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The Clinical and Radiographic Results of a Mean 23 Year Follow-up Period of Children with Legg-Calve-Perthes Disease Applied with Femoral Varus Derotation Osteotomy: What are the Factors Affecting the Long-term Results?

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Abstract

Objective: The aim of this study was to evaluate the long-term results of cases applied with femoral varus derotation osteotomy (FVDO) in our clinic because of Legg-Calve-Perthes disease (LCPD), and to determine the factors affecting the results.

Methods: This retrospective, single centre study included 22 patients (19 males, 3 females) who had undergone FVDO for a diagnosis of LCPD, had regular clinical and radiological follow-up, and could be contacted by telephone. The patients were evaluated with the Waldenström and Lateral Pillar classifications, the Harris Hip score, the Stulberg Hip score, and the short-form-36, and the results were compared.

Results: Evaluation was made of 22 patients, comprising 19 males and 3 females with a mean age of 31.5±6.9 years, operated on because of LCPD. The mean follow-up time was 23±6.2 years and the mean age at first presentation was 6.5±2.6 years. Lateral pillar class C was determined as an indicator of a poor outcome in patients in the Waldenström fragmentation stage. The outcomes were better in patients who were diagnosed at the age of <8 years and operated on <10 yearsin the early stages of the disease.

Conclusion: Although FVDO is an effective surgical method in LPCD cases, the chance of success is higher when diagnosis is made before 8 years old, and surgery is applied before the age of 10 years in the initial stage according to the Waldenström classification. Therefore, the importance of early and diagnosis and treatment in LCPD must be emphasised.

Keywords: Femoral varus-derotation surgery, Legg-Calve-Perthes disease, osteonecrosis

INTRODUCTION

Legg-Calve-Perthes disease (LCPD) is a juvenile hip disorder, which progresses at later stages with pain in the hip joint and findings of early arthrosis (1). Incidence of the disease is 0.5-21/100.000, and it is seen 4-5 fold more in boys than girls, and most often in the 4-8 years age group (2). Although the etiology of the disease is not fully known, reasons have been suggested such as trauma, clotting disorders, factor V Leiden mutation, hyperactivity, low socio-economic level, exposure to cigarette smoke, low birthweight, genetic predisposition, and retarded skeletal age. Patients presenting with complaints such as limping, hip, and anterior knee pain are evaluated with direct pelvis anterior-posterior radiographs. The main principle in treatment is acetabular coverage of the sensitive femoral head with impaired vascularization, to increase the blood flow by decreasing the increasing intra-articular pressure, and thereby obtain a spherical femoral head compatible with the acetabulum (3). The leading surgical methods reported to date

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are Salter osteotomy, combined osteotomy (femoral and pelvic osteotomy), chiari osteotomy, shelf arthroplasty, and femoral varus derotation osteotomy (FVDO).

Although short- and mid-term follow-up results of the surgical treatment of LCPD have often been reported in literature, there are very few studies that have reported the long-term results of surgical treatment (4-6). This study aimed to evaluate the long-term results of cases applied FVDO in our clinic because of LCPD and to determine the factors affecting the results.

METHODS

This retrospective study was approved by the Karadeniz Technical University Faculty of Medicine Institutional Review Board (IRB protocol number: 2018/149) and the requirement for informed consent was waived. We retrospective examined 26 hips of 22 patients who were applied with FVDO for a diagnosis of LCPD between 1983 and 2003, when they were aged 6-12 years. The patients included had at least 15 years of follow-up, attended final follow-up examinations, had direct radiographs available and were not missing any data in the records. Patient records were retrospectively examined. The Waldenström and Lateral Pillar classifications were applied by evaluating the preoperative clinical and demographic data together with the direct radiographic images (Table 1).

Patients were then called by telephone to undergo a follow-up examination. Those who attended were evaluated with the Harris Hip score, the Stulberg Hip score, Tonnis osteoarthritis grading, and parameters affecting activities of daily living [short-form-36 (SF-36)]. Limb length discrepancy was defined with the measurement of the distance between the spina iliaca anterior superior and the medial malleolus.

All the patients were operated on with the intertrochanteric closed wedge derotation varus osteotomy technique. Postoperatively, a pelvipedal plaster cast was applied as far as the toes, with the hip in 30° abduction and 10-15° internal rotation. The cast was removed after approximately 9 weeks according to the X-ray findings. Subsequently, weight-bearing was permitted and joint range of motion (ROM) exercises were initiated.

