd., Kalamazoo, MI) was used to determine the final cup inclination of 36° and anteversion of 28°. A size 44 acetabular cup from the Trinity system (Corin, Tampa, FL), which had a 45-mm diameter with a 20° lipped liner, was placed with the raised portion placed posterior-superiorly. Owing to small and dysplastic nature of the hip, the femur was unable to be broached because of limited access, and the intermedullary canal of the femur was reamed. A size 14 femoral stem from the Wagner Cone Prosthesis system (Zimmer Biomet, Warsaw, IN) with a 28-mm head and Vitamin E on ceramic bearing were then implanted. Estimated blood loss for the procedure was 225 milliliters. Postsurgical films after the first procedure

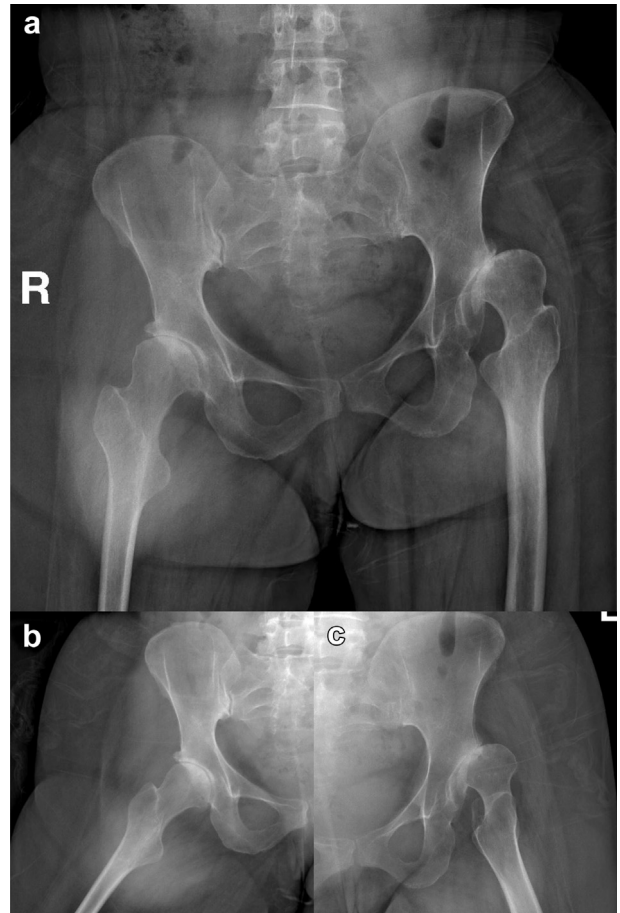


Figure 1. Preoperative films displaying severe bilateral hip osteoarthritis secondary to Crowe 3 DDH of the left hip and Crowe 1 DDH of the right hip. Anteroposterior radiograph of the pelvis seen in top (a), lateral of the right hip seen in bottom left (b), and lateral of left hip seen in bottom right (c).

demonstrated a well-seated acetabular cup with satisfactory component positioning as seen in Figure 2. Patient was weight-bearing as tolerated for bilateral lower extremities during this postoperative course. Her drain was removed 24 hours after the procedure. She had a normal neurovascular exam postoperatively and maintained that status throughout the recovery period. She went through the standard anterior total joint replacement rehabilitation protocol, and after months of physical therapy, she was taken back to the operating room 7 months later because of persistent symptomatic osteoarthritis of her right hip.

