CASE REPORT

Nagoya J. Med. Sci. **84**. 185–199, 2022
doi:10.18999/nagjms.84.1.185

Good result for twelve years after bilateral hip and knee arthroplasties for ankylosis with juvenile idiopathic arthritis: a case report

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ABSTRACT

Juvenile idiopathic arthritis (JIA) can lead to joint deformity and bone destruction, which can cause gait disturbances. To the best of our knowledge, there are no case reports with over 10 years of follow-up on quadruple joint arthroplasties (QJA) for bilateral hip and knee ankylosis associated with JIA. We present the case of a 29-year-old woman with JIA. The patient suffered from bilateral ankylosis of the hips and knees and developed a swing gait requiring double crutches. We performed staged QJA with careful attention to postoperative rehabilitation and her physical features, which included excessive pelvic anteversion, poor bone quality, and short statue of bones. Twelve years after surgery, the patient was able to walk without any support and showed good clinical functional scores. In addition, no radiological loosening following QJA was observed. We hereby introduce a surgical strategy for total hip arthroplasty for excessive pelvic anteversion, which involves two methods to calculate pelvic tilt on a pelvic anteroposterior radiograph. These methods were able to approximately predict postoperative pelvic changes.

Keywords: total hip arthroplasty, total knee arthroplasty, juvenile idiopathic arthritis, pelvic tilt

Abbreviations:
JIA: juvenile idiopathic arthritis
QJA: quadruple joint arthroplasties
THA: total hip arthroplasty
TKA: total knee arthroplasty
ROM: range of motion
AP view: anteroposterior view
HHS: Harris Hip Score
JOA score: Japanese Orthopaedic Association score
the IBBC technique: the interface bioactive bone cement technique
POD: postoperative day

Received: May 11, 2021; accepted: June 28, 2021

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INTRODUCTION

Ankylosis can occur secondary to inflammatory diseases, infection, injury, or surgery. Juvenile idiopathic arthritis (JIA), known as juvenile rheumatoid arthritis, is defined as arthritis persisting for > 6 weeks after onset in patients aged < 16 years. Even though the prognosis of JIA has significantly improved with the advent of biologics, it can still lead to joint destruction.[1] There have been two case reports on the 1.5- and 4-year outcomes of JIA patients after bilateral hip and knee arthroplasties for ankyloses.[2,3] However, no case reports of quadruple joint arthroplasties (QJA) with a 10-year follow-up currently exist. In patients with JIA, there are high rates of revision after total hip arthroplasty (THA) or total knee arthroplasty (TKA).[4-7] We hereby report a case that showed good 12-year outcomes of QJA for bilateral hip and knee ankylosis in a patient with JIA.

CASE REPORT

A 29-year-old woman presented with a 27-year history of JIA. The patient was able to walk normally until 24 years of age. At 25 years of age, she began experiencing bilateral pain in her hips and knees. Disease activity was controlled with methotrexate 8 mg weekly and infliximab 200 mg bimonthly. However, the patient developed progressive deformity in the four joints. She developed a swing gait and required double crutches and had difficulty standing independently.

On physical examination, her height was 149 cm, body weight was 46 kg, and body mass index was 20.7 kg/m^2. Flexion contractures with severely limited ranges of motion (ROM) were observed in the hips and knees (Table 1). Preoperative radiographs of both hips and knees showed that all four joints had significant deformities. Anteroposterior (AP) radiographs of both hips showed the absence of joint spaces, excessive anterior tilt of the pelvis, and narrow medullary cavities of both femurs (Fig.1). Radiographs of both knees showed the absence of patellofemoral and femorotibial joint spaces (Fig.2). Lateral radiographs of the lumbar spine showed a lumbar lordosis angle of 42° with preserved intervertebral disc spaces (Fig.3). Laboratory investigations revealed a well-controlled inflammatory response. The bilateral hips and knees were eligible for arthroplasty.

