Clinical efficacy of early open reduction of dislocated hips using a modified Smith-Peterson approach in arthrogryposis multiplex congenita

CURRENT STATUS: ACCEPTED

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DOI: 10.21203/rs.2.22654/v1

SUBJECT AREAS
Orthopedics

KEYWORDS
Arthrogryposis, Hip dislocation, Open reduction, Modified Smith-Peterson approach, Retained rectus femoris
Abstract
Background: Arthrogryposis multiplex congenita (AMC) is a rare syndrome with multiple joint contractures. There is controversy regarding what the best surgical approach and age is for performing reduction of dislocated hips in AMC. The purpose of this retrospective study was to evaluate the clinical outcome of early open reduction on infant hip dislocation with arthrogryposis multiplex congenita through the modified Smith-Peterson approach, with retained rectus femoris intact. Methods: From 2010 to 2017, we performed this procedure on 28 dislocated hips in 20 infants under 12 months of age with AMC. The clinical and radiology data were retrospectively reviewed. The mean age at surgery was 6.9 ± 5.1 months, with a mean follow-up of 42.4 ± 41.1 months. Results: After open reduction, the average hip acetabular index (AI), the international hip dysplasia institute classification (IHDII), and hip range of motion were significantly improved (all P<0.001). After the surgery, 16 patients were community walkers and four patients were home walkers. Three hips in two patients required secondary revision surgery for residual acetabular dysplasia with combined pelvic osteotomy and femoral osteotomy. Seven of the hips that had been operated on showed signs of avascular necrosis (AVN). Among them, four were degree II, two were degree III, and one was degree IV. Multiple linear regression analysis demonstrated that the older months are risk factors for secondary revision surgery (P=0.032). Conclusions: The modified Smith-Peterson approach with retained rectus femoris intact is an encouraging and safe option for treating hip dislocation in young AMC patients (before 12 months). Even if surgery takes place at less than 12 months old for patients with AMC, earlier open reduction for hip dislocation may reduce the chance of secondary revision surgery. Level of Evidence: IV, retrospective non-randomized study. Key words: Arthrogryposis, Hip dislocation, Open reduction, Modified Smith-Peterson approach, Retained rectus femoris

Background
Arthrogryposis multiplex congenita (AMC) is a rare disorder characterized by congenital, non-progressive, and multiple joint contractures with replacement of the musculature with fibrous bands and fat tissue. AMC is not a homogenous disease, and more than 150 conditions with arthrogryposis have been recognized. [1] Stern (1923) was the first to use the term AMC; he observed and described
the condition in four of his patients, and in doing so, he characterized the syndrome.

The widely accepted classification of AMC was first proposed by Bamshad. [2] AMC with normal neurological functioning is divided into two subtypes: amyoplasia and distal arthrogryposis.

Amyoplasia is the most common of these subtypes and occurs in 38–43% of AMC. [3] The typical appearance of amyoplasia-type AMC is as follows: dislocation of the hip, extension contractures of the knee, severe equinovarus contractures of the feet, adducted and internally rotated shoulder, and extension contracture of elbow. [4]

Classically, hips in AMC are abducted, flexed, and externally rotated. [5] Hip deformity in AMC is defined as a teratological hip dislocation (TDH) condition, which is not reducible by gentle manipulation at birth as it occurs at an early stage of intrauterine life. Hip dislocation is present in approximately 30% of patients with amyoplasia-type AMC, and one half of these will be bilateral. [5] The acetabulum is shallow and small, and the femoral head is hypoplastic and often flattened in its medial part. The anteversion angle may vary greatly and can even be retroverted. [6] These pathological features lead to difficulty in dealing with TDH in AMC.

