Hinged versus fixed arthrodiastasis in late-onset Perthes' disease

Sameh M. Abo El-Fadl

Department of Orthopedic, Faculty of Medicine, Suez Canal University, Ismailia, Egypt

Correspondence to Sameh M. Abo El-Fadl, MD, Department of Orthopedic , Faculty of Medicine, Suez Canal University, 9 Baladia Street, Mostafa Kamel Square, El-Afrang District, Ismailia, Egypt. Tel: +20 100 510 9682; e-mail: drsamehfadl72@yahoo.com

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Background
Late-onset Perthes' disease occurs in patients older than 8 years and constitutes approximately 20% of cases and is known for its aggressive course and poor outcome with chronic hip pain and stiffness. The best treatment is still unknown, and various surgical treatment options have been proposed, but they have some inherent drawbacks. Arthrodiastasis had been introduced as a treatment method for late-onset Perthes' disease.
Patients and methods
This is a prospective study performed in Suez Canal University hospitals during the period from October 2010 to December 2014. Twenty hips were divided into two groups. Group I consisted of 10 hips treated by hinged arthrodiastasis. Group II consisted of 10 hips treated by fixed arthrodiastasis. Arthrodiastasis was done by external ring fixator. At the end of the follow-up period, patients were evaluated clinically and radiologically.
Results
At the end of the follow-up period, which ranged from 24 to 30 months, with a mean of 27.6 months for group I, and a mean of 26.2 months for group II, preoperative and postoperative clinical features and outcome scoring systems in both groups showed improvement and statistically significant relationship in values of limb length discrepancy, total hip range of motion, modified Harris hip score, and epiphyseal index. Comparing postoperative clinical features and outcome scoring systems between both groups at final follow-up showed values in group I better than group II, but there is no statistically significant relationship.
Conclusion
Arthrodiastasis either hinged or fixed using ring external fixator combined with adductor tenotomy is an excellent and reliable method for treatment of late-onset Perthes' disease, where prognosis is usually poor and conventional methods of treatment are not reliable.

Keywords:
arthrodiastasis, late onset, Perthes'

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Introduction

Legg–Calvé–Perthes disease is the eponym given to idiopathic osteonecrosis of the femoral head. It can lead to hip deformities and severe degenerative arthritis. The exact etiology of Perthes' disease remains unclear, but it is likely multifactorial and may include genetic predisposition, environmental exposures, and/or socioeconomic factors [1]. Perthes' disease is defined as being of late onset when it is first diagnosed in patients older than 8 years of age. Older age at initial presentation carries a less favorable prognosis with rapid deterioration of the joint, and early arthritic changes are common, and it is common also that hinge abduction appears in uncontained hip, situation that has a difficult solution with the standard surgical procedures [2]. This group constitutes approximately 20% of cases and is known for its aggressive course and poor outcome with chronic hip pain and stiffness [3]. It is widely accepted that those most at risk of a poor outcome are who develop the disease late [4]. The possible explanations to this fact are that acetabulum is unable to accommodate the congruity of deformed femoral head owing to decreased elasticity of acetabulum [5] and possibly these children have less time remaining for growth and remodeling of head of femur [6]. There are different modalities in the management of Perthes' disease, but the best treatment of Perthes' disease is still unknown. The main principles of treatment have traditionally been relief of loading and containment. Bed rest, traction, and bracing have been used to relieve symptoms of the disease, with a better outcome to be expected in the younger patients [7]. For older

children, the various surgical treatment options have been proposed for this late-onset disease, but they have some inherent drawbacks. These approaches neither reduce the pressure on the head of femur nor change the shape of the femoral head. No traditional treatment modality has shown any statistically significant efficacy in improving the outcome of Perthes' disease [8]. The term 'arthrodiastasis' was initially used to describe a technique involving articulated distraction of the hip joint that was developed by surgeons in Verona, Italy, and is in use since 1979 [9]. The word is a composite from the Greek: arthro (joint), dia (through), and taxis (to stretch out). The technique aims at creating a space between the articular surfaces, minimizing mechanical stresses, and maintaining movements, while the synovial circulation can be restored. This encourages fibrous repair of defects of articular cartilage and the preservation of an intact and congruent femoral head [10]. So, this method is expected to show improvement in patients with Perthes' disease, especially those with late onset, who would have rather poor outcomes by conventional methods of treatment.

