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症 例

Transtrochanteric Curved Varus Femoral Osteotomy for Spondyloepiphyseal Dysplasia Tarda. A Case Report

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Abstract A female with spondyloepiphyseal dysplasia (SED) tarda suffered bilateral hip pain since about the age of ten. Her radiographs revealed subluxation and irregularity of the hip joints with acetabular dysplasia. Transtrochanteric varus osteotomy was performed on her left femur at 13 years of age and on her right femur at 14 years of age. The congruities of both hips improved after surgery. At follow-up, she had no pain in either hip even after walking for an extended period. Improved activity in daily life was noted.

Key words : Spondyloepiphyseal dysplasia tarda, Subluxation of the hip joints, Transtrochanteric varus osteotomy of the femur.

Introduction

Spondyloepiphyseal dysplasia (SED) tarda characteristically causes osteoarthritis at a very early age (5). Degenerative changes occurring at an early age are difficult to treat. On a case diagnosed with SED tarda, we performed transtrochanteric varus osteotomy of the femur (4) to improve congruency of the hip.

Case report

A female with short stature suffered bilateral hip pain since about the age of ten. She had no past history. Her parents were in a consanguineous marriage.

She was born on March 26, 1978. At 1 year old, she began to walk independently. At 3 years old, her short stature was noted. At 10 years old she complained of pain in her left hip, and consulted one of the authors at 11 years old. She suffered pain while walking and also

limped. Range of motion of the hip joints were slightly limited and she felt terminal pain with flexion of the hip. She had dwarfism with a short trunk (height: 114cm, weight: 28kg). Her intelligence was normal and she did not have visual or audile disturbances. From these findings we diagnosed her as SED tarda.

Her radiographs revealed subluxation of the hip joints with acetabular dysplasia (Fig. 1) and

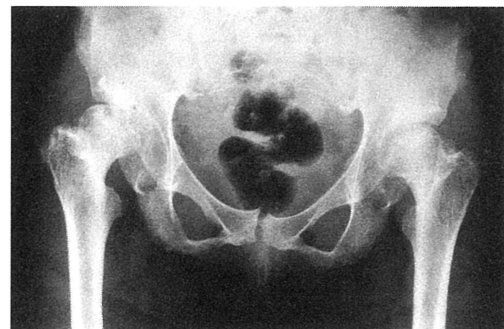


Fig. 1 Pre-operative radiograph at 13 years of age.

platyspondyly of the spine. They showed coxa plana, mild coxa valga, shortenig of the femoral neck, irregularity of the Shenton's line and decreased center-edge angle (4 degrees on the right and 15 degrees on the left) which indicate subluxation of the hip joints, sclerosis of the acetabulum and a cyst on the right femoral head which are findings consistent with osteoarthritis of the hip. Arthrograms showed that the congruence of the hips improved when abducted.

Transtrochanteric curved varus osteotomy was performed on her left femur on December 5, 1991 when she was 13 years old. The same osteotomy was performed on her right femur because of pain in her right hip on March 4, 1993 when she was 14 years old. The congruities of both hips improved after surgery (Fig. 2). The irregularity of the Shenton's line and narrowing of the joint space improved. Skin traction was performed postoperatively for 6 weeks. Partial weight bearing started at 8 weeks and full weight bearing started at 10 weeks.

Final follow-up was made at 6 years and 6 months on her left hip, and 5 years and 3 months on her right hip on June 6, 1998 when she was 20 years old. She did not limp and was using a cane only outside her house. She had no pain in either hip even after walking for an extended period. Improved activity in daily life was

noted. Flexion of both hips decreased from 130 degrees preoperatively to 110 degrees at follow-up, but this did not restrict her activities in daily life. Postoperative follow-up radiographs revealed improved congruency of the hips and preserved joint spaces although both of the femoral heads deformed probably due to the molding effect of the dysplastic acetabulums (Fig. 3).

Discussion

There are several methods of treatment for disorders of the hip in bone dysplasia. Canale (2) recommended total hip arthroplasty instead of arthrodesis for dwarfs with severe arthritis. Bassett and Scott (1) recommended femoral osteotomy or acetabular augmentation.