There is significant inconsistency in prior literature on the surgical approach and age for performing reduction of dislocated hips in AMC. [7] The results of closed reduction of the hip have generally been considered poor, and they often result in increased stiffness and redislocation. On the other hand, open reduction for TDH in AMC is also controversial, especially in cases of bilateral dislocation and increased surgery age. Gruel et al. recommended that bilateral hip dislocations in AMC should not be reduced because of high rates of resulting redislocation, subluxation, and avascular necrosis, [8] whereas Bernstein et al. argued that open reduction should be performed with the AMC patient between six months and one year of age. [9] Bahattin et al. held yet another perspective, as they argued that short-term results of early (before 6 months) open reduction of hips were not promising, given that about half of the AMC children who received early surgery required additional hip surgeries. [7] Wada reported that femoral varus derotation osteotomy and pelvic osteotomy were combined synchronously in most open reductions for TDH in AMC. [10] In addition, the open reduction approach usually depends on surgeon preference. Although most surgeons select the anterolateral
approach (Smith-Petersen approach) for reduction, better clinical results have been reported by the medial approach (Ludloff’s medial approach). [9] Most research regarding the open reduction through the Smith-Petersen approach for TDH in AMC patients has been for children over the age of 12 months. [10-12]

This study aims to review the results of surgical treatment to reduce hip dislocation through the Smith-Peterson approach, where the rectus femoris is kept intact in AMC patients under 12 months old.

Methods
This retrospective study was approved by the Medical Ethical Committee of Shanghai Jiao Tong University School of Medicine and the Affiliated Shanghai Children’s Medical Center. Written informed consent was obtained from the legal guardian of each participant in the study. AMC was diagnosed in accordance with Fisher’s clinical criteria. [13] Hip dislocations were diagnosed by pelvis X-ray and physical examination. The patients included in this study were AMC patients with TDH who were treated at the same hospital by senior pediatric orthopedic surgeons between 2010 and 2017. Inclusion criteria included being less than 12 months old at the time of hip reduction and having at least 24 months of follow-up after the open reduction. None of the patients included had undergone previous treatment. Physical and roentgenographic examinations were performed in outpatient follow-up.

Physical examination
As previously described, the degree of hip functioning was not the only criterion for classifying the results. All the patients were also classified as community walkers, home walkers, non-functional walkers, or non-walkers in the final post-operative follow-up. [6, 14] The preoperative and postoperative clinical evaluations measured the degree of joint mobility. The hip range of motion in flexion and abduction was also assessed at the last follow-up. [6]

Radiological assessment
The IHDI classification, continuity of Shenton’s line, AI, ossific nucleus center edge angle (ONCEA), and signs of AVN were evaluated in the final follow-up. [15-17] AVN was also classified in accordance
with Kalamchi-MacEwen classification. [18] Hips in which the Shenton arc was continuous and the CE angle was $15^\circ$ or greater were considered to be centered. For patients who had bilateral TDH, both hips were reduced in the same surgical treatment.

**Surgical treatment**

All the hips were operated on using a modified Smith-Petersen approach, but the rectus femoris was retained intact through an incision below the iliac crest. [19, 20] This was done by making an anterior bikini incision from the middle of the iliac crest to a point midway between the anterior superior iliac spine and the midline of the pelvis. The iliac apophysis was split and the iliac wing exposed subperiosteally. The straight heads of the rectus femoris were not released and iliopsoas muscle were released, but the tendon attachment was retained (Fig.1a). A T-shaped incision was made from the most medial aspect of the hip capsule to the most lateral, and the incision was extended along the anterior border of the femoral head. The soft tissue, including the ligamentum teres in the acetabulum cavity, was removed. The transverse ligament was also divided. The femoral head was gently reduced without tension and then the hip was moved through a complete range of motion to determine the ‘safe zone’. Open reductions were followed by the application of a hip spica cast in the human position with a gentle posterior mold at the greater trochanteric region of the femur. Adductor tenotomy was done in all hips to reduce tension and to achieve better congruity of the hip. [10] The patients were immobilized after surgery using a hip spica cast in the human position for a period of 12 weeks, after which they wore a brace for six months.