Patients and methods

This is a prospective study performed in Suez Canal University hospitals during the period from October 2010 to December 2014. This study was approved by the Ethical Committee of Suez Canal University. All parents signed an informative consent form. The aim of this study is to compare the results of hinged versus fixed arthrodiastasis in the treatment of late-onset Perthes' disease. Children older than 8 years with a presentation of persistent variable degree of hip pain and/or limited range of motion (ROM) of hip joint were included in our study. Both sexes and all grades according to Herring's classification were also included in this study. Patients younger than 8 years or patients with AVN of the femoral head owing to other causes of Perthes' disease were not included in this study. Ilizarov ring external fixator was used to do arthrodiastasis for the involved hips. Twenty hips were included in our study, and they were divided into two groups. Group I included 10 hips that were treated by hinged arthrodiastasis. It includes 6 males and 4 females, with a male to female ratio of 3:2. Their ages range from 8 to 13 years, with a mean 9.9 years. Right hip was involved in 6 children, whereas left hip was involved in 4 children. According to Herring's classification, one child was A grade, 5 children were B grade, and 4 children were C grade. Group II included 10 hips that were treated by fixed arthrodiastasis. It includes 5 males and 5 females, with a male to female ratio of 1:1. Their ages range from 8 to 13 years, with a mean of 9.8 years. Right hip was involved in 3 children whereas left hip was involved in 7 children. According to Herring's classification, one child was A grade, 5 children were B grade, and 4 children were C grade. The mean follow-up period was 27.6 months for group I and 26.2 months for group II (Table 1).

Table 1 Demographic features of both groups

Features	Hinged arthrodiastasis	Fixed arthrodiastasis
Patients [*n* (%)]	10 (50)	10 (50)
Mean age (years)	9.9	9.8
Male : female	6 : 4	5 : 5
Affected side [*n* (%)]		
Right	6 (60)	3 (30)
Left	4 (40)	7 (70)
Follow-up [mean (SD)] (months)	27.6 (2.1)	26.2 (2.4)
Preoperative Herring classification [*n* (%)]		
A	1 (10)	1 (10)
B	5 (50)	5 (50)
C	4 (40)	4 (40)

