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ORIGINAL STUDY

Arthrodiastasis for late onset Perthes' disease using a simple frame and limited soft tissue release : Early results

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Perthes' disease (Legg-Calvé-Perthes' disease) is relatively common in childhood, affecting 0.8/100.000 children. Late onset Perthes' disease, older than 8 years of age, constitutes about 20% of cases and is known for its aggressive course and poor outcome with chronic hip pain and stiffness. Although containment of the head in the acetabulum is the usual treatment for early cases, its results in treating late-onset cases are universally poor, so that many authors conclude that there is an upper age limit for effectiveness of containment treatment. The emerging philosophy of hip distraction (arthrodiastasis) with release of contractured muscle groups around the hip may be a new concept for treatment of late onset Perthes' disease.

Thirty children with late-onset Perthes' disease (age > 8 years) presenting to the orthopaedic department at Sporting Health Insurance Student Reference Hospital – Alexandria, Egypt, between December 2004 till November 2008 were treated by hip arthrodiastasis using minimal soft tissue release and a simple Ilizarov construct. At the end of an average follow-up period of 3.6 years (range : 2-7 years, SD 1.3) there was an improvement in the range of movement, pain, and superior and lateral subluxation of the head, with a statistically significant difference between pre and post-operative values.

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.

Keywords : Perthes ; late onset ; Ilizarov ; arthrodiastasis.

INTRODUCTION

Perthes' disease (Legg-Calvé-Perthes' disease) is a relatively common disease of the hip in childhood, affecting 0.8/100.000 children. Although the disease was described as early as in 1910, both its aetiology and its course essentially remain unknown and subsequently its treatment is still a matter of debate (*10,25*). Moreover, the so called "late-onset Perthes' disease" (onset after 8 years) has been known for its aggressive course and poor outcome with chronic hip pain and stiffness (*3,14,27,30*). The concept of containment of the femoral head within the acetabulum has been popularized as a principle of treatment in Perthes' disease to direct and guide remodeling of the softened femoral head as it evolves from fragmentation through ossification (*6,15*). Although this concept is widely accepted for treatment of early cases, its application for the treatment of late Perthes' disease

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is still a challenge. This consisted traditionally of either femoral or pelvic osteotomies, or a combination of both but, unfortunately, results were poor (*16,17,25,32-34,39,41*). Salvage procedures such as valgus extension osteotomy or cheilectomy were recommended by several authors but have shown no effect in preventing the early appearance of osteoarthritic changes in hips with a deformed head and hinge abduction (*4,29,31,40*). For this reason, many authors support the concept of hip distraction (arthrodiastasis) as a rationale for treatment of late-onset cases either as a first line of treatment or as a salvage solution after failure of traditional treatment options mentioned above (*1,2,5,7-9,20,21,24,35-38*). The aim of this method is to allow recovery of the femoral head by effectively relieving the mechanical force on the capital femoral epiphysis and allowing at the same time true nonweight-bearing ambulation.

The goal of this study was to evaluate the results of hip arthrodiastasis as a treatment option for cases of late-onset Perthes' disease using a simple frame and a limited soft tissue release.

PATIENTS AND METHODS

Thirty children with late onset Perthes disease (age > 8 years) presenting to the orthopaedic department at Sporting Health Insurance Student Reference Hospital – Alexandria, Egypt, between December 2004 till November 2008 were treated by hip arthrodiastasis using the Ilizarov fixator (Table I). Included were all late onset Perthes' cases older than 8 years of age, suffering from either a persistent variable degree of pain, a limited range of motion (ROM) of the hip, or presenting signs of head at risk and in either the late necrotic or fragmentation stage. Trendelenburg gait was present in all patients. The results were evaluated after a minimum follow-up period of 2 years after the termination of treatment. According to the Herring classification (*11-13,23*), 25 cases were classified as Herring Grade C while 5 were classified as Grade B. There were 20 males and 10 females. Eighteen cases were left sided and 12 right sided. Mean age of the patients at time of operation was 9.7 years (range : 8-12, SD 1.5). The mean follow-up period was 3.6 years (range : 2-7, SD 1.3).