**Statistical analyses**

Numerical variables are shown as mean ± standard deviation. The results were analyzed using an independent-sample t-test and paired t-test. Multiple linear regression models were performed to evaluate the relationships between secondary revision surgery and related factors. The association between the revision osteotomy surgery and related factors including sex, surgery time (months), preoperative AI, and preoperative IHDI classification was evaluated using logistic regression models to determine the odds ratio (OR). Multivariate-statistical significance was assumed at $P<0.05$, $P<0.01$, and $P<0.001$. All analyses were performed using a statistical software package (SPSS for Windows, v.
Results

**Baseline data**

Among the 20 patients who participated in this study, seven were male and 13 were female. Eight patients presented with bilateral TDH and 12 had unilateral dislocation (Table 1). The children’s mean age at the time of surgery was $6.9 \pm 5.1$ months (range: four to 12 months). The mean duration of the follow-up was $42.4 \pm 41.1$ months (range: 25 to 99 months).

| Numbers (patients/hips) | All   | Revision | Non-revision | P value* |
|-------------------------|-------|----------|--------------|----------|
|                         | 20/28 | 2/3      | 18/25        |          |
| Side (left/right)       | 14/14 | 1/2      | 13/12        | 0.324    |
| Sex (male/female)       | 7/13  | 0/2      | 7/11         | 0.032    |
| Open reduction age (months) | 6.9 ± 5.1 | 10 ± 1.7 | 6.5 ± 2.4 | 0.07    |
| Preoperative AI         | 39.9 ± 4.4 | 38.8 ± 5.4 | 40.1 ± 4.4 | 0.913   |
| Preoperative IHDI (II/III/IV) | 1/19/8 | 0/2/1     | 1/17/7      |          |

*represents statistical differences (P < 0.05). OR= Odds ratio

**Ambulatory and hip functional status**

In relation to ambulatory function, 16 patients were community walkers and four were home walkers. Thirteen patients were able to walk without aids and seven were able to walk with braces. Before open reduction, the mean hip range of motion in flexion and extension in the postoperative examination was $78.2 \pm 15.2^\circ$ (range: 50° to 105°) and the mean range of motion in abduction was $23.0 \pm 8.6^\circ$ (range: 10° to 35°). The mean hip range of motion in flexion and extension in the postoperative examination was $102.0 \pm 16.2^\circ$ (range: 65° to 125°), and the mean range of motion in abduction was $37.1 \pm 9.7^\circ$ (range: 15° to 50°). Hip flexion/extension and abduction range were significantly improved after surgery, and these differences were statistically significant (P<0.001 for both factors).
Radiologic evaluation

Regarding radiographic evaluations, before open reduction one hip was IHDI II degree, 19 hips were IHDI III degree, and eight hips were IHDI IV degree. The mean AI value was $39.9 \pm 4.4^\circ$ (range: $30.9^\circ$ to $48.3^\circ$) in the preoperative examination.

After open reduction, 25 hips were classified IHDI I degree and three hips were IHDI II degree. The mean AI was $22.5 \pm 5.1^\circ$ (range: $13.6^\circ$ to $31.9^\circ$) in 28 hips. The mean IHDI classification and AI degree were significantly reduced after surgery, and there were statistical differences ($P<0.001$ in both cases). The mean ONCEA was $17.1^\circ$ (range: $9.4^\circ$ to $30.4^\circ$) in 27 hips of children with appearance of ossification center of femoral head. The Shenton’s line was found to be intact for all patients in the final follow-up. Seven hips showed signs of avascular necrosis. Among them, four were degree II, two were degree III, and one was degree IV.

Complications and additional procedures

There was no redislocation but one subluxation among the 28 hips. Three hips in two children needed a pelvic and femoral osteotomy with varus and rotation for residual acetabular dysplasia around the age of 3.4 years. There were two Salter and one Dega osteotomies in pelvic procedures. Multiple linear regression models were performed to evaluate the relationships between revision osteotomy surgery and related factors including sex, surgery time, preoperative AI, and preoperative IHDI classification (Table 1). There was a significant difference in the age of surgical time between the secondary revision surgery patients and the non-revision surgery patients ($P = 0.032$). The logistic regression model analysis showed that the OR value of open reduction time for the revision groups as non-revision groups was 4.588 (Table 1). The results show that the age of hip open reduction is significantly associated with secondary revision surgery.