Table 1 The progressions of ROM, HHS and JOA knee score before and after bilateral total hip arthroplasties

			Pre.	5 m.	1 y.	2 y.	3 y.	4 y.	8 y.	12 y.
ROM (degrees)	Hip	flex	30/40	60/70	80/90	80/90	90/95	90/90	90/95	95/100
		ext.	–20/–30	0/0	0/0	2/2	2/3	3/3	4/5	8/10
		abd.	10/10	15/15	20/25	15/20	30/30	30/30	30/30	25/30
		add.	–10/–10	10/10	15/15	10/15	30/30	40/30	40/20	30/25
		ex. r.	0/0	3/0	10/10	10/5	10/5	5/5	10/15	10/15
		in. r.	0/0	10/15	20/25	10/10	10/15	10/20	20/20	15/20
	Knee	flex	30/40	50/55	70/80	80/70	80/75	75/75	70/80	70/75
		ext.	–20/–30	–20/–30	–10/–10	0/0	0/0	0/0	0/0	0/0
HHS (points)			15/15	58/64	79/81	85/87	95/95	95/95	97/97	94/98
JOA knee scores (points)			10/10	20/20	60/70	80/70	80/80	80/80	70/80	70/80

This table is shown in the manner of right/left.
ROM: range of motion
HHS: Harris Hip score
JOA: Japan Orthopedic Association
Pre.: pre-operation
m.: month
y.: year
ex. r.: external rotation
in. r.: internal rotation

Fig. 1a

Fig. 1b

Fig. 1 Preoperative anteroposterior hip radiographs
Fig. 1a: The hip in the supine.
Fig. 1b: The hip in the standing position.

Fig. 2a

Fig. 2b

Fig. 2c

Fig. 2d

Fig. 2 Preoperative radiographs of the knees

Fig. 2a: The right knee in the anteroposterior (AP) view.
Fig. 2b: The left knee in the AP view.
Fig. 2c: The right knee in the lateral view.
Fig. 2d: The left knee in the lateral view.

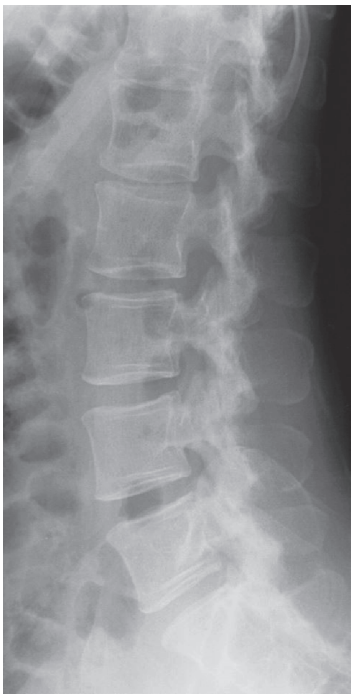

Fig. 3 Preoperative lateral radiograph of the lumbar spine showing a lumbar lordosis angle of 42° with normal intervertebral spaces

Clinical and radiological assessments

Clinical evaluations included the Harris Hip Score (HHS) and the Japanese Orthopedic Association (JOA) knee score.[8,9] The JOA knee score involved the following four categories: pain on walking (30 points), pain while walking up and down the stairs (25 points), ROM (35 points), and degree of joint swelling (10 points). A maximum of 100 points implied a healthy hip or knee.