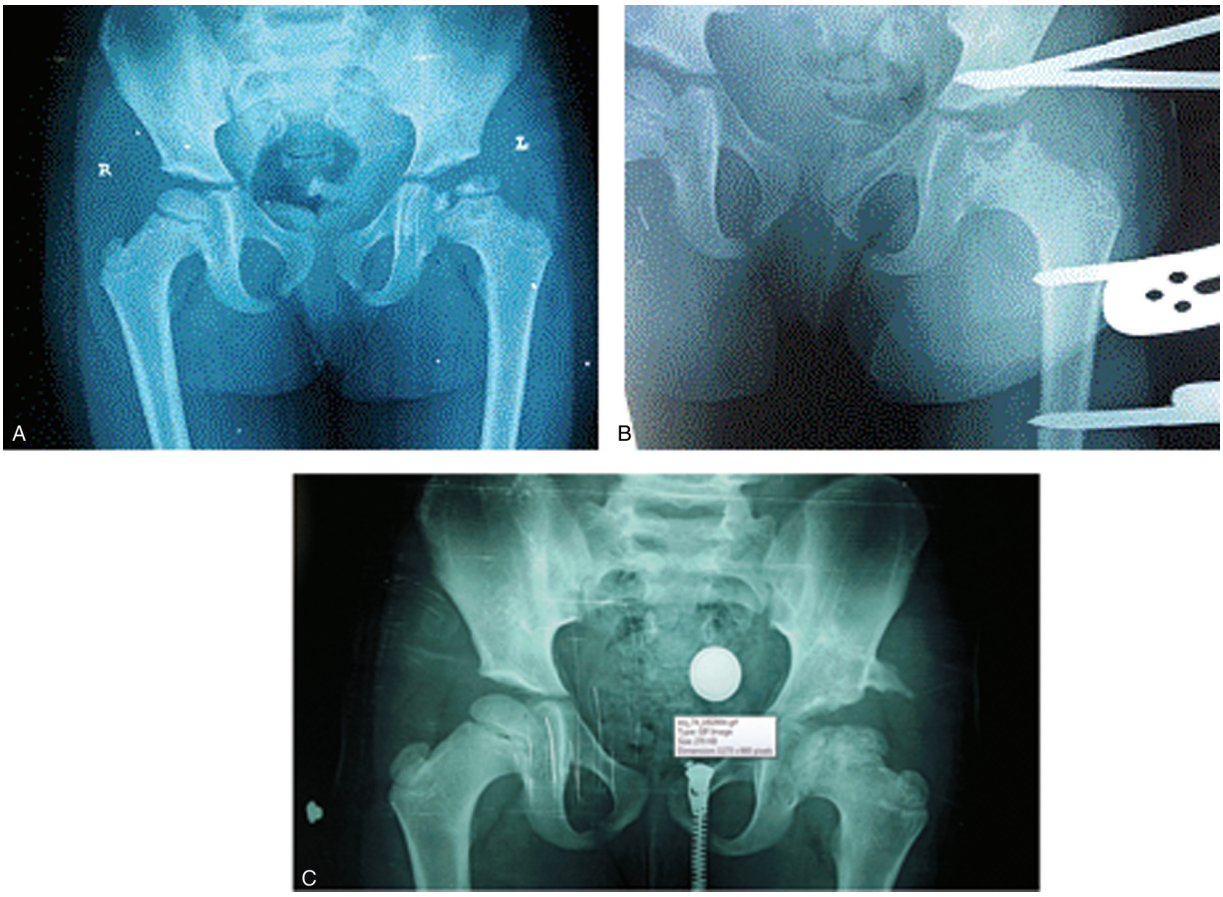
The procedure is evaluated both clinically and radiologically. The clinical evaluation includes limb length discrepancy, Modified Harris hip score, and the hip total range of motion, which was the sum of range of motions in all directions, i.e., flexion, extension, adduction, abduction, internal rotation, and external rotation. In the normal hip, it is around 270°.

The radiological evaluation depends on epiphyseal index, which was measured preoperatively, after doing arthrodiastasis for 4–5 months until lateral pillar reossification appeared, and at the end of the follow-up period.

Operative procedure

Under general anesthesia and the patient placed supine on the operating table, the involved extremity, the iliac crest, and the groin are prepared. Adductor tenotomy is to be done in all cases. A simple frame Ilizarov with two arches connected by four rods and two hinges in between that allows flexion-extension is constructed in patients of group I, who were treated with hinged arthrodiastasis. Arches connected by two rods without hinges in between were used in patients of group II, who were treated by fixed arthrodiastasis. Using the image intensifier, a perpendicular line is drawn from the shaft of the femur to the center of the femoral head. This is the line of the axis of flexion-extension of the hip. A 2-mm guide wire is inserted from the lateral side toward the center of the femoral head. It should be parallel to the floor and perpendicular to the femur

Figure 1



(a) Preoperative plain radiography of a 9-year-old female child with a left-sided Perthes' disease. (b) Postoperative plain radiography immediately after application of hinged ring external fixator of left hip. (c) Plain radiography at the end of the follow-up period showing remodeling and containment of the femoral head into the acetabulum.

while held in the 15° abduction position. The frame is positioned so that the guide wire runs through the hinges that act as center of axis of flexion and extension. Three or two supra-acetabular half pins are inserted (lateral, anterolateral and posterolateral) at multiple levels and connected to the upper arch (Fig. 1b). Two half pins are then inserted to the subtrochanteric region and another one or two pins inserted to the shaft of the femur and connected to the distal arch (Fig. 1b). In hinged arthrodiastasis, passive flexion-extension movements are done to ensure movement arising from the hip joint and the frame is modified accordingly. Acute distraction up to 5 mm is done guided by image intensifier. Sterile dressings are applied around pins.

