Modified wedge osteotomy for osteoarthritis of elbow secondary to osteochondritis dissecans in adolescent with multiple epiphyseal dysplasia. A case report

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ABSTRACT

Multiple epiphyseal dysplasia (MED) may be complicated by osteochondritis dissecans (OCD), most commonly reported in the knee, but involvement of the elbow is very rare. Optimal treatment for MED-associated elbow OCD and treatment outcome have not been established. This report describes the case of an adolescent male patient with elbow OCD treated by arthroscopic drilling at a previous clinic. Progression of osteoarthritis and radial head subluxation were observed. Resection of the osteophytes and modified wedge osteotomy of the lateral condyle were then performed, and a favorable result was obtained by decompression and sufficient congruency of the radiohumeral joint.

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1. Introduction

Multiple epiphyseal dysplasia (MED) is a genetic disorder associated with symmetric ossification centers and multiple growth abnormalities. The mode of inheritance is usually autosomal dominant or recessive, but sporadic cases have also been reported.^{1–3} The prevalence of MED is 9 in 100,000 births.⁴ MED most often affects large joints, especially the hips, and is often detected due to gait abnormalities and joint contractures during childhood.^{3,5} Early-onset osteoarthritis (OA) appears most often in the hips, knees, and ankles, but the shoulder, elbow, and wrist joints may also be involved.^{3,6–8} MED may also be complicated by osteochondritis dissecans (OCD), most commonly reported in the knee,^{9–11} but involvement of the elbow is very rare.^{6,9} Optimal treatment for MED-associated elbow OCD and treatment outcomes have not been established.

This report describes the case of an adolescent male patient with MED who presented with left elbow pain and limited range of motion (ROM). He had no improvement despite ROM training at an osteopathic clinic. Although elbow OCD had been treated by

arthroscopic drilling at a previous clinic, elbow contracture continued to worsen, and progression of OA and radial head subluxation were observed. Resection of the osteophytes and modified wedge osteotomy of the lateral condyle were then performed, and a favorable result was obtained by decompression and sufficient congruency of the radiohumeral joint.

2. Case report

The patient was a 13-year-old boy who had been found to have an abnormal gait in kindergarten, with abnormal ossification centers of the hips radiographically. At around 12 years 10 months of age, the patient experienced left elbow pain and limited ROM for no apparent reason. He began active ROM training at 13 years of age at an osteopathic clinic. However, the left elbow pain did not improve, and the limited ROM worsened. At age 13 years 2 months, he was diagnosed with OCD of the left elbow at a previous clinic. One month later, arthroscopic drilling and a synovectomy of the left elbow were performed. Although the elbow pain improved slightly, limited ROM of the elbow persisted.

Three months after surgery, the patient was referred to and initially evaluated by our department. The family history was unremarkable. He underwent surgery for an undescended left testicle during early childhood. The patient played tennis and was

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left hand-dominant. Blood biochemistry testing showed no abnormalities.

General physical examination revealed a Japanese adolescent male with normal growth, having a height of 163 cm and weight of 46 kg. The patient had a waddling gait. The right hip pain was not present, but ROM was severely limited: right sided flexion 60° (left 125°), extension 0° (left 10°), abduction 35° (left 35°), adduction 25° (left 20°), internal rotation 40° (left 70°), and external rotation 20° (left 15°). The Patrick, Trendelenburg, and Duchenne signs were absent.

Examination of the left elbow showed mild swelling laterally, with a palpable bony protrusion. Elbow ROM from extension to flexion was 15° to 150° on the right side and –20° to 50° on the left side. In particular, left elbow flexion was severely limited. Forearm pronation was 90° on the right and mildly limited to 70° on the left.

Imaging findings: Radiographs of the left elbow at the previous clinic showed a nondisplaced fragment with radiolucency of the surrounding fragment in the humeral capitellum, which was classified as Minami grade II.^{12,13} The transverse diameter of the radial head was slightly increased (Fig. 1A,B). Although arthroscopic drilling for OCD was performed 1 month later, at the initial evaluation 3 months postoperatively, radiographs showed joint space narrowing, osteophyte formation at the anterior condyle and olecranon fossa, and a tendency for anterior subluxation of the radial head (Fig. 1C,D). At 7 months after surgery, radiographs showed bone collapse of the OCD lesion, osteophyte enlargement in the surrounding joint, progression of OA, and anterior subluxation of the radial head. (Fig. 1E,F)

Hip radiographs showed abnormal ossification of the proximal femoral epiphyses bilaterally. There was anteromedial flattening and no epiphyseal segmentation. Radiographs of the spine showed slightly irregular end plates.

Computed tomography (CT) of the elbow showed osteophyte formation on the anterior part of the lateral condyle and radial notch, collapse of the humeral capitellum, and marginal bone sclerosis (Fig. 2A,B). On 3-dimensional CT, the osteophyte formation around the elbow and anterior subluxation of the radial head were more clearly observed (Fig. 2C,D).