Preoperative and follow-up radiographic evaluations included an AP view of both hips in the supine and standing positions, as well as AP and lateral views of the knees in the supine position. Pelvic tilt was retrospectively estimated on AP radiographs using Konishi's formula and Muir's nomogram.[10,11] Pelvic tilt calculated by Konishi's formula was expressed as pelvic angle. Cup anteversion was calculated as the ratio of the length of the major axis to that of the minor axis of the cup ellipse on the AP radiographs.[12] Cup inclination was directly measured.[13]

Preoperative planning

Two issues needed to be addressed during THA: the excessive pelvic anteversion and the narrow medullary cavity. The pelvis would tilt backward following surgical correction of the flexion contracture of the hip. The preoperative pelvic tilt in the supine position was calculated to be –1.6° using Konishi's formula.[10] By considering a normal pelvic tilt of 15–20º based on our experience, we estimated that her pelvic tilt would increase approximately 20° posteriorly. Previous studies have shown that the ratio of cup anteversion changes based on pelvic tilt change was approximately 0.7.[14] Therefore, we targeted the intraoperative cup anteversion angle at 10° instead of the usual 25° in our institute. For the narrow cavity, we prepared a thin cementless stem (S-ROM; DePuy, Leeds, United Kingdom) in addition to a cemented stem (Exeter; Stryker

Orthopaedics, Mahwah, NJ, USA), which is a standard practice in our institute. We also prepared ultrahigh-molecular-weight polyethylene (UHMWPE) fiber cables (NESPLON Cable System; Alfresa Pharma Co., Osaka, Japan) for possible femoral fractures.

Special attention was paid to the patient's small tibia during TKA. On computed tomography, the tibial mediolateral measurements were 58 mm/60 mm (right/left), while the AP measurements were 35 mm/38 mm (right/left). An anatomical report indicated that the average tibial mediolateral measurement was 69 mm, and the tibial AP measurement was 45 mm in East Asian women.[15] For the narrow tibia, we chose the smallest posterior-stabilized component available in our country at the time (Hyflex Knee; DePuy, Leeds, United Kingdom), rather than the standard component used in our institute (KU knee; Kyocera Co., Kyoto, Japan).

We planned to fix these implants with cement using the interface bioactive bone cement (IBBC) technique. The IBBC technique applies hydroxyapatite to the bony surface before cementing.[16,17]

Clinical course

The patient underwent staged bilateral THAs, followed by staged bilateral TKAs. She first underwent a right THA, which was performed using the modified Dall's approach in the lateral position.[18] The anteversion of the acetabulum component was targeted at 5° intraoperatively with a mechanical guide on the pelvic fixator. An all-polyethylene cup (Aeonian; Kyocera Co. Ltd, Kyoto, Japan) with an outer diameter of 48 mm was fixed with Simplex P bone cement (Stryker Orthopaedics, Mahwah, NJ, USA) using the IBBC technique. When attempting temporary reduction with a cementless trial stem (S-ROM), a longitudinal fracture occurred in the medial femoral cortex. We reinforced the fractured shaft with two UHMWPE fiber cables of 3.0 mm in width and then fixed a cemented stem (Exeter) of size 33 using the IBBC technique. The right THA was completed using an alumina ceramic head (Stryker Orthopaedics, Mahwah, New Jersey) of 28 mm in diameter. The patient was mobilized on postoperative day (POD) 2. On POD5, she started weight-bearing with a 3-point gait. She could then ambulate on a flat surface with a 2-point alternate crutch gait.

Twenty-one days after the right THA, the patient underwent left THA using the same acetabular procedure. Based on our experience with right hip fractures, a cemented stem (Exeter) of size 30 was fixed using the IBBC technique. Using a metal head (Exeter) of 28 mm in diameter, the left THA was completed without any fractures. The patient was mobilized on POD2. On POD12, she could go up and down the stairs with a 2-point alternate crutch gait without any complications. She was discharged on POD20 and continued outpatient rehabilitation. Postoperative AP radiographs of the hips two months after THA are shown in Figure 4.

Five months after bilateral THA, the knee flexion angles improved from 30° to 50° on the right and from 40° to 55° on the left (Table 1). However, no changes in the knee extension angles were observed. Left TKA was performed using the standard medial parapatellar approach. The smallest components (Hyflex Knee) were fixed using the IBBC technique. Additionally, we performed a lateral release of the patellofemoral joint to improve tracking. On POD1, the patient began ROM training using a continuous passive motion machine for 1 h per day and was mobilized. On POD2, she started weight-bearing with a 3-point gait. She could then walk on a flat surface with a 2-point alternate crutch gait.

