

Salvage procedure in a case with complicated hip stiffness due to late-onset Legg-Calve-Perthes disease: a case report

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Case:

We report a case of a 13-year-old boy with late-presenting Legg-Calvé-Perthes' disease with femoral head extrusion and painful femoro-acetabular impingement. The patient exhibited significant limitations in performing daily activities due to a limping gait requiring elbow crutches. The patient was treated with nonarticulated hip arthrodiastasis. At follow-up after skeletal maturity, the patient demonstrated marked clinical improvement, including a return to sports activity, and radiographs revealed a well-contained and congruent hip joint. Arthrodiastasis is a valuable treatment option for complex cases of late-onset Perthes' disease with femoral head extrusion and significant hip stiffness.

Keywords:

adolescent, arthrodiastasis, perthes, stiffness

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Introduction

In Legg-Calve-Perthes Disease (LCPD), the prognosis generally deteriorates with increasing age at disease onset [1]. Consequently, the optimal treatment approach for late-onset LCPD remains a subject of debate [1]. The presence of hip stiffness due to hinged abduction is difficult to treat [2]. The options are limited to cheilectomy, proximal femur valgus osteotomy, hip arthroscopy, or arthroplasty [2,3]. The main complication of LCPD is deformation of the femoral head which is happened due to extrusion of the femoral head [4]. If more than 20% of the width of the epiphysis extrudes outside the acetabulum, irreversible femoral head deformation is almost inevitable [5,6]. Arthrodiastasis of the hip or hip distraction utilizing an external fixator was initially elucidated by Aldegheri in 1981 as an innovative therapeutic approach for certain hip pathologies [7]. From a Biomechanical perspective, this distraction technique serves to alleviate load from the hip joint, subsequently facilitating the regenerative process within the bone and cartilage [8]. The literature has highlighted numerous applications of this method, advocating its use for the treatment of various hip disorders including LCPD, avascular necrosis (AVN) of the femoral head, and idiopathic chondrolysis [9]. Moreover, hip arthrodiastasis can either stand alone as a therapeutic intervention or be amalgamated with other modalities, such as core decompression, and proximal femur cheilectomy [8]. While an extensive body of research exists on LCPD, a unanimous consensus on the optimal treatment for its late manifestation, specifically in adolescents, remains elusive [10]. Notably, late-onset LCPD, presenting after the age of nine, is typically associated with an unfavorable prognosis. This is primarily attributed to

the limited duration available for the remodeling phase preceding skeletal maturity [10]. The overarching objective of hip arthrodiastasis is to expedite the hip's range of motion recovery, forestall or decelerate the onset of early hip osteoarthritis, alleviate pain, and enhance gait mechanics [11].

In this paper, we describe a 13-year-old male diagnosed with late-presenting LCPD. The patient underwent hip arthrodiastasis without soft tissue release. The main aim of treatment was to regain some hip motion and pain improvement [12]. Written informed consent was obtained from the parents after an explanation of the difficulty of the situation.

Case report

A 13-year-old male, an avid football player with no known prior medical conditions, presented to our clinic with complaints of persistent pain in the left hip, concomitant with noticeable limping and stiffness. Tracing back to when the patient was 11 years old, he was a football player, he recounted the pain as having an insidious onset that progressively worsened and limping gait. He vehemently denied any history of trauma, fever, or systemic symptoms, and lamented his inability to engage in regular sporting activities. Upon physical assessment, the patient exhibited an antalgic gait on the left side. Moreover, stiffness during gait was notable. His reliance on crutches for walking

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was evident. A significant limitation in the painful internal rotation of the left hip was limited to -20° , in stark contrast to the right hip's 45° . Abduction in the left hip was restricted to 10° during both flexion and extension, markedly reduced when compared with the 60° observed on the contralateral side, which is demonstrated in his antalgic limping gait (Figs 1, 2). Radiological investigations, specifically anteroposterior pelvis radiography, and frog lateral views from prior consultations (Figs 3, 4), illuminated signs of AVN of the left femoral head, characterized by prominent sclerosis and an increased medial joint space of the left hip. Initial management elsewhere adopted a conservative approach with close outpatient monitoring. However, 1 year later, the patient's clinical symptoms had exacerbated. Subsequent radiographs (Fig. 5) unveiled irregular ossification of the femoral head, evident femoral head collapse, pronounced sclerosis, and limited abduction. Despite these findings, surgical intervention was not pursued at that juncture. At the age of 13, the patient's clinical condition had deteriorated significantly. He exhibited a pronounced limp and experienced severe pain during hip movements. The radiographic findings were consistent with a Lateral Pillar Classification C, Catrall Classification 4, with a severely flattened, collapsed left femoral head and hinged extruded lateral femoral segment (as shown in Fig. 5). Given the

Figure 1



Front clinical picture for an 11-year-old boy with a flexed hip and knee left side revealed a painful limp left lower limb.

severity, surgical intervention in the form of left hip arthrodiastasis using an external fixator was proposed and executed. Postoperatively, anteroposterior pelvis

Figure 2



Clinical picture back view for 11-year-old boy flexed hip and knee left side indicated painful limp left lower limb.