Magnetic resonance imaging (MRI) of the elbow before arthroscopic drilling showed extensive OCD with low-intensity

T1 and T2-weighted images of the humeral capitellum on the affected site (Fig. 3A–C). A smaller area of OCD with low signal intensity was also observed on the unaffected right site (Fig. 3D–F). MRI after drilling and our evaluation showed low-intensity T1 and high-intensity T2 star-weighted images, severe collapse of the capitellum, and degenerative changes of the radiohumeral joint.

At 8 months after arthroscopic drilling, arthroplasty was performed with osteophyte resection and wedge osteotomy of the lateral condyle, together with radial head reduction. The elbow joint was exposed by a lateral approach. Intraoperative findings showed radial head hypertrophy, convex deformation of the radial joint surface toward the humeral capitellum, and degeneration and wear of the cartilage. The capitellum had loss of joint cartilage with subsidence and degeneration. Flexion of the elbow caused impingement between the anterior osteophyte and the radial head, and flexion was limited to 50° by these bony changes (Fig. 4A, B). Bone spurs of the radial notch and olecranon fossa consistent with the imaging findings were also present. These osteophytes were resected, and bone wax was applied to prevent bone spur reformation. Even after resection of the osteophytes, radiohumeral joint congruency was poor, without improvement in flexion. Therefore, wedge osteotomy of the lateral condyle was performed, with positioning in anterior rotation to prevent bony impingement with the radial head, and the condyle was stabilized with two screws (AO/ASIF cancellous screw ϕ 4.0 mm short). Thus, intraoperative elbow flexion up to 100° was achieved (Fig. 4C,D).

The pathological findings of the anterior osteophyte showed cartilage tissue covered by a capsule, with trabecular bone and adipose tissue directly beneath. This suggested the possibility of an intra-articular osteochondroma with a cartilage cap (Fig. 5).

The patient underwent physical therapy with left elbow ROM training for about 6 months, and by 7 months after revision surgery, he was able to return to playing tennis.

At final evaluation 5 years postoperatively, elbow ROM was up to 100° flexion and –5° extension, and the patient had no major problems with daily activities such as tying a necktie. Radiographs showed no new osteophyte formation or OA progression. The patient has had a good clinical course, including spontaneous improvement of OCD on the right side (Fig. 6).

3. Discussion

MED is a bone dysplasia often presenting with short stature and abnormal epiphyseal ossification in long bones. MED is typically associated with multiple bilaterally symmetrical abnormal epiphyses and early degenerative changes because of poor joint congruency.^{1–3,8} The more severe Fairbank type is associated with the 3 cardinal findings of dwarfism, short digits, and mottling or irregularities in density and outline of several of the developing epiphyses.¹ There is also a milder Ribbing type.² The present patient had no genetic predisposition to MED, his height and growth were normal, and he had no short digits. However, the patient had an abnormal gait from early childhood, and radiographs showed abnormal ossification centers of the hips with limited ROM. The epiphyses of the hips appeared flat rather than small, and slight spinal degeneration and bilateral OCD of the elbows were also present; this case was consistent with Ribbing type MED.

Most case reports of MED-associated OCD involve the knees,^{9–11} and Taketomi et al.¹⁰ reported using autograft reconstruction for early-stage osteochondral lesions with good long-term outcomes. However, no report was found of the surgical treatment for MED-associated OCD of the elbow.

The present patient with MED had an extensive OCD lesion of the left elbow. Without medical consultation, he received ROM training at an osteopathic clinic, which caused continuous

Fig 1. Serial radiographs of the left elbow. A, B: Radiographs at the previous clinic showed a nondisplaced fragment with radiolucency of the surrounding fragment in the humeral capitellum (Minami grade II). C, D: Radiographs at the initial evaluation 3 months after drilling showed joint space narrowing, osteophyte formation at the anterior condyle and olecranon fossa, and a tendency for anterior subluxation of the radial head. E, F: Radiographs at 7 months after drilling showed bone collapse of the OCD lesion, osteophyte enlargement in the surrounding joint, progression of OA, and anterior subluxation of the radial head.

Fig. 2. A B: CT of the elbow showed osteophyte formation on the anterior part of the lateral condyle, collapse of the humeral capitellum, and marginal bone sclerosis (arrow). C, D: 3-dimensional CT showed osteophyte formation around the elbow and anterior subluxation of the radial head (arrow).

Fig. 3. A B, C: MRI of the elbow before drilling showed extensive OCD with low-intensity T1 and T2-weighted images of the humeral capitellum on the affected site (arrow). D, E, F: A smaller area of OCD with low signal intensity was also observed on the unaffected right site (arrow).

dynamic loading of the elbow. Furthermore, bone drilling for OCD was performed. Unfortunately, these factors led to collapse of the humeral capitellum. This resulted in poor congruency of the radiohumeral joint, progression of OA, and anterior subluxation of the radial head.