During her right THA, the patient was given general and spinal anesthesia for the procedure. A direct anterior approach was performed with great difficulty per surgeon record, and a Stryker navigation system was used to determine the final cup inclination of 44° and anteversion of 25°. All implants of identical type and size as her first THA were placed successfully in the same manner as the first procedure. The patient did, however, sustain a calcar fracture intraoperatively, which required placement of a cerclage cable (Fig. 3). Estimated blood loss was 225 milliliters. Postoperatively, she was made to avoid weight-bearing on her right lower extremity for 6 weeks to allow for healing before beginning progressive weight-bearing. Her drain was removed 24 hours after the procedure. She was neurovascularly intact postoperatively and remained intact throughout her recovery. Her leg lengths were equal clinically after this operation. Her rehabilitation course was

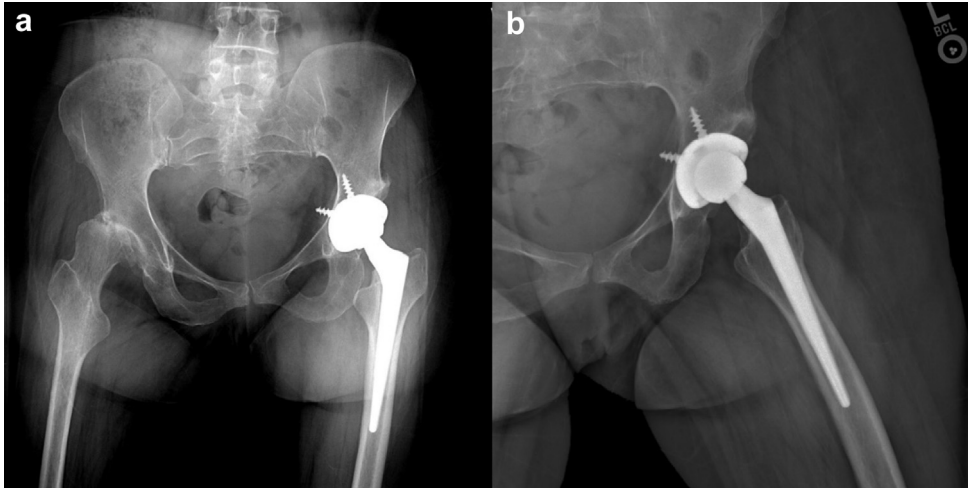


Figure 2. One-month postoperative films after left direct anterior total hip arthroplasty. Anteroposterior radiograph of the pelvis seen in top (a) and lateral of left hip seen in bottom (b).

uncomplicated with no requirements of transfusion and no post-surgical events.

During her follow-up visit at 1 year from her second procedure, the patient was independently ambulating with the use of her 4-point walker and had no complaints of hip pain. Radiographic

and clinical examination confirmed bilateral stable hips with no evidence of loosening, infection, subluxation, dislocation, or peri-prosthetic fracture (Fig. 4). She is now nearly 5 years from her THAs and has not had any dislocations or adverse events. She is highly satisfied with her results without any complaints.

Discussion

In the present case, after being born with type 3 distal phocomelia and bilateral hip dysplasia, this patient had difficulty performing ADLs with bilateral hip pain during ambulation. Her condition was exacerbated by the routine use of her legs to perform activities that would generally be associated with arm usage, which became progressively more painful and difficult. After failing nonsurgical management with a goal of pain-free ambulation, the patient elected to proceed with staged bilateral THA. Owing to the severity of pain and difficulty with function, as well as her leg length was most affected by the left leg, the patient elected to move forward with the left hip as the first procedure. Robotic assisted THA and computer navigation were initially designed to aid orthopedic surgeons in improved surgical planning, component positioning, and accurate implantation of prosthetics [16,17]. With a dysplastic acetabulum and likely need for a femoral stem with the ability to alter version, it was felt that this could be done safely through an anterior approach using computer navigation assistance for placement of the acetabulum. A direct anterior approach was specifically selected for each hip as it was felt that a posterior approach may not have given the patient the postoperative stability of an anterior approach with regard to her risk of posterior dislocation while performing her normal activities with her legs especially in the immediate postoperative period [18–21].