Postoperative management

On postoperative day 2, patients are allowed to walk with partial weight bearing on crutches. With hinged arthrodiastasis, flexion-extension exercises are performed, with careful attention to preserve knee ROM in patients of both groups. Distraction will be started on postoperative day 2 at a range of 0.25 mm 4 times per day. In patients of group I, hip flexion-extension movements were encouraged, but motion is restricted to 45° for fear of damaging the newly formed cartilage. After the patients' parents are educated about pin-site care and the rehabilitation program, the patients are discharged on postoperative day 3. Distraction is to be continued until the Shenton ?s line is radiographically overreduced by ~1–2 mm, controlling the continuity of reduction. Patients are allowed to walk without any restrictions. After the hip is reduced to the overcorrected Shenton ?s line position, it will be held in that position until the date of removal. The apparatus will be left in place for 4–5 months until lateral pillar reossification appeared. Clinical visits are twice per month for the first 2 months and once per month until fixator removal. In the clinical visit, the procedure is to be followed up by examination of the fixator for any defects or infection, and a new radiography is to be done for documentation of the reshaping of the femoral head. Under anesthesia, the external fixator was removed and manipulation of the hip was done. Resumption of weight bearing will begin on a gradual basis immediately after removal, and full weight

bearing is to be achieved ~1 month after removal. Data were collected and analyzed using SPSS software (Release 20.0 for Windows; SPSS Inc., Chicago, Illinois, USA). Patients' demographics were summarized with frequencies or means±standard deviations. When comparing the outcome scores before and after management, a Student *t* test was performed. A difference was considered significant when *P* value less than or equal to 0.05.

Results

This study was performed to compare the usage of hinged or fixed arthrodiastasis as a method of treatment of late-onset Perthes' disease. At the end of the follow-up period, which ranged from 24 to 30 months with a mean of 27.6 months for group I and from 24 to 30 months with a mean of 26.2 months for group II (Table 1), preoperative and postoperative clinical features and outcome scoring systems in both groups were compared. Table 2 shows that in group I, the preoperative mean value for limb length discrepancy was 1.7 with a SD 0.92 and at the final follow-up was 0.4 with a SD 0.52, and *P* value 0.0009. In group II, the preoperative mean value for limb length discrepancy was 1.7 with a SD 1.06 and at the final follow-up was 0.55 with a SD 0.76 and *P* value 0.012. So, limb length discrepancy was found to be improved with a statistically significant relationship between preoperative and at final follow-up values in group I and group II. Regarding total hip ROM, in group I, the preoperative mean value was 197 with a SD 25.41 and at the final follow-up was 247 with a SD 20.03 and *P* value 0.0000 (Table 2). In group II, the preoperative mean value for total hip ROM was 208 with a SD 29.74 and at the final follow up was 251 with a SD 19.12 and *P* value 0.0008 (Table 2). There are statistically significant relationships in both groups as regard total hip ROM which improved in both groups. Modified Harris hip score was found in group I that the preoperative mean value was 65.14 with a SD 17.58 and at the final follow up was 89.68 with a SD 12.47 and *P* value 0.0015 (Table 2). In group II, the preoperative mean value was 63.51 with a SD 17.31, and at the final follow-up was 87.29 with a SD 12.25, and *P* value was 0.0018 (Table 2). Modified Harris hip score was found to be improved in both groups as there are statistically significant relationships between preoperative values and values at the final follow-up. Finally, epiphyseal index was measured in both groups, and it was found that in group I, the preoperative mean value was 28.07 with a SD 3.56, at removal of the external fixator was 31.4 with a SD 4.19, and at the final follow-up was 35.59 with a SD 6.07, and *P* value was 0.0048 (Table 2). In group II, the preoperative mean value was 28.91 with a SD 2.49, at removal of the external fixator was 31.46 with a SD 3.09, and at the final follow-up was 34.48 with a SD 4.87 and *P* value 0.0067 (Table 2). So, there are improvements in the preoperative, at removal of external fixator, and at the final follow-up period values of the epiphyseal index in both groups. When comparing postoperative clinical features and outcome scoring systems between both groups at final follow-up, we found that values in group I were better than group II, but there is no statistically significant relationship (Table 3). Complications were reported in both groups and include loosening of femoral pins in one patient in group I and was treated by changing pins under general anesthesia. Three patients in group I and six patients in group II developed pin-site infection, which was treated by culture and sensitivity, appropriate systemic antibiotic, and frequent dressing with good response.

