CASE REPORT

Surface arthroplasty in a cerebral palsied patient with hip dysplasia and dislocation [version 1; referees: 1 approved, 1 approved with reservations]

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Abstract

Hip dysplasia is considered to be an etiological factor in the progress of developmental osteoarthrosis of the hip. In patients with cerebral palsy, hip dislocation plus other anatomical alterations such as acetabular dysplasia, coxa valga, excessive femoral anteversion and muscle imbalance can be factors leading to coxarthrosis and pain. Total hip arthroplasty is a valuable method of treatment in some of these patients. However, sometimes young age, bone anatomy and shape, and the possible lack of stability of conventional designs in the context of big surgeries with multiple muscle releases and possible osteotomies make this arthroplasty a very difficult indication to reproduce as a surgeon. Hip resurfacing is a good option because it preserves as much bone stock as possible, doesn’t need corrective osteotomies and gives the prosthesis improved stability. The main concern about this type of prosthesis is the suspected lower survival rate of the femoral component, compared with regular total hip arthroplasties. We present a cerebral palsied patient with hip dysplasia who underwent resurfacing surgery. At a follow up one year after surgery, the patient was pain free, had recovered his motion and functionality, and was able to walk.
Introduction

Cerebral palsy is a syndrome caused by a non-progressive upper motor neuron lesion, originating before the central nervous system is mature. Patients suffer abnormal control of motor and sensory function and sometimes abnormal intellectual development. There are different etiological factors but perinatal anoxia is one of the most common. The spastic type is the most frequent and is characterized by rigid muscles with an inability to relax that produces a variety of anatomical and clinical signs. The incidence of hip dysplasia is very high in these patients, including adduction contractures, increased femoral anteversion, coxa valga and acetabular dysplasia. Pain is highly associated with these abnormalities and up to 50% of patients have hip dislocation. A dislocated painful hip in a patient with cerebral palsy can be treated by muscle releases, femoral or pelvic osteotomies and reduction, Girdlestone, arthrodesis or total hip arthroplasty. In a young patient with a good quality of life who was previously able to walk, we think that the best option is a total hip arthroplasty in order to relieve pain, allow perineal care and facilitate mobility and the ability to walk independently.

Case report

In 2011, a 15-year-old boy, Caucasian with triplegic cerebral palsy came to the clinic in a wheelchair, accompanied by his family. He complained of intense pain in his right hip that had been increasing over the last years. Unfortunately, the pain had translated to poor function. He had sparing of his upper left extremity of the hip. He described his hip function as limited and poor, and as continuously worsening. He tried not to bear weight on his right leg, although he was still able to walk around and transfer himself, usually with a Kay posture walker, in his house and around a baseball field.

In addition to the pain, he also had a history of right hip subluxation. Ten years previously, he had bilateral hip soft tissue and muscle releases, and osteotomies which were varus producing. Four years before, he had spinal fusion to help his alignment, and that did seem to help him to some degree. At that point his doctors noticed that his hip was coming out of the socket. His parents were very fearful of him losing his hip functionality as they saw that it had been getting worse in the four years after the spinal fusion. They had seen other doctors who had recommended Girdlestone rather than total hip replacement.

Subsequent X-rays demonstrated hip dysplasia and a dislocated joint with bowing of his femur in the subtrochanteric area. The head was completely out-of-round (Figure 1). The patient also had mild mitral regurgitation and suffered some spasticities whilst on a baclofen pump.

On physical examination he had some spasticity in his lower extremities and very limited movement of his right hip mainly because of pain. His muscle strength was 4/5 in the right leg. His adductor tendons could be felt but they did not seem overly tight.

At this point, total hip replacement was recommended, and the family were informed that he would most likely require subtrochanteric osteotomy, realignment of his femur, placement of a modular-type implant, formation of a new socket and soft tissue releases.

During the reviewing and planning of the case and surgery, we thought about the possibility of implanting a resurfacing hip prosthesis with the idea of maximizing the benefits of this type of device in our patient. Doing so would decrease the duration of the surgery and we wouldn’t need to perform an osteotomy on the femur with consequent lower blood loss and reduced perioperative risks. Also we could achieve the best intrinsic stability and maintain as much proximal femoral bone stock as possible. In addition, the bearing surfaces were perfect as he was a very young patient. Our main concern was the durability of the replacement in terms of loosening or fracture.

The family and the patient were carefully informed about the pros and the cons of both surgical options and they accepted our indication of hip resurfacing rather than conventional hip replacement.

The surgery was conducted in the usual manner, approaching the hip posteriorly without any special issues. The new hip was relocated, moderate soft tissue release was realized and intraoperative regular mobility and stability were accomplished. Postoperative X-rays demonstrated that the implants were well positioned and normal anatomy was almost perfectly recovered (Figure 2 and Figure 3).

Four to five days following surgery, the patient started walking and was put on physical therapy in the typical time and fashion. He had to use an abduction brace for 6 weeks post-surgery. He recovered well but slowly.

At a follow up one year after surgery, the patient was pain-free, walked with a one-handed assist, played without restrictions and had recovered a very good functional status. The passive range of motion in his right hip was much better than it was preoperatively, allowing him to sit and facilitating perineal care. His improvements had been very slow but his gait was still getting better each day.