Preoperative computerized tomography (CT) and intraoperative computer navigation were used for accurate acetabular cup positioning with appropriate cup inclination and anteversion. The CT scan was also useful in determining if there were deficiencies of the anterior or posterior walls of the acetabulum, which would have precluded the ability to use an acetabular cup without augments, a femoral head autograft technique, or a cage construct. In addition, the CT scan helped estimate the cup size during surgical planning. Owing to the Corin Trinity system at the operative surgeon's institution containing a 44-mm acetabular cup as the smallest sized implant in the set, another acetabular system may have had to be readily available if this patient would have required it. Although the operative surgeon uses computer navigation in all hip arthroplasty

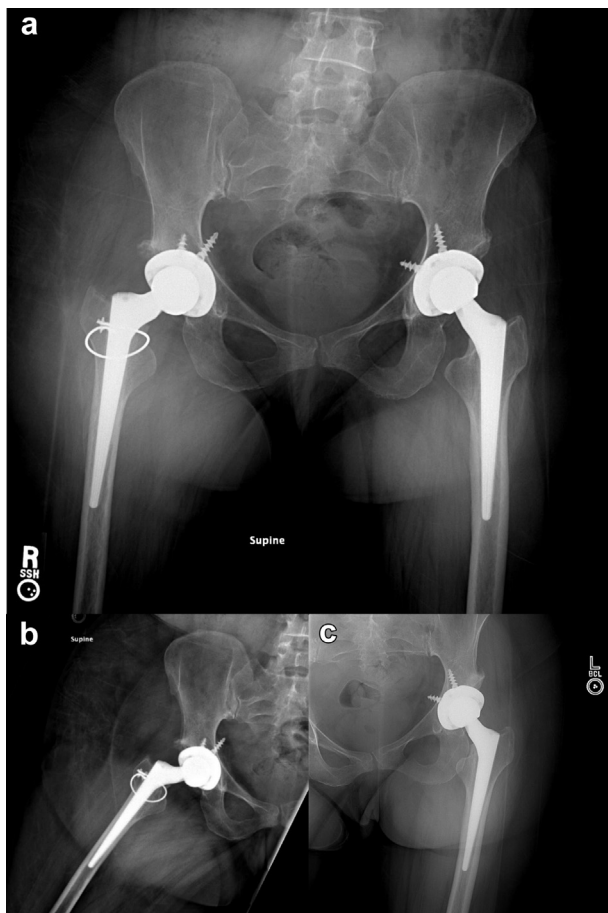


Figure 3. One-month postoperative films after right direct anterior total hip arthroplasty. Anteroposterior radiograph of the pelvis seen in top (a), lateral of the right hip seen in bottom left (b), and lateral of left hip seen in bottom right (c). Of note, cerclage cable intact over calcar region of right hip.

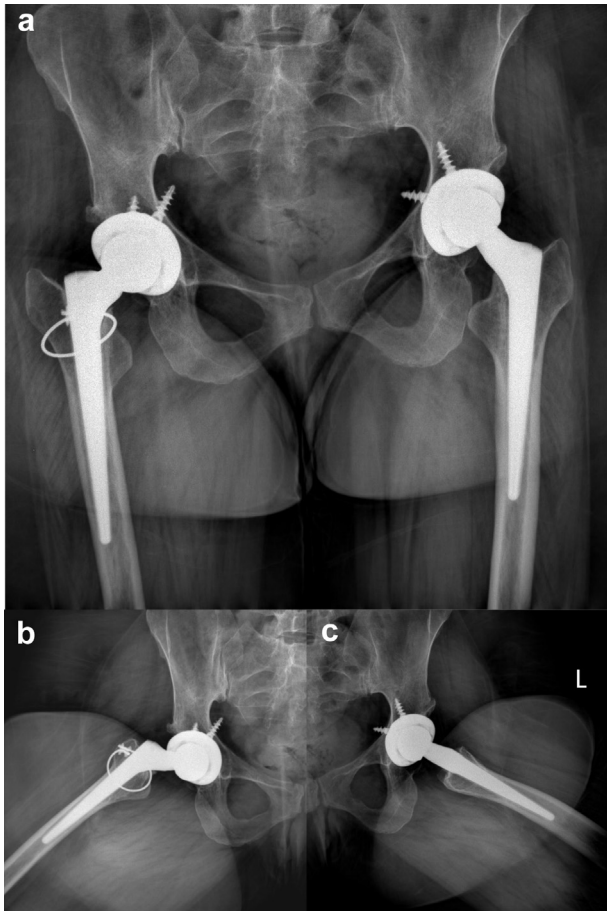


Figure 4. One-year postoperative films after right direct anterior total hip arthroplasty. Anteroposterior radiograph of the pelvis seen in top (a), lateral of the right hip seen in bottom left (b), and lateral of left hip seen in bottom right (c).