The right TKA was performed 21 days after the left TKA in a similar fashion without any perioperative complications. On POD17, the patient could go up and down the stairs with a 2-point alternate crutch gait. She was discharged with a single crutch on POD28 and continued outpatient rehabilitation. Postoperative radiographs of the knees at two months after TKA are shown in Figure 5.

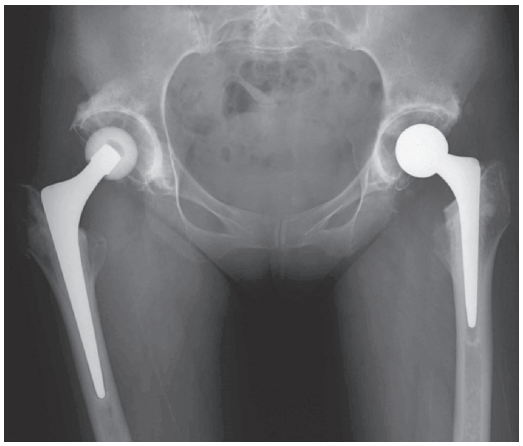

Fig. 4a

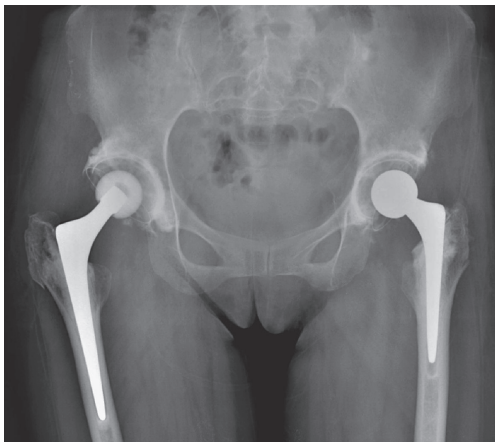

Fig. 4b

Fig. 4 Postoperative anteroposterior radiographs of the hips 2 months after total hip arthroplasty
Fig. 4a: The hip in the supine position.
Fig. 4b: The hip in the standing position.

Rehabilitation continued for one year after bilateral THA. The patient finally began to walk unsupported 1.5 years after THA and walk up and down the stairs unsupported three years after THA. She resumed desk work and commuting by train. She got married nine years after THA. At the last follow-up, 12 years after THA and 11.5 years after TKA, she had successfully reintegrated into the community without limitations.

Outcomes

The progressions of ROM, HHS, and JOA knee scores are shown in Table 1. Just before the first TKA, five months after the last THA, the knee flexion angles had improved, although the extension angles did not change. Both parameters improved after the last TKA. Although the ROM of the hips gradually increased, the ROM of the knees reached a plateau two years after the last TKA. Her HHS and JOA knee scores improved. At the last follow-up, her hip and knee function were assessed as excellent.

The progressions of pelvic tilt, pelvic angle, cup anteversion, and cup inclination are shown in Table 2. The pelvis had retroverted after THA and reached a plateau one year later in the supine position and four years later in the standing position (Figure 6). Cup anteversion and inclination angles were within normal ranges at the last follow-up. It was appropriate to set the target of the intraoperative cup anteversion at 5°.

Fig. 5a

Fig. 5b

Fig. 5c

Fig. 5d

Fig. 5 Postoperative radiographs of the knees two months after total knee arthroplasty
Fig. 5a: The right knee in the anteroposterior (AP) view.
Fig. 5b: The left knee in the AP view.
Fig. 5c: The right knee in the lateral view.
Fig. 5d: The left knee in the lateral view.