Apart from one subluxation, there were no serious complications (e.g., anesthesia accident, compartment syndrome, hip infection, osteomyelitis). However, there were four superficial wound infections. Additional procedures outside the hip joint in 14 patients are summarized in Table 2.
**Table 2 Additional procedures outside the hip joint in 14 patients**

| Operative site | Numbers/Surgical procedure |
|----------------|----------------------------|
| Knee           | 1/Correction of congenital dislocation of patella |
|                | 2/Reconstruction of anterior cruciate ligament |
|                | 2/Quadriceplasty |
|                | 2/Flap surgery and external fixation for knee flexion |
| Shank          | 2/Reconstruction of dysplasia of fibula |
| Foot           | 4/Astragalectomy |
|                | 6/Vertical talus reduction and Kirschner wire fixation |
|                | 12/Achilles tendon lengthening |
|                | 2/Release through internal posterior approach |
| Elbow          | 2/Posterior elbow release |
| Wrist          | 2/Wrist extension osteotomy |

**Case Presentation**

Case 1. A three-month-old boy with AMC was brought to our clinic department. Bilateral IHDI III degree hip dislocation was detected by pelvic X-ray (Fig.2a). He had undergone a bilateral Smith-Peterson approach open reduction surgery when he was five months old and followed up for 55 months after open reduction (Fig. 2f). He had not received secondary revision operation, such as pelvic and femoral osteotomy. He could walk without a brace during the last follow-up visit.

Case 2. A four-month-old boy with AMC with vertical talus presented at our clinic for treatment. Bilateral hip dislocation with IHDI III degree was detected by pelvic X-ray (Fig.3a). He had undergone a bilateral open reduction surgery through the Smith-Peterson approach when he was six months old and followed up for 99 months after the open reduction (Fig. 3f). He had not received hip revision operation, but he had two surgical operations for vertical talus. He could walk with crutches during his last visit.

Case 3. This case presented a nine-month-old AMC girl. Bilateral hip abduction was limited to 20° and bilateral hip dislocation with IHDI III degree was detected by pelvic X-ray (Fig.4a). She had undergone bilateral open reduction surgery through the Smith-Peterson approach at nine months of age, and she followed up for 93 months after open reduction. She had received a bilateral hip revision operation at
33 months and 42 months old (Fig.4e, g). She could walk without crutches during her last visit.

Discussion
There is little prior research on the treatment for hip dislocation through the Smith-Peterson approach in AMC under the age of 12 months. Our study addresses this research gap, as it presents the experience in treating TDH in AMC using the aforementioned approach for children under 12 months old.

The teratologic dislocations in patients with AMC are much more rigid and irreducible than developmental dysplasia of the hip. Therefore, hip closed reduction generally results in increased stiffness and a high rate of subluxation and redislocation. Consequently, hip closed reduction is normally considered invalid. [21] Open reduction for teratologic dislocations was introduced for patients with AMC as a valid option and this approach is considered to prevent pelvic obliquity, sitting imbalance, gait abnormality, and secondary scoliosis. [10]

Surgical time
Concerning surgical time, Bahattin et al. suggest that early reduction (before six months) in patients with AMC does not lead to less hip surgery. [7] Several authors have suggested that operative treatment of hip dislocations in AMC can be performed at three to ten months. [3, 22, 23] The hip joint development is the result of an intricate balance between growing bony changes of acetabulum with the proximal femur. [24] Considering the severe deformity of hip dislocation in AMC and the corresponding need for earlier and longer acetabulum molding, we agree with their treatment time window which can be performed at three to ten months. In fact, the results of this study also confirm this viewpoint. The two patients (three hips) with secondary revision surgery were more than eight months old when they received open reduction; however, fifteen out of the total 18 participants did not require secondary revision surgery at less than eight months old. Recall that there was a significant difference in the age of surgery between the secondary revision surgery patients and the non-revision surgery patients. However, sometimes we could not receive the patient for treatment as early as we intended to because some children were abandoned by their families and sent to welfare institutions. In the literature, a relatively high proportion of the reported AMC case series required
additional femoral or pelvic osteotomies along with open reduction. This result suggests that the effects of reduction age on acetabular shaping in TDH and developmental hip dysplasia are similar; in other words, reduction surgery at an older age is more likely to lead to residual acetabular deformities. [25]