All patients were assessed clinically for hip range of motion (ROM), gait, limb and the degree of pain using the visual analogue scale (VAS). Hip ROM 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°. Radiological evaluation of the patients was done by an anteroposterior and lateral radiograph to determine the Herring and Waldenström staging and the superior (Shenton line breakage) and lateral (distance between tear drop and femoral head) subluxation.

Operative technique

The patient was placed supine on a transparent operating table, the ipsilateral hip being elevated by a pillow. The involved extremity was draped free till above the iliac crest. A traction table was used for heavy weight children. After general anaesthesia, adductor contracture was assessed and released percutaneously if present. This was done in 25 cases. In no cases were an iliopsoas or extensive anterior hip release needed. The leg was kept in 20° of abduction with the patella pointing upwards. Under image intensifier, two or three 6 or 5 mm Schanz screws were placed in the supra-acetabular area, just penetrating the inner iliac table. To do this, the C-arm was oriented 45° externally to the vertical plane (iliac oblique view) during screw insertion to avoid deep penetration far beyond the inner table of the iliac bone with the risk of pelvic visceral injury. These screws then were connected to the proximal arch of a pre-formed Ilizarov construct made up by 2 small femoral arches and 2 or 3 connecting rods. After this, the distal arch of the construct was fixed by 3 Schanz screws to the lateral side and perpendicular to the proximal 1/3 of the femur (Fig. 1). The joint space was immediately distracted

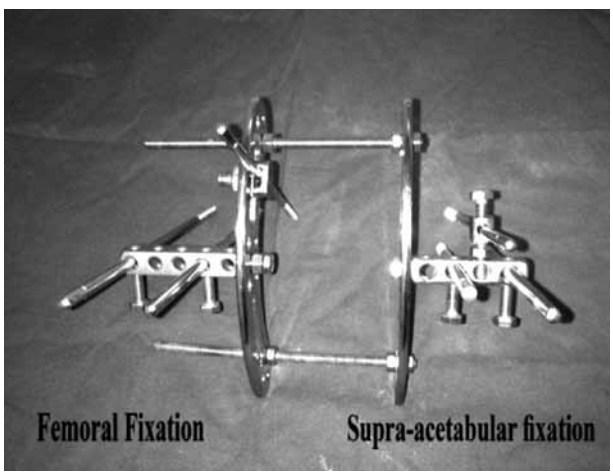

Fig. 1. — Ilizarov construct

Table I. — Data of the patients

No.	Age	Sex	Side	Follow up (years)	Pain (VAS) Pre	Pain (VAS) Post	Total ROM Pre-	Total ROM Post-	Trendelenburg Gait Resolution	Herring Class	Waldenstrom staging	Reduction in Superior subluxation (mm) Pre-	Reduction in Superior subluxation (mm) Post-	Reduction in lateral subluxation (mm) Pre-	Reduction in lateral subluxation (mm) Post-
1	11	M	L	4	9	1	240	270	+	C	F	3	1	2	0
2	8	M	L	3	8	2	220	250	+	C	F	15	2	8	4
3	8	F	L	6	8	2	230	250	+	C	F	3	0	2	0
4	12	M	L	2	9	2	140	210	-	C	F	14	3	9	3
5	9	M	R	6	6	1	150	260	-	C	F	3	0	0	0
6	10	M	R	3	4	1	250	270	+	C	N	2	0	0	0
7	12	M	L	3	8	2	130	200	-	C	N	0	0	0	0
8	8	M	R	7	9	1	240	260	-	C	N	3	0	2	0
9	8	M	R	3	10	2	160	260	+	C	F	2	0	0	0
10	9	M	L	3	8	0	220	270	+	B	F	0	0	0	0
11	11	F	L	3	7	3	170	250	+	C	F	4	2	2	0
12	8	M	L	2	6	4	240	270	+	C	F	3	0	4	1
13	11	F	R	3	6	1	200	260	-	C	F	3	1	0	0
14	9	M	L	4	7	1	170	250	-	C	F	13	2	6	2
15	11	F	L	3	8	1	220	270	+	B	F	12	0	4	1
16	8	M	R	2	10	2	210	250	-	C	F	4	0	2	0
17	10	M	R	4	3	2	230	270	-	C	F	2	1	0	0
18	9	F	L	2	2	1	170	240	-	C	F	0	0	0	0
19	11	M	L	3	3	1	220	270	+	C	N	0	0	0	0
20	8	F	L	4	1	2	240	270	-	B	N	0	0	0	0
21	12	M	L	2	4	1	190	260	-	B	N	0	0	4	2
22	9	M	R	6	1	2	180	250	-	C	F	3	2	3	0
23	9	F	R	4	3	2	170	250	+	C	F	5	3	0	0
24	8	M	R	3	7	2	160	240	+	C	F	11	3	2	0
25	8	M	R	3	9	1	220	270	+	B	F	2	1	0	0
26	11	M	L	2	10	2	190	250	+	C	F	0	0	0	0
27	10	M	L	3	10	1	240	270	+	C	F	0	0	0	0
28	9	F	L	4	4	2	220	270	-	C	N	0	0	0	0
29	12	F	R	4	6	2	160	240	-	C	F	3	0	0	0
30	12	M	L	5	3	1	170	250	-	C	F	2	0	0	0