To compare the effect of the preoperative lateral pillar and Waldenström classifications on the clinical and radiographic results, the patients were separated into groups of age at diagnosis and age at operation. The groups were defined with reference to similar studies in the literature (5,6) as age at diagnosis of <8 years and ≥8 years, and age at operation of <10 years and ≥10 years.

Table 1. Demographic and preoperative radiographic characteristics of the patients

Variables	Data
Age (years)	31.5±6.9
Gender - Female - Male	 3 (13.6) 19 (86.4)
Age at onset of complaints	6.5±2.6 (1.0-11.0)
Age at diagnosis - <8 years - ≥8 years	 14 (63.6) 8 (36.4)
Follow-up period (years)	23±6.2 (15.0-35.0)
Mean age at operation (years)	8.2 (6.0-12.0)
Age at operation <10 years ≥10 years	 13 (59.1) 9 (40.9)
Operation side Right Left Bilateral	 8 (36.4) 10 (45.5) 4 (18.2)
Waldenström classification (hip) Initial Fragmentation	 7 (26.9) 19 (73.1)
Lateral pillar classification (hip) A B B/C C	 2 (7.7) 10 (38.5) 2 (7.7) 12 (46.1)

Statistical Analysis

In the comparison of variables not showing a normal distribution, the Mann-Whitney U test and the Kruskal-Wallis test were applied as non-parametric tests. In the evaluation of repeated measurements showing a normal distribution, the Paired t-test was applied to dependent groups. Fisher's exact test or the chi-square test were applied in the comparison of categorical data. A value of $p<0.05$ was set as statistically significant.

RESULTS

According to the Harris Hip score at the final follow-up examination, the mean score was 86.8 and none of the patients had a poor result (0-40) (evaluation range, 1-100). One hip was evaluated as fair (41-60), one as good (61-70), 5 as excellent (71-85) and 19 as excellent (86-100). In the Stulberg scoring, 15 hips were evaluated as a good outcome (11 hips grade 1, 4 hips grade 2) and 11 hips were evaluated as a poor outcome (4 hips grade 3, 4 hips grade 4, 3 hips grade 5) (Figures 1, 3).

Of the 26 hips, 12 (46.1%) were classified as lateral pillar grade C, with shortness determined in the affected extremity.

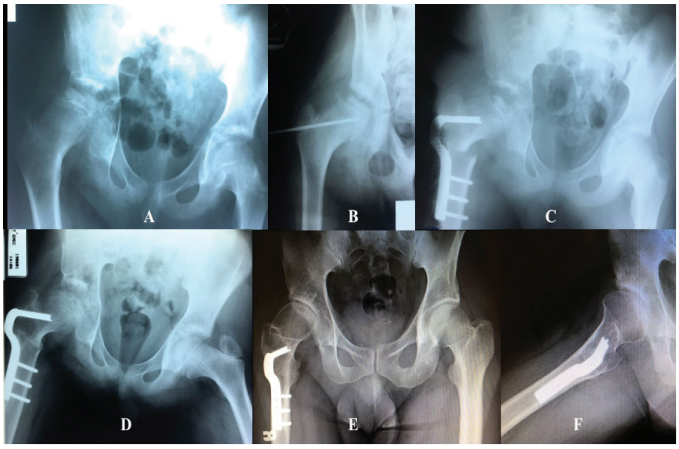

Figure 1. Preoperative image of a 10-year old patient in the fragmentation stage, lateral pillar C (A). Intraoperative radiograph of the patient at 11 years old showing the guide K-wire used for osteotomy (B). Early postoperative radiograph (C). Follow-up radiograph at 13 years old (D). Final follow-up examination radiographs (E, F) showing flattening of the femoral head and changes in the acetabulum. The result was aspherical compatible, Stulberg grade 3

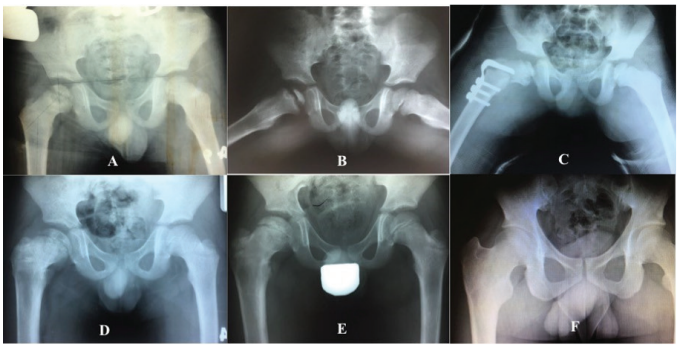

Figure 2. Radiograph of 4-year-old patient at Waldenström grade 1, lateral pillar grade B, showing the metaphyseal defect below the epiphysis (A). Frog leg position view (B). Early postoperative radiograph (C). Follow-up radiographs at age 9 years(D) and 13 years (E). Final follow-up examination radiograph (F) showing full femur head and acetabulum congruence. Spherical compatible, Stulberg grade 1