cases, it was especially useful in achieving the target socket position, anteversion, and inclination parameters as well as checking for the position of components and leg length equalization. Computer navigation during this case provided the operative surgeon assistance not only during the direct anterior surgical approach but also during implant fixation. This patient's distorted anatomy impaired direct visualization and made it difficult to use surgical landmarks during the exposure. Through a smaller surgical exposure than that anticipated, the operative surgeon used navigation to help predict the hip center and height so accurate component positioning could be achieved.

The intraoperative calcar fracture sustained during this patient's right THA is not uncommon in patients with dysplasia undergoing THA. Ollivier reviewed 28 consecutive cementless THAs performed in patients with Crowe IV DDH and found intraoperative fractures in 4 of 28 surgeries (14.3%) [22]. In present case, the operative surgeon chose to address the intraoperative femur fracture by placing a cerclage cable. Faldini reported the utility of cerclage wiring and 5 weeks of restricted weight-bearing in treating an intraoperative proximal femur fracture sustained during stem implantation in a patient with DDH [23]. In the current case, the patient was made to avoid weight-bearing for 6 weeks with progression to full weight-bearing thereafter. Schmotzer's study further demonstrated cerclage cables to augment fixation by providing additional strength and stability to intraoperative femur fractures [24]. Therefore, cerclage cabling may be a suitable option for surgeons facing similar circumstances.

Congenital abnormalities such as DDH pose a challenge to orthopedic surgeons because of the frequent requirements of patients with these conditions to have multiple musculoskeletal surgeries to improve functionality [25]. Patients with DDH develop secondary osteoarthritis and undergo THA at the earlier average age of 53 years in comparison to patients who undergo THA for idiopathic osteoarthritis at an average age of 65.5 years [26]. THA is a technically challenging procedure in the treatment of DDH because of several reasons including but not limited to deficient acetabular bone stock, increased anteversion, muscle contractures, leg-length discrepancy, reconstructing a distorted acetabulum, and component failure [27]. Although THA is difficult in patients with DDH, multiple studies have reported improvements in long-term outcomes when using cementless stems at 10-year follow-up with no revisions or evidence of aseptic loosening [23,28]. In the present case, the narrow, tubular, and sclerotic nature of the femoral canal of this patient's proximal femur prevented broaching access by even the smallest implant in the set. Therefore, the operative surgeon selected to use a 360° tapered stem, which did not have a proximal flare similar to a broached implant. This modular implant also has a variety of components allowing for multiple offsets and length options to correct the patient's distorted anatomy. The Wagner cone stem was specifically selected in this patient for survivorship and because of her anatomical aberrations, such as a cylindrical configuration of the proximal medullary canal of the femur with coxa valga and increased anteversion. This femoral stem also specifically allowed for adjustment to the angle of version as reported in the literature [29,30]. Furthermore, Heckmann reported abnormal spinopelvic motion as a risk factor for late hip dislocations with spinopelvic stiffness specifically associated with increased age and increased femoral motion, which could lead to potential impingement and dislocation [31]. With the patient's excessive lordosis in the present case, there was concern for pelvic instability with a risk of dislocation, and therefore, the acetabular cup was placed in more anteversion to adjust for pelvic tilt.