4–5 mm under image control. Distraction was continued 1 mm per day until the Shenton line was overcorrected 2-5 mm (Fig. 2). The patient was allowed partial weight bearing the second post-operative day. The fixator was left in place for 4-5 months until lateral pillar reossification appeared (Fig. 3). During this period, there were two clinical visits a month in the first 2 months and then once a month until removal of the apparatus at the end of treatment. During these visits, pin site care, radiological evaluation and knee range of motion were evaluated. The fixator was removed in the operating room and manipulation of the hip was done under anaesthesia. Postoperatively, the patient continued a program of protective weight bearing and intensive physical and hydrotherapy for an additional 6 weeks. At this stage full weight bearing was allowed with continued physiotherapy for another 6 months.

Student's t-test (paired sample t-test & independent t-test) was used to evaluate the results. We used SPSS version 17 for Windows on IBM compatible computer

Fig. 2. — Overcorrection of Shenton line of about 4 mm of the right hip.

Fig. 3. — Lateral pillar re-ossification in the left hip. A : rapid resorption and osteoporosis of the lateral pillar denoting revascularization ; B : early re-ossification of the lateral pillar.

for analysis and statistical significance. The results were considered significant if $p \leq 0.05$.

RESULTS

At the end of follow-up, all patients showed improvement in ROM. The mean pre-operative hip ROM was 198.33° (range 130°-250°, SD 35°). Post operatively, the hip ROM improved to a mean of 255° (range 200°-270°, SD 17°), which was found to be statistically significant ($t = -12.72$, $p = 0.001$). The mean pre-operative pain score was 6.3, which fell to a mean of 1.6 (range 0-4, SD.77) at the end of post-operative follow-up,also statistically significant ($t = 8.7$ $p = 0.000$). Radiologically, there was also a significant difference between the pre- and post-operative measurements of both of the superior and lateral subluxation ($p = 0.001$). The mean pre- and post-operative superior subluxation was 3.7 mm (range 0-15 mm, SD 4.49) and 0.77 mm (range 0-3 mm, SD 1.00) respectively. The mean pre- and post-operative lateral subluxation was 1.6 mm (range 0-9 mm, SD 2.44) and 0.33 mm (range 0-3 mm, SD 0.80) respectively (Fig. 4 & 5). The Trendelenburg gait was resolved in 50% of cases. There was no significant difference related to age, sex, and follow-up duration on the recorded post-operative result for pain, range of motion or superior and lateral subluxation ($p > 0.05$). In this study, not all cases reached skeletal maturity at their final follow-up, so it is too early to assess the effect of this method on the final sphericity of the femoral head using the Stulberg classification.