Table 2 The progressions of pelvic tilt, cup anteversion and cup inclination before and after bilateral total hip arthroplasties

		Pre.	2 m.	6 m.	1 y.	2 y.	4 y.	6 y.	8 y.	12 y.
Pelvic Tilt	Sup.	30.6	22.5	16.2	8.9	10.2	11.0	10.0	10.4	8.6
	Stand	33.3	15.7	10.7	10.6	12.4	4.5	6.5	3.7	3.8
Pelvic Angle	Sup.	–1.6	9.6	8.8	14.4	13.2	14.3	12.2	14.3	14.5
	Stand	–7.2	14.5	14.4	17.8	11.7	20.7	17.3	20.0	18.7
Cup Antev.	Sup.	-	7.6 /9.4	11.6 /18.6	12.7 /24.4	15.2 /22.1	17.7 /17.6	14.7 /21.6	13.8 /23.0	15.7 /22.1
	Stand	-	14.9 /14.5	16.9 /21.1	13.6 /23.6	16.1 /23.9	20.6 /22.8	18.3 /20.9	20.0 /26.3	20.8 /24.2
Cup Inclin.	Sup.	-	40.0 /55.8	42.1 /56.8	42.4 /59.5	43.1 /59.0	42.5 /58.8	42.1 /59.0	43.4 /59.3	43.2 /59.2
	Stand	-	41.0 /58.0	42.6 /58.8	43.0 /59.4	43.4 /59.2	43.1 /60.2	43.0 /60.0	43.7 /59.7	43.3 /60.7

This table is shown in the manner of right/left. All numbers are in degrees.
Pelvic Tilt was estimated by Muir's nomogram. This indicates the angle of the anterior pelvic plane relative to a vertical axis, positive for anterior tilt. Pelvic Angle was calculated by Konishi's formula. This indicates the amount of change from the average pelvic tilt, positive for posterior tilt.
Pre.: pre-operation
m.: month
y.: year
Sup.: supine
Antev.: anteversion
Inclin.: inclination

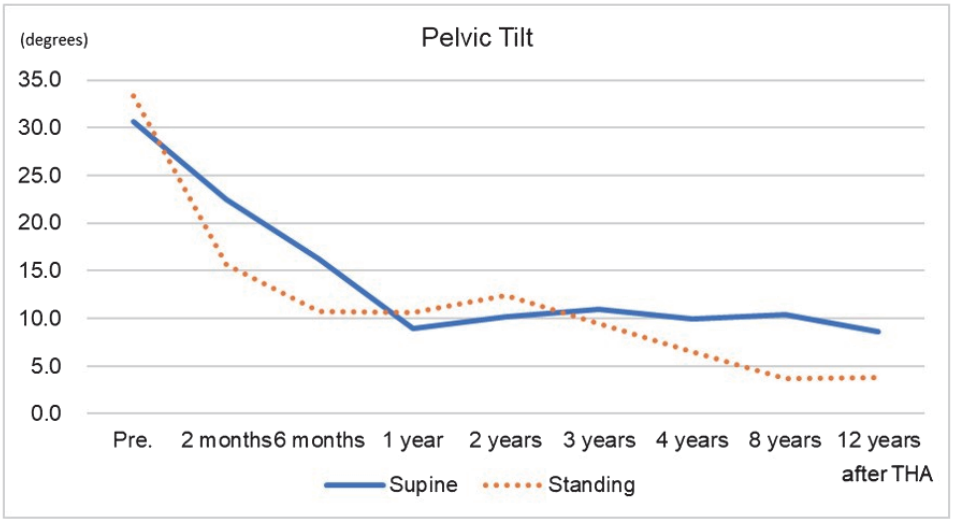

Fig. 6 A chart of pelvic tilt estimated using Muir's nomogram
Pelvic tilt change reached a plateau one year after total hip arthroplasty (THA) in the supine position and four years after THA in the standing position.