Figure 3



Pelvis radiography anteroposterior and frog lateral views show signs of left femoral head avascular necrosis in the form of sclerosis and widening of left hip medial joint space (space between teardrop and ossification center).

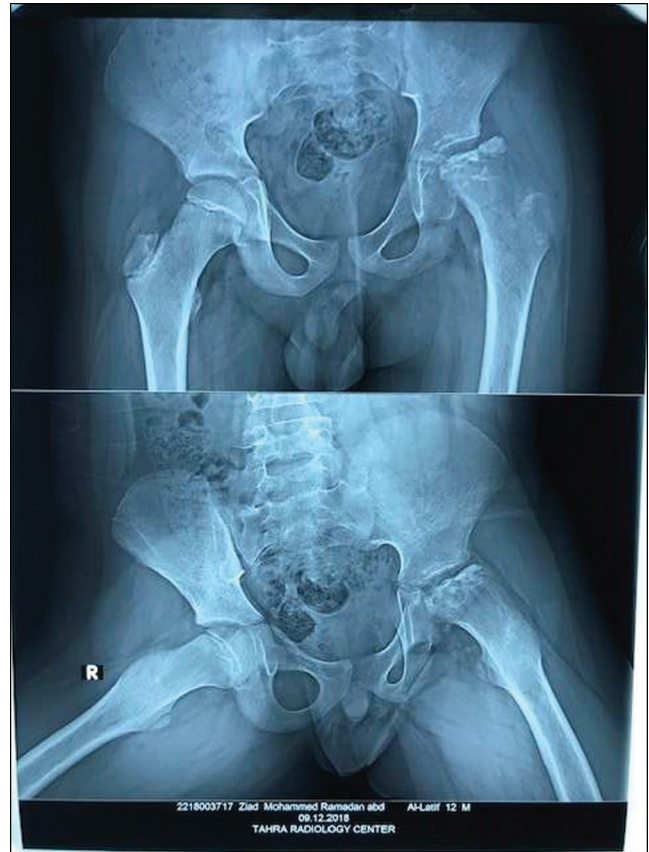
Figure 4



Pelvis radiography anteroposterior and frog lateral views show femur head ossification center irregularity, femur head collapse and sclerosis, and limited abduction.

radiography depicted nonarticulated hip distraction arthrodiastasis achieved through an external fixation device. The fixation was secured using three supra-acetabular Schanz pins and three pins in the femoral shaft (Fig. 6). Distraction was oriented using Shenton's line as a reference radiographic landmark. Subsequent computed tomography and MRI scans corroborated the presence of extensive femoral head osteonecrosis accompanied by a complete lateral pillar collapse. Following a 4-month treatment period, the external fixator was gradually removed. Initially, the rods were loosened to allow full weight bearing. After 1 week, with no radiographic changes or clinical pain, the patient underwent removal of the hip arthrodiastasis in the operating room. The procedure involved sequential removal of connectors, rods, the three supra-acetabular pins, and the remaining Schanz pins from the femoral shaft. Recovery was uneventful with no complications. At the 4-month postoperative mark, radiographs depicted initial re-ossification of the femoral head and preserved left hip joint space (Fig. 7). Remarkably, by the 15-month postsurgical follow-up, the patient exhibited significant mobility without pain, improved hip range of motion, and radiological evidence signifying enhanced hip congruency (Fig. 8). Fast-forwarding to 4.5 years postsurgery, the patient,

Figure 5



A 13-year-old boy with left hip late-onset Legg-Calve-Perth's Disease showed Lateral pillar classification C, Catrall classification 4, severe flattening left femur head with Hinged Abduction.

having attained skeletal maturity, was completely pain-free, ambulated with a normal gait without any support, and showcased a considerably improved left hip range of motion. A mild limitation in the internal rotation was noted on the left side 15°. He returned to his previous functional level as a football player. Radiographic evidence depicted a well-contained and congruent hip joint (Fig. 9). The patient resumed his former athletic activity, returning to football without any residual complaints.