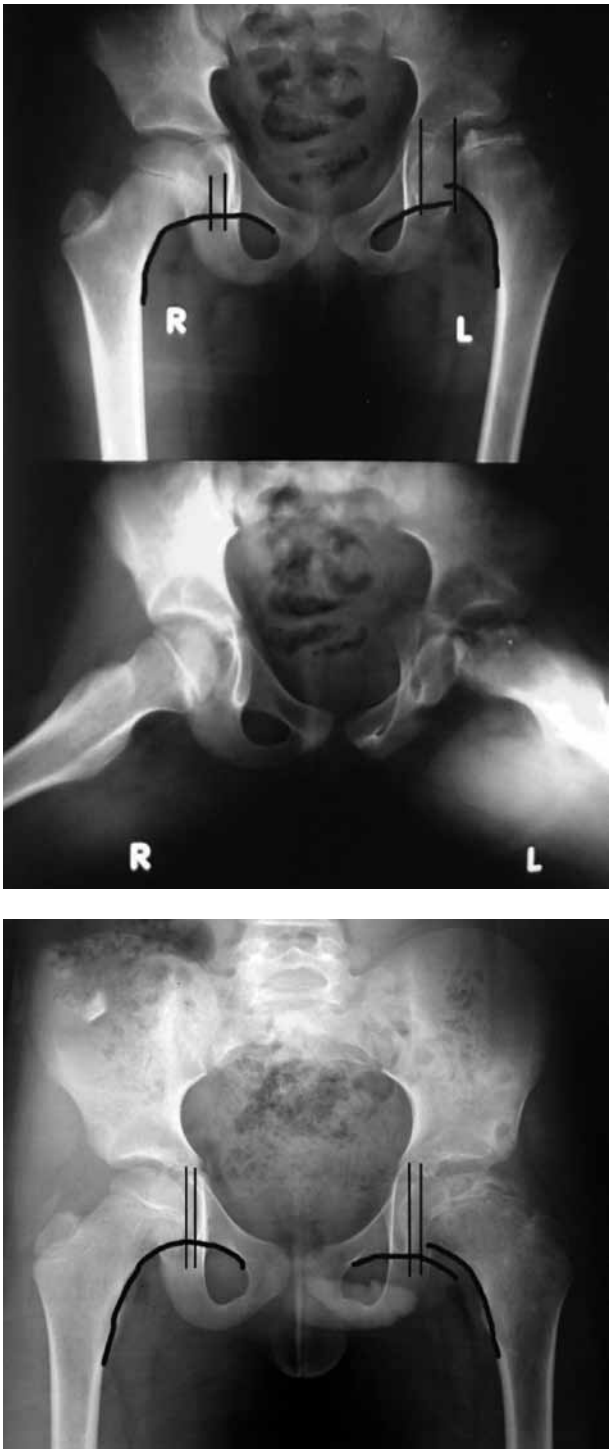

Fig. 4. — (Case 1) 8-year- old male. A : Pre-operative radiograph showing Herring class C Perthes' disease of the left hip, with 15 mm lateral subluxation and 8 mm superior subluxation. B : Follow-up radiograph after 3 years showing much improvement in lateral subluxation which reduced to 4 mm and in superior subluxation to 5 mm.

Fig. 5. — (Case 2) 11- year-old female. A : Pre-operative radiograph showing Herring class B Perthes' disease of the left hip, with 12 mm lateral subluxation and 4 mm superior subluxation. B : Follow-up radiograph after 4 years showing no superior subluxation and much improvement in the lateral subluxation to 1 mm.

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 and was

treated by extending the construct distally to fix the fracture ; the fracture united and the patient continued the treatment well.