According to the Tonnis grading, no findings of degenerative osteoarthritis were determined in 12 hips, findings of mild osteoarthritis were seen in 7 hips, and grade 2 or 3 degenerative osteoarthritis was determined in 7 hips (5 hips grade 2, 2 hips grade 3). The mean SF-36 score was calculated as 78.3 (maximum: 100) (Table 2).

The patients were classified according to age at diagnosis (<8 years and ≥8 years) and age at operation (<10 years and ≥10 years), and the preoperative lateral pillar and Waldenström classifications were compared with the final follow-up clinical and radiographic results (Table 3).

The Harris Hip score in patients diagnosed <8 years was statistically significantly higher than that of the patients diagnosed at ≥8 years (p=0.007). Of the 26 hips diagnosed <8 years, 14 (77.8%) had a good Stulberg score (grade 1-2) and 1 (12.5%) of those diagnosed at ≥8 years had a good result (p=0.003). According to the Tonnis grading, significantly fewer of the patients diagnosed at <8 years had findings of degenerative osteoarthritis (p=0.004).

According to the age at operation, the Harris Hip score was significantly lower in the group operated on at ≥10 years (p=0.016). The rate of patients with a good Stulberg score (1-2) was significantly higher in the <10 year age operation group compared to the ≥ decade age at the operation group (76.5% vs. 22.2%) (p=0.014). A higher rate of degenerative osteoarthritis was determined in the ≥ decade age at the operation group compared to those operated on at <10 years (p=0.023). No difference in was determined between the two groups of age at operation with respect to the Stulberg score (p=0.057). The SF-36 score was found to be higher in the < decade age at the operation group than those operated on at ≥10 years (81.31±16.4 vs. 74.02±18.2) but the difference was not statistically significant (p=0.209).

Harris Hip scores were generally higher in patients at the initial stage according to the Waldenström classification, the Stulberg score was lower, and the rate of good results was statistically significantly higher. Of the 14 hips with lateral pillar grade A, grade B, and grade B/C, 12 (85.7%) were evaluated as excellent according to the Harris Hip score, and 7 (58.3%) of the 12 hips with grade C obtained an excellent result. The rate of excellent results according to the Harris Hip score was lower in the lateral pillar grade C cases, but not statistically significant (p=0.902). The rate of those with a good Stulberg score in the lateral pillar C group was significantly lower than that in the other groups (p=0.012).

Table 2. Evaluation of the clinical and radiographic characteristics at the final follow-up examination

Variables	Data
Tonnis osteoarthritis classification (hip)	
- 0	12 (46.1)
- 1	7 (26.9)
- 2	5 (19.2)
- 3	2 (7.7)
Harris Hip score (0-100)	
- Poor	0 (0)
- Fair	1 (3.8)
- Good	1 (3.8)
- Very good	5 (19.2)
- Excellent	19 (73.1)
Stulberg score (hip)	
- 1	11 (42.3)
- 2	4 (15.4)
- 3	4 (15.4)
- 4	4 (15.4)
- 5	3 (11.5)
Short-form-36 score (mean)	78.3±17.2

Table 3. Analysis of the clinical and radiological factors affecting the long-term results

		No of hips	Harris	p	Stulberg grade 3-5	p	Degenerative osteoarthritis	SF-36		
			Mean ± SD					p	Mean ± SD	p
Age at diagnosis	<8 y (n=14)	18	90.6±9.9	**0.002**	4	**0.003**	2	**0.014**	78.1±19.0	0.285
	≥8 y (n=8)	8	78.6±8.6		7		5		75.3±12.8	
Age at operation	<10 y (n=13)	17	90.5±10.4	**0.004**	4	**0.014**	2	**0.028**	78.9±16.8	0.376
	≥10 y (n=9)	9	80.1±8.8		7		5		74.0±18.3	
Lateral pillar grade	A	2	91.5±6.4	0.076	0	**0.017**	0	0.103	95.0±1.4	0.076
	B	10	92.0±7.2		2		1		83.3±14.4	
	B/C	2	91.5±2.1		0		0		87.1±6.9	
	C	12	81.1±12.5		9		6		67.6±16.8	
Waldenström classification	Initial	7	95.1±4.3	**0.003**	1	**0.010**	0	0.134	93.6±3.0	**<0.001**
	Fragmentation	19	83.4±11.1		11		7		71.2±16.2	