With regard to bone spurs of the anterior part of the lateral condyle, sequential imaging findings showed osteophytic changes as bone spurs with impingement due to poor congruency between the radial head and humeral capitellum. Pathological examination suggested a possible osteochondroma covered by a cartilage cap. These protruding lesions made it unlikely that the bone spurs were

Fig. 4. Operative findings. A, B: Flexion of the elbow caused impingement between the anterior osteophyte and the radial head, and flexion was limited to 50° by these bony changes. C, D: Modified wedge osteotomy of the lateral condyle was performed, with positioning in anterior rotation to prevent bony impingement with the radial head.

(hematoxylin-eosin stain; low power)

Fig. 5. The pathological findings of the anterior osteophyte showed cartilage tissue covered by a capsule, with trabecular bone and adipose tissue directly beneath.

simply associated with OA. Various factors, including MED, continuous dynamic loading of the elbow, and irritation by bone drilling may have resulted in this osteochondroma. As previously indicated in Shariatzadeh's report, intra-articular osteochondroma of the elbow is very rare.¹⁴

There have been numerous reports of the surgical treatments and favorable outcomes in OCD of the humeral capitellum.^{12,15–17} However, poor treatment outcomes with bone drilling or curettage for extensive OCD lesions have been reported.¹⁷ Mechanical irritation, including bone drilling, of the epiphysis may cause lesion collapse, possibly leading to osteochondromas in this type of patient. Therefore, surgical treatment must be carefully selected.

There have been no reports of the surgical treatment for an extensive OCD lesion of the elbow associated with MED. Moreover, treatment in patients with radial head subluxation¹⁸ secondary to

Fig. 6. Radiographs of bilateral elbows at 5 years after revision surgery showed no new osteophyte formation or OA progression, including spontaneous improvement of OCD on the right side.

OCD can be difficult. Revision surgery includes closing-wedge osteotomy of the lateral condyle,^{19,20} reconstruction with osteochondral autograft,^{21,22} and radial head excision.²³ As for radial head subluxation, ulnar corrective osteotomy,²⁴ radial shortening osteotomy,²⁴ and ulnar distraction osteogenesis²⁵ are also considered. However, whether a transplanted osteochondral autograft can adapt to axial loading and survive remains uncertain in patients with degenerative joint changes, marked radial head hypertrophy, and convex deformity of the radial head. The indication for radial head excision is also limited in such early

adolescent patients. In addition, radial shortening osteotomy and ulnar distraction osteogenesis are additional treatments for intra-articular bony impingement. Although closing-wedge osteotomy may be a problem in osteonecrosis of the lateral condyle, favorable outcomes in OCD have been reported.^{19,20} The present patient already had loss of congruency of the radiohumeral joint due to degenerative changes and bony impingement. Therefore, radial head excision would be indicated in an adult case, but because the present patient was an early adolescent, modified wedge osteotomy of the lateral condyle was performed. First, bone spurs causing bony impingement were resected to improve restricted elbow motion. Then, a modified wedge osteotomy of the lateral condyle was performed, with anterior rotation for joint congruency in a flexion position. Flexion up to 100° was possible, but because of a long-term joint contracture, there was hypertonia of the triceps brachii muscle, so further intraoperative restoration of ROM could not be achieved. V-Y advancement of the triceps brachii fascia was also considered, but this is highly invasive and was not performed.

Elbow pain improved soon after the revision surgery, and because bony impingement had been relieved, active ROM training was started. Now, as an adult 5 years after surgery, the patient has had a good clinical course. Flexion ROM is limited to 100°, but radiographs show good elbow joint congruency and no progression of OA. OCD on the unaffected side has resolved spontaneously, and no epiphyseal morphological defect is present.

Elbow OCD is a frequently encountered disorder in young athletes.^{12,16} Diagnosis and treatment have often been reported, and improved treatment outcomes have also recently been described. However, useful treatment for elbow OCD as a complication of MED has not been established. In patients such as the present case with elbow OCD due to MED, physical irritation can cause spur-like osteochondromas and rapid progression of OA, so that careful diagnosis and treatment are required in these patients.

4. Conclusions

1. A patient with MED associated with elbow OCD who developed elbow OA and anterior subluxation of the radial head was presented.
2. Bone drilling of the osteochondral lesion and continuous dynamic loading were factors leading to osteochondroma of the elbow.
3. Resection of bony protrusions and modified wedge osteotomy of the lateral condyle improved flexion contracture of the elbow.
4. Treatment of elbow OCD should include evaluation for the presence of MED. Surgical treatment must be carefully selected in these patients.

Conflict of interest

The authors have none to declare.

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