DISCUSSION

The optimal treatment for Perthes' disease is still unknown. Late-onset Perthes' disease is not a common condition, constituting about 20% of Perthes' cases (*24*). The long-term results of late-onset Perthes' disease were found to be unfavourable for several reasons : with the disease starting at age 8 years or older, there is a short period remaining for remodeling, there are short pathological phases especially the necrotic phase that is easily overlooked, the course is more severe, and usually more than 50% of the head is involved. For these reasons, non-operative treatment, which may be appropriate in younger patients, is less successful and disappointing in the late onset age group. It has been suggested that this group has a better outcome when treated surgically (*3,19,24,27,30,35,36*). Although the philosophy of management of Perthes' disease is containment of the soft femoral head into the acetabulum as a means of preserving the sphericity of the femoral head in the younger age group, this is not effective in treating late-onset cases. The success of this approach depends on resorption of the necrotic avascular bone, replacement with new bone and subsequent remodelling while the femoral head is protected from deformation by adequate containment. The repair process appears to be grossly impaired in the late-onset cases. As a result they did not benefit from treatment by containment (*3,14*). On the other hand, the femoral head in an attempt to facilitate remodeling will overgrow, always resulting in enlargement of the epiphysis. This requires remodeling of the acetabulum to provide adequate lateral cover (*27*). Unfortunately, in late-onset Perthes' disease the corresponding acetabular remodeling both in shape and size is seldom sufficient to match the enlarged femoral head because there is not enough time for the triradiate cartilage to carry on sufficient acetabular remodeling. Thus there will often be a disparity in size between the enlarged femoral head and the acetabulum at maturity. Even if partial acetabular remodeling occurs, this will produce a sloping, unstable acetabular roof that encourages lateral subluxation (*3,14,27,35*). On the other hand, many authors postulate that cases which develop a saddle-shaped subluxating femoral head cannot be contained in the acetabulum and if this is tried, it will exacerbate pain and symptoms and invite early osteoarthritis, as this will increase the incongruity (*35,36*). For the above mentioned causes, the results of containment of the head in the acetabulum in late onset Perthes' disease using redirection osteotomies either on the femoral or pelvic side are often poor, inciting many authors to conclude that there is an upper age limit for effectiveness of containment treatment (*26,27,30,32,34,39*). No treatment has been shown convincingly to be significantly effective in improving the outcome of Perthes' disease or in influencing its course (*24*). Varus realignment of the hip may even increase the incongruency to the acetabulum and shorten the leg. Valgus osteotomy, will succeed in unloading the deformed part but it may increase the subluxation without influencing the basic avascular process. Acetabular lateral shelf, Salter, Chiari and triple osteotomies are procedures that aim to reorient or increase the size of the acetabulum and produce more support to the head. However, these approaches fail to reduce the pressure or change the shape of the femoral head (*26,28,35,36*). Arthrodiastasis utilizing Ilizarov frame can offer an alternative solution for this condition (*1,2,5,7-9,20,21,24,35-38*). Several studies have been performed to assess the role of arthrodiastasis in Perthes' disease but unfortunately, they lack universal methodological criteria regarding patient selection, evaluation of the final outcome, and presence of a control group. They evaluate the results using descriptive analytic statistics and tests of significance and consequently, direct comparisons of our results with others are not valid at this stage (*18,24*). In this study there was a significant improvement of the ROM, pain, superior and lateral subluxation at final follow-up, whereas age, sex, duration of follow-up did not have a statistically significant effect on the results. The philosophy of treatment of late-onset Perthes' cases is not containment but joint distraction. The mechanism of this kind of treatment for late onset Perthes' cases

was explained by many authors. Segev *et al* (*35*) postulated that the unloading of the cartilage inside the joint induced growth of this tissue on both sides of the joint. They showed by *in vivo* experiments in animal models, that the cartilage tissue, which is maintained under hydrostatic pressure transforms itself into bone when the compressive force is switched to distraction. The unloaded cartilage proliferates on the side of the joint space while endochondral ossification allows the epiphysis to reshape on the subchondral side. The newly transformed subchondral bone may now rebuild itself under a condition of almost total unloading (*24,27,35*). We believe that combined 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. This method, furthermore, does not affect the joint anatomy, thus allowing future surgery if needed.

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