SD: Standard deviation, SF-36: Short-form-36 score

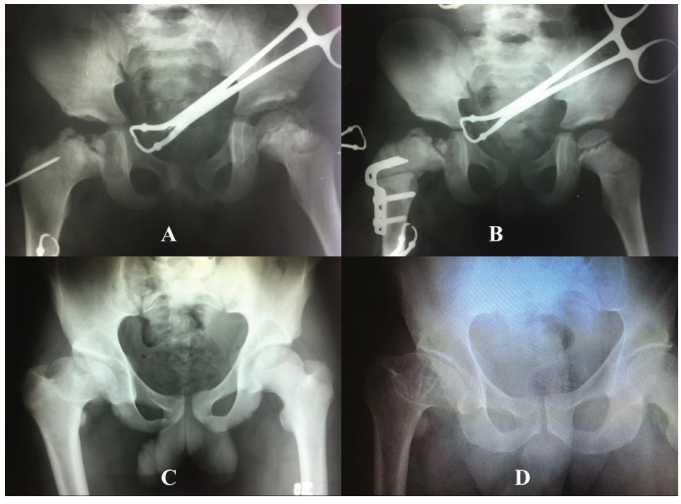

Figure 3. Preoperative radiograph of an 11-year old male patients with Waldenström fragmentation grade and lateral pillar grade C (A). Early postoperative image (B). Follow-up radiograph at age 30 years showing flattening of the femoral head and shortness in the neck (C). Final follow-up examination radiograph at age 46 years (D) showing flattening of the femoral head and shortness in the neck. Tönnis grade 1 osteoarthritis is present. Partial congruence with the acetabulum was achieved, aspherical compatible, Stulberg grade 4.

DISCUSSION

The aim of treatment for LCPD is to prevent the possibility of coxarthrosis developing by obtaining a normal or near normal hip joint. When the disease was first identified, bed rest and devices to avoid weight-bearing were used (7), and this later changed to abduction plaster cast and orthoses. However, over time studies of conservative treatment have shown that orthosis treatment is not sufficiently effective (8). As the results obtained with conservative treatment were not sufficiently satisfactory, this led to an increased tendency for surgery.

In a multicentre, prospective study, the clinical and radiographic results of the clinical treatment of LCPD were evaluated in 451 hips of 438 patients. The results of patients diagnosed at more than 8 years and with lateral pillar B or B/C were better than the surgical treatment results in the literature, but it was emphasized that those with lateral pillar C had a worse result independent of age (9). In this study, the mean follow-up period was 23 years and the mean age at surgery was 7.5 years. At the final follow-up examination, the mean patient age was 31.5 years, and 9 of the 11 patients with a poor Stulberg score had lateral pillar grade C. Of the 12 hips with lateral pillar grade B and grade B/C, 2 had a poor outcome according to the Stulberg score. Thus, it can be concluded that surgical treatment in this group was at least as successful as conservative treatment. Similarly, in patients with lateral pillar grade C, a direct relationship was determined by a poor outcome (p=0.018).

The Stulberg score of patients in the initial Waldenström grade was found to be statistically significantly lower than that of patients diagnosed in the fragmentation grade. In this context, organizing treatment in the initial stage immediately after a simple pelvis radiograph taken of a child presenting with limping, which is an early clinical sign of the disease, provide the clinician with a great advantage for treatment with better results.

The application of the principle of surgical coverage was started in 1952 by Soeur and De Racker (10) with the application of proximal FVDO. In 1962, Salter (11) brought a new dimension to the coverage principle by performing innominate osteotomy. Sponseller et al. (12) compared FVDO and innominate osteotomies, and despite no functional difference determined,

recommended that FVDO be preferred in patients with limb length discrepancy when the growth plate was closed. The coverage principle can be applied without problems in mild and moderate degree patients, but a series of problems have been encountered in severe cases (13,14). Even more shortening of the extremity in patients applied with proximal FVDO, and the Salter innominate osteotomy not providing sufficient coverage in patients affected to an advanced degree constitute problems in surgical treatment (12). In this study, 14 (63.6%) of 22 patients were lateral pillar grade C and shortness was determined in the affected extremity. As extremity shortness is related to limping, this result can be said to be the most important disadvantage attributable to the FVDO operation.