**Surgical approach with muscle protection**

We used a modified anterior Smith-Peterson approach with rectus femoris intact for all the TDH in this research. There are several advantages to using this method to treat patients with AMC. Firstly, more attention should be paid to concentric reduction and reduction stability in hip dislocation in AMC, and the method used here addresses this issue. The acetabulum in AMC is small, shallow, and filled with fibrous-fatty tissue. The femoral head in AMC is hypoplastic and often flattened in its medial portion, which can be demonstrated by arthrography (Fig. 1b and 1c). [6] Compared with medial-approach open reduction, the anterior Smith-Peterson approach is demonstrably more effective in hip joint exposure, hip obstacle removal, and circumferential capsulotomy. The reports in the literature show that there are more frequent secondary procedures for progressive subluxation after open reduction when the anteromedial access route is used. [3, 8] Additionally, the medial approach is likely related to injury of the medial circumflex artery, which causes iatrogenic AVN especially in infants younger than 12 months with unclear hierarchical anatomy. The reported rate of significant AVN in medial-approach open reduction is as high as 43%. [26, 27] For the present study, we have kept rectus femoris and the tendon attachment of iliopsoas intact in this modified anterior Smith-Peterson approach, which minimizes muscle damage near the hip joint.

Avascular necrosis of the femoral head in open reduction through the Smith-Peterson approach in TDH is a risk when doing a complete capsulotomy of the hip. However, Akazawa et al. thought that hip capsulotomy adjacent to the acetabular rim does not affect the blood supply to the femoral head if the incision has a proper distance from the base of the femoral neck. The lateral epiphyseal artery comes from the femur greater trochanter and passes through the posterior capsule at the femoral neck base. [10, 12] In our series, seven hips demonstrate AVN, but only one hip showed Kalamchi and MacEwen grade IV AVN.
Open reduction was usually associated with increased stiffness of the hip joint in AMC. [21] In terms of the open reduction approach, Staheli et al. reported that the range of motion of AMC patients who received the medial approach was better than that in those treated using an anterolateral approach. [23] We consider that the limitation of hip joint motion is related to more soft tissue injury than that which occurs with the anterolateral approach. Therefore, we have retained rectus femoris and the attachment of iliopsoas in our Smith-Peterson approach, which also maximizes the postoperative hip joint function. In our series, most of the children retained a certain degree of hip joint activity, and none of them encountered joint stiffness after the operation.

Bahattin et al. suggested that open reduction for TDH at a late age may be preferable because open reduction and femoral osteotomy procedures can be performed simultaneously to reduce the need for additional surgeries. [7] However, when the femora-acetabular harmony is created early through the open reduction of the femoral head, the remodeling capacity of the femoral head and the acetabulum could be maximized and the need for additional surgeries may be reduced. In our study, 25 hips were IHDI I degree and three hips were IHDI II degree, without IHDI III or IV classification postoperatively. Only three hips received secondary revision surgery including femoral and pelvic osteotomy in the latest follow-up. These results indicate favorable results for femora-acetabular harmony after early open reduction through a modified Smith-Peterson approach with retained rectus femoris intact.

**Bilateral and unilateral side**

The reduction of bilateral teratologic hip dislocations in AMC remains controversial. Many authors have argued that bilateral TDH should be left untreated because the pelvis remains level and motion is satisfactory; leaving it untreated also circumvents the high rate of complications after surgery. [1] Some authors suggest that bilateral TDH should be reduced to restore femora-acetabular harmony and decrease the risk of later pain or stiffness. [3, 12] In our study group, eight patients with bilateral TDH received open reduction surgery simultaneously and seven of these patients did not receive secondary revision surgery. There was no statistically significant difference in surgical age and revision surgery between the bilateral and unilateral hip groups (P=0.188 and P=0.736). These results may suggest that simultaneous open reduction of bilateral hip joints does not affect the
clinical results.

TDH in AMC is accompanied by multiple musculoskeletal disorders, such as contractures and other joint dislocations. Management of arthrogryposis is difficult because numerous surgical procedures are necessary for concomitant knee, shank, foot, elbow, and wrist deformities. [10] We dealt with lower limb deformities in the following order: foot, hip joint, and knee joint.