The difference in pelvic tilt between two months and 12 years after THA was 13.9° in the supine position and 11.9° in the standing position. The difference in pelvic angle was 4.9° and 4.2°, respectively. Furthermore, the difference in cup anteversion in the right hip was 8.1° and 5.9°, respectively, while that in the left hip was 12.7° and 9.7°, respectively. The ratio of cup anteversion change to pelvic tilt change was 0.7 with Muir's nomogram and 2.0 with Konishi's formula.

There was no evidence of osteolysis or radiolucent lines after QJA at the last follow-up (Figures 7 and 8) and dislocation did not occur. The patient was satisfied with her ability to perform activities of daily living independently without any pain.

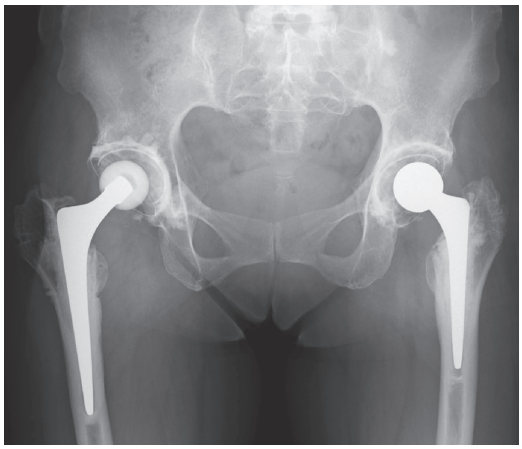

Fig. 7a

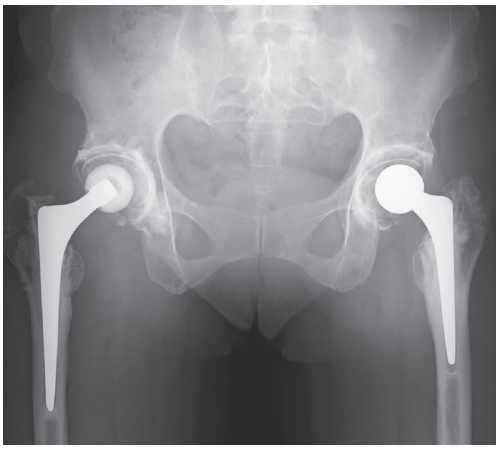

Fig. 7b

Fig. 7 Postoperative anteroposterior radiographs of the hips 12 years after total hip arthroplasty
Fig. 7a: The hip in the supine position.
Fig. 7b: The hip in the standing position.

Fig. 8a

Fig. 8b

Fig. 8c

Fig. 8d

Fig. 8 Postoperative radiographs of the knees11.5 years after total knee arthroplasty
Fig. 8a: The right knee in anteroposterior (AP) view.
Fig. 8b: The left knee in AP view.
Fig. 8c: The rightknee in lateral view.
Fig. 8d: The left knee in lateral view.

DISCUSSION

There are many studies on arthroplasty for JIA, including either THA-only or TKA-only procedures.[5-7] However, there are few studies on arthroplasties of all four joints for JIA.[2-4] Here,

we report a case with over 12 years of follow-up, with valuable findings on postoperative pelvic tilt changes.

There have been three reports of QJA in patients with JIA in the last 20 years. Karva et al presented a case report of a 1.5-year follow-up with good results and proposed that the hip should be treated initially.[2] Malhotra et al presented a case report of a four-year follow-up with satisfactory results and suggested that simultaneous QJA is an effective treatment for stiff legs.[3] These represent the short-term results of QJA. Malhall et al reported the 15-year outcomes of six patients with JIA who underwent QJA, including bipolar hemiarthroplasty in two of the six patients[4]; however, five of the six cases underwent revision THA at a mean follow-up of 12 years after primary arthroplasty. QJA for JIA may have poor long-term survivorship because of abnormal anatomical structures, long life span, and poor bone quality secondary to a combination of osteoporosis, immobility, and steroid use.[5,6]

Many reports of THA for JIA have shown acceptable survivorship at approximately ten years of follow-up.[5] However, several studies with over ten years of follow-up have reported a high rate of failure. De Ranieri et al showed that failure was observed in 32% of 37 primary THAs for JIA patients, with multiple joint destructions reported at 9.5–19.6 years of follow-up.[6] In addition, revision hip surgery in patients with JIA is even more challenging. In the study series by Goodman et al involving 24 revision THAs for 15 JIA patients, reoperation was required in 29% of patients.[20] Therefore, surgeons should treat patients with JIA with greater precision than those with osteoarthritis.