In this case, we performed femoral varus osteotomy as a treatment of choice. The reasons why we selected this method are: (i) it improves congruency of the hip, (ii) this patient weighed only 28 kg and it was presumed that pelvic osteotomy would be too invasive, and (iii) total hip arthroplasty was not preferable for this patient because of her young age. If her body had been bigger, pelvic osteotomy might have been the best choice.

Goldberg (3) emphasized the importance of arthrography in considering osteotomy. In this case, arthrograms of the hips showed better

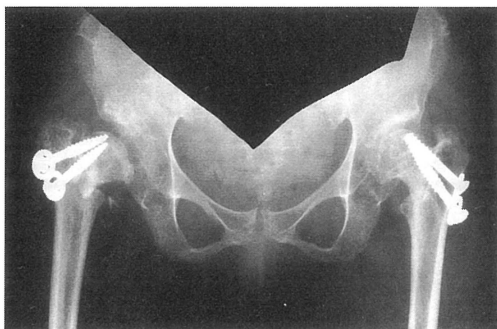


Fig. 2 Post-operative radiograph at 14 years of age.



Fig. 3 Radiograph at 19 years of age.

congruity when the hips were abducted.

A diagram of transtrochanteric curved varus osteotomy of the femur (4) is shown in (Fig 4). It shows that after osteotomy the greater trochanter apparently moves proximally a great deal, but in actual fact, the insertion of the abductor muscles moves only a little. Furthermore, the greater trochanter moves laterally which increases the lever arm effect. The advantages of this procedure over conventional osteotomies are as follows. (i) This procedure is less surgically invasive than pelvic osteotomy. (ii) Postoperative shortening of the leg is minimal. (iii) Elevation of the insertion of the gluteus medius and minimus muscles on the greater trochanter is small (Fig. 4). (iv) Fixation is easily achieved with screws. (v) The broad contact area at the osteotomy site allows rapid bone union. (vi) There is little risk of damaging the nutrient arteries to the femoral head. The first three are advantageous especially for a case of dwarfism.

Goldberg (3) pointed out that caution should be made because joint stiffness may be accelerated after hip surgery, with disappointing results. In our case, ranges of motion of both hips were a little restricted but still good after surgery. At 20 years old, she used a cane only outside her

house and could walk without any pain in either hip. This reveals that this method gives good intermediate term results because there would be no further remodeling of the joint, although longer follow-up will be necessary to determine the ultimate results. If conservative methods were chosen, the degenerative changes might have progressed leading to severe coxalgia and much restricted hip motion. We think this method can be a treatment of choice to improve activity in daily life at school age and delays the timing of total hip arthroplasty.

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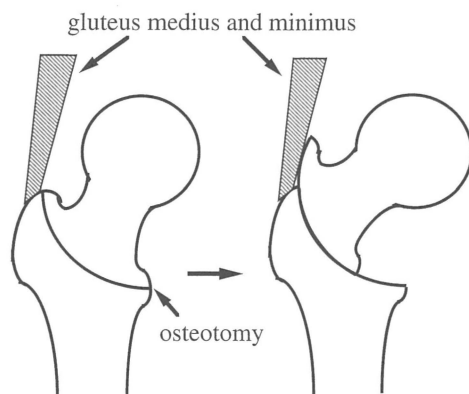


Fig. 4 Diagram of transtrochanteric curved varus osteotomy of the femur.

(和文抄録)

転子間彎曲内反骨切り術を施行した遅発性脊椎骨端異形成症の1例

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遅発性脊椎骨端異形成症の股関節症に対し、転子間彎曲内反骨切り術を施行し良好な成績を得たので報告する。症例は身長114 cm、体重28 kgの女性。10歳頃両股関節痛出現。レントゲン上、両股関節の亜脱臼、不整像、臼蓋形成不全を認めた。13歳時に左大腿骨に、また14歳時に右大腿骨に、転子間彎曲内反骨切り

術を施行。術後、両股の適合性は改善し、20歳現在、長時間の歩行後も疼痛はなく、日常生活の活動性は大きいに改善した。転子間彎曲内反骨切り術は、侵襲が少なく、脚短縮や大転子高位を起こしにくいため、小児症の股関節症の治療の選択肢になると思われた。