In this patient, a staged bilateral THA was chosen in comparison to a simultaneous bilateral THA to address the debilitating hip pain secondary to Crowe 3 DDH osteoarthritis with concurrent radiographic evidence to minimize the burden of surgical stress on the body and allow for the patient to rehabilitate one limb at a time. Multiple studies have reported a reduction in hospital stay, shorter anesthesia duration, and an improvement in cost-effectiveness when performing simultaneous bilateral THA for the average patient [32,33]. However, with her increased medical comorbidities, a staged bilateral THA was selected for this patient taking into account the risks for systemic complications [34]. Moreover, with lower extremity nerve injury being attributed to leg lengthening at a threshold as low as 0.5 cm during THA, there was concern for tension on the sciatic nerve and development of a foot drop in our patient [35]. Therefore, a staged bilateral THA was offered to this patient to allow her adequate rehabilitation while minimizing the risks of neuropraxia and falls.

When performing THA on patients with DDH, acetabular cup placement may require additional fixation with transacetabular screws when insufficient osseous coverage or osteopenic bone is present as demonstrated in this case. Although placement of transacetabular screws augments fixation, it also increases the risk of intrapelvic screw penetration and the possibility of injuring the surrounding neurovascular structures. In Wasielewski's first anatomical and radiographical study for transacetabular screw placement during uncemented THA in anatomically normal patients, the posterior-superior and posterior-inferior acetabular quadrants were determined to be the original safe zones compared with the anterior-superior and anterior-inferior quadrants, which posed danger to external iliac artery and vein in addition to the

obturator nerve, artery, and vein [36]. However, it is important to note safe zones change when a patient has a severe DDH or a high hip center. In a subsequent study by Wasielewski et al. [37] examining transacetabular screw placement in patients with high hip centers, the anatomic antero-superior and antero-inferior quadrants were reported to be located in the original posterior-superior quadrant. Owing to this positioning, it was determined the peripheral half of the posterior-superior quadrant was the safest location for the transacetabular screw placement. Furthermore, Liu et al. [38] later demonstrated the quadrant system developed by Wasielewski et al. may be less applicable for patients with DDH as the original safe zones were shifted in cases of dysplasia, and the posterior-superior quadrant was no longer safe. In the present case, there was medial screw penetration as the shortest screw length available with the instrumentation in the Corin system used is 15 mm, and its use was required for additional acetabular cup stability. In dysplastic cases such as this, with known smaller anatomical features, it would be beneficial to also have smaller screws available for use than what is customarily included in a set.

This case was particularly challenging because of multiple factors such that the patient was shorter than the average height resulting in smaller anatomical structures. Owing to the smaller than anticipated muscle planes developed during the surgical approach, there was a tight window to operate. This created a challenge when using the standard instruments and retractors for THA as there were no pediatric instruments available for this patient's hip arthroplasty. In addition, this patient's distorted anatomy made it difficult to judge the cutting position when removing the femoral head during the femoral osteotomy. Placement of the acetabular cup proved to be difficult because of the patient's dysplasia and created a narrow margin of error during the reaming process. In the operative surgeon's experience, patients with an abnormal anatomy and hip dysplasia typically have poor bone quality. When this poor bone quality is coupled with a small acetabular cup size, there is an increased chance to ream out the posterior acetabular wall. In addition, this patient presented with significant femoral anteversion. Intraoperatively, this was difficult to assess as the operative surgeon had to judge, predict, and correct this femoral anteversion to assure hip stability. Owing to this patient's condition, various implants had to be readily available, which provided the surgeon with the opportunity to adjust for distorted anatomy by having the freedom to select from different leg length offsets and femoral version options.

Throughout this patient's life, she was reliant on her adjustable 4-point cane for ambulation. However, designs of canes can vary among different manufacturers. This patient's absent forearm presented a unique challenge as she was unable to use a traditional one-point cane. An adjustable cane was critical to her recovery as the cane could be lengthened to reach the distal aspect of her upper limbs to provide support by reducing weight-bearing forces in the lower extremities. Moreover, a 4-point cane provided additional stability by creating a broader base with a low center of gravity. In one study, Kumar et al. [39] reported that canes should be fitted by measuring the ideal cane length from the floor to the distal wrist crease to produce the optimal 20° to 30° of elbow flexion. However, owing to the absent forearms in this patient, the ideal method of estimating the proper cane length could not be used. Furthermore, in patients with severe type 1 phocomelia, canes may need to be manufactured on a custom basis, which was done in this case, and this could certainly be an area of further research and development.