Discussion

Femoral head extrusion with marked hip stiffness in severe late-onset Perthes' disease is a real challenge to the pediatric orthopedic surgeon. Hip arthrodiastasis, as underscored in various studies [12–14] is particularly efficacious during the early fragmentation stage of LCPD. However, the prolonged use of an external fixator at this juncture often culminates in device intolerance, which could lead to premature removal. Such an untimely removal, prior to femoral head reconstruction, may exacerbate the risk of recurrence of epiphyseal collapse [5,6,11,13] In light of these findings, we strongly advocate for initiating

Figure 6



Postoperative anteroposterior pelvis radiography showed 3 Hip distraction arthrodiastasis using iliazrove external fixation device. Three supra-acetabular Schanz pins and three femur shaft Schanz pins with hip distraction using Shenoton's line as a reference for the hip arthrodiastasis.

hip distraction during the impending femoral head collapse in the fragmentation stage, especially when accompanied by any of Catterall's head-at-risk signs [15]. Age of onset in LCPD holds paramount importance, dictating the natural progression, prognosis, and the extent of femoral head involvement [16,17]. An exhaustive review of literature converges on the consensus that an onset after 9 years, especially with comprehensive femur head involvement, portends a poor prognosis [16,18–20]. The patient in focus manifested with notably delayed presentation, with radiological indicators pointing to extensive femoral head AVN. Joseph *et al.* delineated three distinctive patterns of adolescent LCPD: late-onset, segmental collapse, and the more severe destructive pattern [14]. Joseph's research underscores the notion that management protocols tailored for younger patients are not seamlessly translatable to adolescent LCPD [14]. In addition, he recommended starting any other surgical treatment at the early collapse stage [14]. In our case, although hip arthrodiastasis started after the epiphyseal collapse, the clinical and radiological outcome were excellent. The final follow-up pelvis radiography showed type two according to Stulberg classification left hip [14].

Joseph added that a proximal femur valgus osteotomy in cases of lateral femoral head hinging abduction

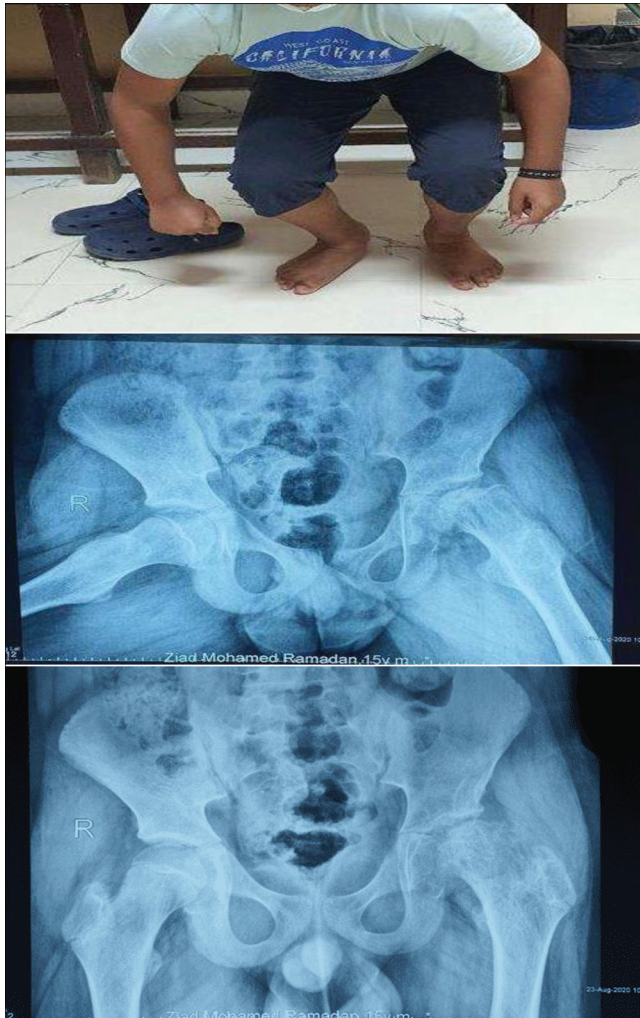
Figure 7



4 months postsurgery, anteroposterior pelvic radiography showed starting re-ossification of the femur head and maintenance of the left hip joint space.

can convert to a congruent hip and the acetabulum can be treated with a shelf procedure [4]. However, hip arthrodiastasis does not have enough literature to prove its efficacy [4]. The case detailed here wants to prove the efficacy of hip arthrodiastasis in such late presentations. The salient takeaway is that intervention when timed appropriately, can proffer better outcomes than abstaining from treatment. This notion has garnered concordance from multiple authors [14]. Our case was treated conservatively initially with a worsening scenario; in our opinion, the best time for intervention is the stage where any surgical intervention will give a better result than no treatment. This was agreed by many authors [21–23]. Moreover, this case underscores the preference for hip arthrodiastasis over other containment strategies based on empirical evidence and specific patient factors. Hosny *et al.* chronicled a series of 21 cases of late-onset Perthes' treated with nonarticulated hip arthrodiastasis. Remarkably, 20 out of the 21 cases culminated in congruent hips with satisfactory clinical outcomes at the final follow-up [24–26]. In addition, femur head subluxation and femur head collapse both are contraindicated for surgical hip containments.