Figure 1. X-ray showing a dislocated and dysplastic joint.
ambulatory patients\textsuperscript{4,5}. Resurfacing hip prosthesis is a type of hip replacement that should be considered in these patients because of its benefits in terms of stability\textsuperscript{6}, preservation of bone stock and low wear expectations\textsuperscript{11,12}. The presence of femoral deformities could be considered as a relative contraindication for resurfacing of the hip\textsuperscript{9}, but in some cases the surgeon should consider that hip resurfacing avoids the difficulty sometimes encountered in patients with dysplasia during total hip replacement of a narrow and frequently curved proximal femoral canal\textsuperscript{6}. The survivorship of this implant at five years in patients with different levels of dysplasia is similar to the rates for total hip replacements, always above 95\%\textsuperscript{6,7}. The clinical outcomes of resurfacing with respect to pain scores, restoration of the joint biomechanics, range of motion and walking and sport activities are also very good at five and ten years\textsuperscript{2,4,6,8}, even some measurements such as range of motion can be better after hip resurfacing than after total hip replacement\textsuperscript{7}. Furthermore, there is the possibility of an easier conversion to a total hip arthroplasty if needed.

There are no reports about the use of hip resurfacing in cerebral palsied patients but considering that these devices have demonstrated good results in dysplastic hips, we think that cerebral palsy may not be a complicating factor and the results or resurfacing could also be comparable to those obtained in not palsied individuals with dysplasia. For instance, it has been shown that other procedures such as acetabuloplasty, pelvic osteotomies and femoral osteotomies with or without open reduction of the hip have similar outcomes in patients with and without cerebral palsy\textsuperscript{3}.

Further studies will be needed to demonstrate conclusively that resurfacing is as valuable as total hip replacement in the majority of cerebral palsied hips.

**Consent**

Written informed consent for publication of their clinical details and clinical images was obtained from the patient and his parents.

**Author contributions**

Both authors followed the patient for more than a year. CGR was the main person involved in writing the case and doing the review of the literature, PGR revised the manuscript. Both authors agreed the final manuscript for publication.

**Competing interests**

No competing interests were disclosed.

**Grant information**

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References

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Current Referee Status: ✅

Version 1

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The presented interesting case study deals with a 15-year-old Caucasian boy, with triplegic cerebral palsy. Athetosis is not mentioned.

From my personal experience of nearly 40 years I favour THA for the treatment of painful hips with subluxation or dislocation with spasticity, combined with the necessary soft tissues and muscles release; a procedure which has long term acceptable outcomes. The survivorship for THA, in cerebral palsy patients, is referred to be 85% for 10 years with relief from preoperative pain and improved function. Nevertheless, because of the difficulty of preoperative assessment and of selecting the right surgical indications, orthopaedic surgeons have been reluctant to recommend THA in these patients. In the present case study the authors attempted to treat an adolescent boy with a resurfacing hip arthroplasty, combined with the necessary soft tissue and muscles release, a surgical procedure which has not been used previously for such a case. So far, according to the present authors, the proposed surgical procedure is successful.

Since the long term results for cerebral palsied patients treated with THA, cemented or not, are acceptable, I cannot see why a resurfacing arthroplasty would not be successful even in an adolescent patient of this group. Further studies are definitely needed as well as confirmation of long term successful results.

I have the following additional comments:

1. There is a lack of information about the preoperative status of the spinal column, the extent of spinal fusion and any benefits its correction had for the patient. Also, the x-ray of the pelvis shows a degree of obliquity, which might allow greater than average forces to act upon the hip.

2. One of the most important things to evaluate is the functional situation of a patient suffering from spasticity, which can lead to subluxation or dislocation and also to exclude athetosis, which complicates treatment or makes it impossible. Although it is not mentioned in this report I assume that the previous operative procedures have improved the muscle balance of the patient, a very important element, hopefully towards the right direction. Hip subluxation or dislocation in the cerebral palsy population is a problem for these patients and the reported incidence is from 18% to 59%. For painful hip subluxation or dislocation with arthrosis in the adolescent or adult, procedures such as hip arthrodesis, valgus osteotomy, proximal femoral resection, or total hip arthroplasty have all been done to relieve pain and improve function.
This brief case study outlines an alternate surgical approach of surface arthroplasty for a young cerebral palsied patient who was severely disabled by hip pain and dislocation.

While it is beneficial for clinicians to read about successful surgical interventions particularly for someone who is young and has difficult anatomy, the authors have been very sparing in their details about the case. As the patient had spastic triplegia, I am unclear as to which limb was spared from the statement "He had sparing of his upper left extremity of the hip" - does this mean his left arm or his left leg? Also it is difficult to understand how someone who has "limited and poor hip function" and "did not want to bear weight on his right leg" could "walk around with a Kay posture walker on a baseball field - which is usually quite large in area."

Some detail about how many levels of the spine were fused is required in the history. The authors should also comment on the patient's skeletal maturity as I would suspect that over the last four years the increase in long bone development and change in muscle mass may have exacerbated his hip joint position, symptoms and functionality. Was the patient skeletally mature when they performed the surface arthroplasty? The authors mentioned that when the spinal fusion was performed, "the hip was coming out of the socket" and continued to worsen after the spinal fusion, so perhaps they should have commented further on whether a spinal fusion is suitable for an immature cerebral palsied individual and on the effect a fusion may have on acetabular and femoral position.

I am surprised that all the muscles in his right leg were 4/5, particularly the hip external rotators and the extensors (gluteus maximus). The adductor tendon tightness with the changed hip joint centre, as the hip was dislocated, would be difficult to assess. The authors are referred to Delp & Maloney (1993), who investigated changing hip joint centre and moment arms of the muscles around the hip, specifically for consideration in diplegic patients.

I have read this submission. I believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Competing Interests: No competing interests were disclosed.