Current controversies and future considerations

The surgical approach used in THA for patients with DDH is controversial, and in this particular case, it was exacerbated by concurrent phocomelia. THA to correct hip dysplasia has traditionally been performed through either a lateral, posterolateral, or posterior approach [40]. The posterolateral approach has yielded excellent outcomes with a component survivorship of 91.4% at 98 months [41]. However, traditional approaches are associated with muscle injury and have a postoperative dislocation rate up to 16.6% in patients with DDH [15,40]. The direct anterior is an alternative approach, but may be challenging for high-grade hip dysplasia because of multiplanar difficulties, complex lumbar-pelvis-hip pathology, morphological abnormalities, and implant difficulties requiring stems with version control [42]. Although direct anterior approach may be technically difficult, computer navigation could provide utility in anatomical mapping to accurately place prosthetic components. Owing to the lack of literature on managing DDH in patients with phocomelia, this case may provide insight into the surgical planning. However, future follow-up and additional cases of patients with this rare combination of conditions need to be performed to further determine the long-term success and longevity of THA.

Summary

Developmental hip dysplasia in patients with phocomelia is a rare condition and may lead to hyperflexion requirements during development as an adaptation for ADLs. Staged bilateral THA could be considered in these patients to minimize surgical stress and allow for adequate recovery. This case report highlighted the difficulties of a patient with phocomelia who had severe osteoarthritis secondary to Crowe 3 DDH, the importance of surgical planning, and intraoperative techniques used to optimize a successful outcome.

KEY POINTS

- Phocomelia is a rare condition that poses unique challenges to orthopedic surgeons in regard to surgical planning and postoperative rehabilitation because of absent limbs.
- Direct anterior THA is a viable option in phocomelia patients with DDH especially when there are hip hyperflexion requirements for ADLs.
- Preoperative CT and intraoperative computer navigation can be effectively used in patients with distorted anatomy to accurately place components with less extensile approaches.
- It is imperative to recognize difficult arthroplasty cases in advance as they may require great expertise and more extensive surgical planning.
- The traditional quadrants and safe zones for transacetabular screw fixation may be less applicable in patients with high dislocation or severe DDH, and therefore, it may be advisable to have smaller screws readily available than customarily provided in a set to minimize the risk of intrapelvic screw penetration.

Conflict of interests

S. W. Kreuzer received royalties from Stryker Co., Zimmer, Corin, Synvasive, and Smith & Nephew; is in the speakers' bureau of or gave paid presentations for Brainlab, Inc., Medtronic, Inc., Corin, THINK Surgical Inc., and Anterior Hip Foundation, Inc.; is a paid consultant for Corin USA Ltd., Pacira Pharmaceuticals Inc., Smith & Nephew PLC, Stryker Co., THINK Surgical Inc., Zimmer Biomet Inc., UOC USA, Inc., Anterior Hip Foundation, Inc., BodyCad, Inc., Intelijoint Surgical Inc., and HD Research Group; has stock or stock options in Innovative Orthopedic Technologies (IOT), Corin USA Ltd., INOV8 Orthopedics, INOV8 Surgical, INOV8 Healthcare, K and S Solutions; received research support from THINK Surgical Inc. and Smith & Nephew PLC; is a board member of International Society for Technology in Arthroplasty (ISTA), Memorial Bone & Joint Research Foundation (MBJRF), Corin USA Ltd., UOC USA, Inc., International Congress for Joint Reconstruction (ICJR).

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Image 1. Front view of phocomelia patient with foreshortened bilateral upper extremities. Patient dependent on adjustable 4-point cane currently in use by patient's left upper extremity for ambulation and balance.



Image 2. Side view of phocomelia patient in a flexed forward positioning of the lumbar spine and increased lordosis.