Table 2 Preoperative and postoperative clinical features and outcome scoring systems in both groups

Features	Hinged arthrodiastasis	Fixed arthrodiastasis
LLD [mean (SD)]		
Preoperative	1.7 (0.92)	1.7 (1.06)
Final follow-up	0.4 (0.52)	0.55 (0.76)
P value	0.0009	0.012
Hip ROM [mean (SD)]		
Preoperative	197 (25.41)	208 (29.74)
Final follow-up	247 (20.03)	251 (19.12)
P value	0.0000	0.0008
MHHS [mean (SD)]		
Preoperative	65.14 (17.58)	63.51 (17.31)
Final follow-up	89.68 (12.47)	87.29 (12.25)
P value	0.0015	0.0018
Epiphyseal index [mean (SD)]		
Preoperative	28.07 (3.56)	28.91 (2.49)
At removal of fixator	31.4 (4.19)	31.46 (3.09)
Final follow-up	35.59 (6.07)	34.48 (4.87)
P value	0.0048	0.0067

LLD, limb length discrepancy; MHHS, modified Harris hip score; ROM, range of motion.

Table 3 Comparison of postoperative clinical features and outcome scoring systems between both groups at final follow-up

Features	Hinged arthrodiastasis	Fixed arthrodiastasis	*P* value
LLD [mean (SD)]	0.4 (0.52)	0.55 (0.76)	0.613
Hip ROM [mean (SD)]	247 (20.03)	251 (19.12)	0.652
MHHS [mean (SD)]	89.68 (12.47)	87.29 (12.25)	0.669
Epiphyseal index [mean (SD)]	35.59 (6.07)	34.48 (4.87)	0.656

LLD, limb length discrepancy; MHHS, modified Harris hip score; ROM, range of motion.