The Stulberg score results were evaluated as good (grade 1-2) in 58% of the hips evaluated in this study and poor (grade 3-5) in 42%. According to the Harris Hip scores, no patient had a poor result. The mean SF-36 value was 78.33±17.15. Despite the 11 poor results according to the Stulberg score, the reason that their wereno poor results in the Harris Hip score can be explained by the fact that a radiographic result may be reflected differently clinically, the pain threshold of the local population is high, and they have high general resistance and muscle strength.

In this study, patients diagnosed at <8 years and operated on at <10 years, better results were determined with respect of the Harris Hip score, Tonnis grade and SF-36 values. The Stulberg score of the patients diagnosed at <8 years was significantly better than that of patients diagnosed at ≥8 years ($p=0.003$), but no significant difference was determined between those operated on at <10 years or ≥10 years ($p>0.05$). It can be concluded that this result was due to close follow-up and muscle-joint exercises applied to patients with an early diagnosis, that they responded well to traction and anti-inflammatory treatment, and this directly affected the surgical outcome.

However, a significant difference was determined between the patients operated on at <10 years and ≥10 years regarding the Harris Hip score in contrast to the Stulberg score, the Harris Hip score was statistically significantly higher in those operated in <10 years ($p<0.05$). Generally, this scoring system evaluates the hip joint and the effect of the joint dynamics on walking and normal life, and is considered clinically separate from the Stulberg classification, which is a radiological evaluation criteria. The quality of life of a patient who has no pain, can walk without limping, and can easily perform daily tasks, including heavy work, will be just as high in terms of this disease. Consequently, the evaluation of radiological worsening will probably be more valuable in long-term follow-up.

Similar to the current study, Aydin et al. (6) conducted another multicentre study in Turkey and evaluated 21 hip joints applied with FVDO because of LCPD. In that study, the male-female ratio was 18:2, the mean age at surgery was 8.8 years, and the mean follow-up period was 25.1 years. Better results were determined in patients aged <10 years when operated on, and the clinical and radiological results were worse and degenerative osteoarthritis was more frequent in those with lateral pillar grade C in the preoperative evaluation. Degenerative osteoarthritis was determined in 33% of the patients.

Degenerative osteoarthritis is the most significant complication of LCPD and affects the long-term outcomes (15). Advanced degenerative osteoarthritis will cause severe hip pain, eventually resulting in total hip prosthesis for the patient, and will be a significant factor determining the quality of life of the patient in the long term. In this study, 1 patient was progressing to arthroplasty and 4 (18.2%) had hip pain that was not severe. These degenerative osteoarthritis had developed in 14 (53.9%) of the 26 hips would appears to be a worse result than that reported by Aydin et al. (6). However, of the 14 hips seen having osteoarthritis, 7 were Tonnis grade 1 (mild). Nevertheless, it is difficult to comment on the results as previously reported studies have excluded the same grade and age groups, and the skills of different surgeons can directly change operation outcomes, so this remains open to debate.

Study Limitations

A primary limitation of this study was the lack of a control group. Therefore, in the evaluation of the surgical treatment results, there can be no comparison with conservative treatment or other surgical treatments. Although the number of patients was relatively low, when the 23 year follow-up period is considered, it can be concluded that the number of patients is reasonable. This is one of the very few studies in the literature to have included the long-term follow-up results of more than 20 years.

CONCLUSION

In conclusion, better results were obtained in patients diagnosed at <8 years and when operated on at <10 years. Therefore, the importance of the early diagnosis and treatment of LCPD must be emphasized. When deciding for treatment of patients, it must be considered that in FVDO treatment, there are fewer painful hips, less osteoarthritis, and lower rates of progression to arthroplasty compared with conservative treatment, especially in lateral pillar grade C and patients aged more than 8 years at the fragmentation stage.

Ethics

Ethics Committee Approval: Karadeniz Technical University Faculty of Medicine Institutional Review Board (IRB protocol number: 2018/149).

Informed Consent: Informed consent was waived.

Peer-review: Externally and internally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: E.T., H.A., Concept: E.T., H.A., Design: E.T., H.A., Data Collection or Processing: E.T., H.A., Analysis or Interpretation: E.T., H.A., Literature Search: E.T., H.A., Writing: E.T., H.A.

Conflict of Interest: No conflict of interest was declared by the authors.

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