Our study had several limitations. Firstly, because it was a retrospective study, there was selective bias and no standardized indication for secondary revision surgery. Secondly, the AMC sample size was too small. Having a larger number of patients in future research may yield more definitive results concerning the best time for early open reduction surgery. Thirdly, we only evaluated medium-term clinical outcomes. Longer follow-up may lead to increased incidence of secondary revision surgery and complications and may provide additional clinical information.

Conclusion
Our study indicates that early open reduction of dislocated hips through a modified Smith-Peterson approach with retained rectus femoris intact in AMC could restore femora-acetabular harmony and provide sufficient clinical and radiological results. Earlier open reduction for hip dislocation may reduce the chance of secondary revision surgery.

Abbreviations
AMC: Arthrogryposis multiplex congenita; Al: Acetabular index; AVN: Avascular necrosis; TDH: Teratological hip dislocation; IHDI: The international hip dysplasia institute classification; OR: Odds ratio

Declarations
Acknowledgements
Not applicable

Funding
Approval of financial support was given by the Medium and Long-Term Clinical Research Foundation of Shanghai Children’s Medical Center (Grant No. ZCQ-SCMC2018-1), the Clinical Research Cultivation Foundation of the Shanghai Shenkang Hospital Development Center (Grant No. SHDC12018X31), the
Natural Science Foundation of Shanghai (Grant No. 17ZR1417900) and the National Natural Science Foundation of China (Grant No. 81801919). This research received no specific grant from any funding agency in the commercial sectors.

Availability of data and materials
The datasets generated and/or analyzed during the current study are not publicly available because they contain patients’ personal information, but are available from the corresponding author on reasonable request.

Authors’ contributions
MM: Data collection, major contributor to writing of manuscript. HC (Haiqing Cai) and ZW: Data collection and interpretation, assistance with writing manuscript. LH and JB: Data analysis and interpretation. HC (Haoqi Cai): Study concept and design, assistance with writing manuscript. All authors read and approved the final manuscript.

Ethics approval and consent to participate
The clinical study proposal was approved by the Ethical Committee of Shanghai Jiao Tong University School of Medicine Affiliated Shanghai Children’s Medical Center, and written consent was obtained from all patients.

Consent for publication
Written consent to publish images of patients was obtained from study participants and was approved by the ethics committee.

Competing interests
The authors declare that they have no competing interests.
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Figures
(a) Photos during operation indicated the straight heads of rectus femoris were not released (the white solid arrow) and exposed the femoral head after opening the hip joint capsule through T-shaped incision. (b,c) Preoperative arthrography shows the hip inverted limbus with widened acetabulum medial pooling in AMC. Tentative closed reduction shows that the dislocated hips were stiff and irreducible.
Case 1. (a) Preoperative pelvic anteroposterior view in case 1 o five months old. (b) Immediate MRI after surgery confirmed satisfactory hip reduction. (c and d) Radiograph at 16 months and 26 months after open reduction, respectively. (e) MRI assessment at 55 months after open reduction. (f) Radiograph at 55 months after open reduction.
Figure 3

(a) Preoperative pelvic anteroposterior view in case 2 on six months with lower limbs orthopedic gypsum for foot deformity correction. (b) Immediate CT scan after surgery confirmed satisfactory hip reduction. (c and d) Radiograph on 14 months and 26 months after open reduction, respectively. (e) MRI assessment at 28 months after open reduction. (f) Radiograph at 83 months after open reduction.
(a) Preoperative pelvic anteroposterior view in case 3 on a nine-month-old. (b) Immediate MRI scan after surgery confirmed satisfactory hip reduction. (c) Radiograph at 12 months after open reduction. (d) Radiograph at 24 months after open reduction indicates the right hip subluxation. (e) Radiograph of right Dega pelvic and femoral osteotomy on a 33-month-old. (f) Radiograph at 33 months after open reduction that indicates left hip residual deformity. (g) Radiograph of left Salter pelvic and femoral osteotomy on a 42-month-old. (h and i) Radiograph at 39 and 88 months after open reduction, respectively.