Discussion

Late-onset Perthes' disease is not a common condition, constituting approximately 20% of Perthes' cases [10]. Late-onset Perthes' disease poses a special challenge. Prognosis is usually poor because of (a) the short time available for remodeling before skeletal maturity and (b) the severity of the disease with involvement of 50% or more of the femoral head or collapse of the lateral pillar, and lateral subluxation with hinge abduction [11–13]. Treatment of Perthes' disease has always been controversial. Most orthopedic surgeons agree that not all patients need operative treatment nor on the contrary can be treated conservatively or left alone [14]. In this study, hinged as well as fixed arthrodiastasis using ring external fixator was used as a method of treatment for late-onset Perthes' disease. The rationale of arthrodiastasis using an external fixator in Perthes' disease is that it is expected to provide true non-weight bearing, giving the femoral head a chance to heal in a relatively short time and even to remodel. It can also help to achieve containment in hips that are noncontainable with the conventional methods. Another proven advantage of arthrodiastasis is the neovascularization in the distracted tissue as the space left by the distracted pins is filled by vascular granulation tissue [10]. Arthrodiastasis reduces the mechanical stress across the hip joint, which may facilitate cartilage proliferation and endochondral ossification of the proximal femoral epiphysis [15,16]. According to Herring's classification, group I consisted of one child with A grade, five children with B grade, and four children with C grade, and group II consisted of one child with A grade, five children with B grade, and four children with C grade. So, hinged arthrodiastasis and fixed arthrodiastasis were used in all grades according to Herring classification. Joseph *et al.* [17] have recommended that treatment should be started before epiphyseal collapse had occurred, as the potential to remodel in the older age group is limited. Maxwell *et al.* [10] have applied the articulated distractor to hips with minimal collapse to maintain epiphyseal height. They reported early results with preservation of epiphyseal height and arrest of epiphyseal collapse. Sudesh *et al.* [14] used arthrodiastasis in patients with Herring grade C and Catterall grade IV, where already substantial epiphyseal collapse had occurred, and they found that arthrodiastasis in such patients shows improvement in pain and increased ROM of the hip along with elimination of the hinge abduction, which allowed them to proceed with a surgical containment later on. Amer and Khanfour [3] in their work showed that children showed improvement in total hip ROM as the mean preoperative hip ROM was 198.33° (range, 130°–250°; SD, 35°). Postoperatively, the hip ROM improved to a mean of 255° (range, 200°–270°; SD, 17°), which was found to be statistically significant (t=–12.72, P=0.001). They found improvement of pain score, as the mean preoperative pain score was 6.3, which fell to a mean of 1.6 (range, 0–4; SD, 0.77) at the end of postoperative follow-up, and also was statistically significant (t=8.7, P=0.000). They concluded that minimal soft tissue release and hip distraction can be regarded as a salvage procedure for late-onset Perthes' disease with hip pain, at the stage of necrosis or fragmentation. Furthermore, this method did not result in any alteration in the joint anatomy, thus allowing the possibility for future surgery, if needed. These results were matched with our results as our patients in both groups showed improvement in limb length discrepancy, total hip ROM, and modified Harris hip score. Moreover, there was improvement in the mean epiphyseal index in both groups (Fig. 1c).

Laklouk and Hosny [18] concluded in their study that nonarticulated hip distraction without soft tissue release seems to be a valid treatment option in cases with Legg–Calvé–Perthes disease where poor results are expected from conventional treatment. In a study by Singh *et al.* [8] 12 children with age more than 8 years with Perthes' disease of less than 1 year were treated with hip distraction by a hinged monolateral external fixator. Mean duration of distraction was 13.9 days. These children were evaluated for a mean period of 32.4 months. There was a significant improvement in the range of movements, in Harris hip score, and in mean epiphyseal index, but the change in the percentage of uncovered head femur was insignificant. They concluded that hip distraction by hinged monolateral external fixator seems to be a valid treatment option in cases with Perthes' disease in the selected group of patients, where poor results are expected from conventional treatment. These results consistant with our results as there were significant improvement in limb length discrepancy, total hip ROM, and modified Harris hip score. Moreover, in our study, there was improvement in the mean of epiphyseal index in both groups, which were treated by hinged and fixed arthrodiastasis using ring external fixator. However, when comparing postoperative clinical features and outcome scoring systems between both groups at final follow-up, we found that values in group I were better than group II, but there is no statistically significant relationship, and this could be explained by the small sample size of both groups. Reported complications in this study include one patient in group I developed pin loosening and nine patients in both groups developed pin-site

infection. Amer and Khanfour [3] reported pin-track infection occurred in nearly all cases but resolved with systemic antibiotics and frequent dressing. Breakage of a Schanz screw occurred in one case during extraction of the construct under image control. A fracture of the femur distal to the construct occurred in one case after a fall. Maxwell *et al.* [10] mentioned that complications have occurred, although infrequently, and these must be balanced against the benefits that may eventually be achieved.

Conclusion

Arthrodiastasis, either hinged or fixed using external ring fixator combined with adductor tenotomy is an excellent and reliable method for treatment of late-onset Perthes' disease where prognosis is usually poor and conventional methods of treatment are not reliable.

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Conflicts of interest

There are no conflicts of interest.

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