Figure 8



15-month follow-up posthip arthrodiastasis showed symmetrical improvement clinically, radiography showed re-ossified femur head 5-month after external device removal.

As stated by Joseph, despite repeated emphasis by multiple authors on the importance of early containment in LCPD, it has often been disregarded. This is concerning, as the odds ratio of avoiding femoral head deformation is 16.58 times higher if containment is achieved early than if it is achieved late Stage [4,14]. Furthermore, range of motion must be restored before effective containment can be achieved. Ippolito *et al.* reviewed 11 patients diagnosed with adolescent LCPD and treated with long periods of immobilization using casts and nonweight bearing [16]. All cases had ended up with marked stiffness, painful hips, and marked muscular atrophy [16]. This also supports that; the surgery is superior to conservative treatment. In addition, Joseph advocates for the combination of weight-bearing avoidance and containment, as some reports suggest that this combination may be beneficial in the treatment of LCPD [4,14]. In the presented case, we implemented a comprehensive treatment approach. Immediately following frame application, the patient

Figure 9



Patient has reached skeletal maturity, he was pain-free and mobilizing with normal gait without assistance. Examination of the left hip showed a full range of motion with no pain nor stiffness. Radiography showed a well-contained and congruent hip joint.

was encouraged to commence partial weight-bearing. After 5 days, distraction was initiated at a rate of 1 mm/day until the joint space was 5 mm wider than the normal side. Weight-bearing gradually increased until full weight-bearing was achieved after 2 months of the operation. Following frame removal, the patient underwent hydrotherapy and partial weight-bearing using two elbow crutches for 2 months. The patient then used one elbow crutch on the opposite side for 1.5 months. This progressive approach was designed to optimize the patient's recovery and minimize the risk of complications.

Garry reported a 15-year-old boy with LCPD treated with hip arthrogram which demonstrated a loose fragment within the hip, and he recommends planning the osteotomy based on the hip arthrography in all adolescent LCPD with collapsed femur heads. We thought that hip arthrodiastasis is a valid alternative option to operate at once without intra-operative hip examination [27]. Hosny *et al.*, reported a series of 21 cases with late-onset LCPD who underwent nonarticulated hip arthrodiastasis without soft tissue release with 20 cases resulting in a congruent hip and clinically satisfactory at the final follow-up [28]. We believe that our case is unique because it is clinical and radiological follow-up until skeletal maturity, adolescent LCPD is approached with different treatment strategies and the boy returns finally to his previous functional level as a football player. As reported in the literature, the age at presentation

was classified into two groups: Presentation between 6 and 9 years and the other group more than 9 years, with obviously better clinical and radiological result after hip arthrodisastis with the younger group. This clearly described the effect of age on the clinical and radiological response [29]. Our case was in the group of older children with follow-up until skeletal maturity following hip arthrodiastasis. Roy *et al.* reported 9-cases with adolescent LCPD evaluated by hip arthroscopy and they found four cases with ligamentum teres tear, 2 cases with labral tears, and others with osteochondritis dissecans [30]. They reported that 90% of cases were pain-free and returned to their previous sports activities after addressing intraarticular pathology [30]. The arthroscopic technique was using joint distension and distraction which is going with same biomechanics of hip arthrodiastasis by external fixator.

Quevedo *et al.*, presented 15 year old with late presenting LCPD and preferred to preserve the hip using surgical hip dislocation with a mosaicplasty utilizing the extruded lateral femoral segment as an autograft, this was to address the intra-articular obstacles while doing femur neck lengthening and trochanteric advancement for the extra-articular problems in both hips [31]. He had a Harris hip score of 85% and a hip outcome score was 94% for daily activities and 86% for sports [31]. In our detailed case the Harris hip score was 93% and the hip outcome score was 100% and patient satisfaction was excellent.

The existing literature on hip arthrodiastasis for late-onset LCPD is often limited by short follow-up periods and heterogeneity of treatment indications [32]. This case report describes a patient who presented at 13 years old, underwent intervention and was subsequently followed until skeletal maturity, achieving remarkable clinical and radiographic outcomes.

Conclusion

We herein report a unique case of late-onset LCPD, characterized by complete femoral head collapse and radiographic evidence of total femoral head involvement. Given the patient's age and the advanced stage of the disease, the decision was made to perform hip distraction using external fixation. At the 5-year follow-up, which extended until skeletal maturity, the patient exhibited marked clinical improvement and radiographic evidence of a well-contained and congruent hip joint.

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Conflicts of interest

All the authors declare that they have no known competing financial interests or personal relationships that could have to influence the work.

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