Although THA is one of the most successful treatments for hip disorders with excellent long-term outcomes concerning pain relief and improvement of function and quality of life, whether cement or cementless fixation is better remains controversial.[21] Such discussion is the same in THA for JIA. De Ranieri et al suggested that miniature cementless porous-coated stems can be used successfully.[6] However, especially for the femoral stem, Murray et al reported that many situations deviated from the indication for cementless stems, although cemented stems can be used in almost all cases.[22] Schmitz et al showed that with the use of Exeter stems in patients < 40 years of age, the 17-year survival rate was 92.5% and 100% when the endpoints were revision for any reason and aseptic femoral component loosening, respectively.[23] Our patient had no evidence of aseptic loosening at 12 years of follow-up. We believe that cement fixation can be better in special cases such as JIA.

Whether the use of cement with or without the IBBC technique is superior in the long term is also controversial. Oonishi et al showed that the IBBC technique could prevent radiolucent line and osteolysis for 24–29 years.[17] Our case might benefit from the IBBC technique.

Few studies have been published on QJA. All data reported before 1990 included 85–100% of RA cases, which not only included JIA. No surveys from 1990 to 2000 are available, and two studies reported since 2000 comprised 80% of osteoarthritis and 20% of RA cases.[24] Grauer et al reported the 6.8-year results of 26 patients who underwent QJA, 85% of whom were for RA or JIA.[25] They described that aseptic loosening occurred in 22.5% and 13.5% in the hips and knees, respectively. Although QJA is an effective treatment for severe arthritis in the four major lower joints, the survival rate of QJA in RA patients remains low because of poor bone quality.[5,6]

Intraoperative periprosthetic femoral fracture is a known complication of cementless THA. Miettinen et al described that patients with intraoperative fractures had more deviated femoral anatomies and thinner bone cortices in the proximal femur observed during radiological analyses.[26] They concluded that “to avoid intraoperative fractures, special attention should be paid when cementless stems are used with deviant-shaped proximal femurs and with thin cortices.” In this case, the cause of the intraoperative fracture seemed to be the extremely narrow medullary cavity of the proximal femoral diaphysis. When performing THA on patients with deformed proximal

femurs, surgeons should opt for cemented fixation instead of cementless fixation.

Regarding the order of QJA, the hips should be treated first. Karva et al emphasized that performing TKA with a stiff hip involves abnormal stress, and rehabilitation of the hip is possible with a stiff knee.[2] We observed an increase in knee flexion angles with THA and postoperative rehabilitation (Table 1). Such an increase in flexion angles was advantageous for the ROM of the knees after TKA. It seemed better to perform THA initially if ipsilateral THA and TKA were performed at different timepoints, considering postoperative rehabilitation. Alternatively, ipsilateral THA and TKA under single anesthesia or simultaneous QJA have also been reported.[3,27] These strategies should be considered as alternatives.

There is no debate regarding the importance of rehabilitation after total joint arthroplasty. The most important part of rehabilitation is ROM training to slightly increase the ROM obtained intraoperatively, because it is impossible to completely eliminate joint contractures of ankylosis intraoperatively. Therefore, surgeons should determine the residual ROM restrictions intraoperatively, as this will help prevent postoperative dislocation and set goals for postoperative rehabilitation. It is controversial as to which strength training should be performed for walking ability. In this case, quadriceps training was employed. The effects of quadriceps muscle strength on gait have been reported by two studies.[28,29]

Postoperative changes in pelvic tilt should be taken into account when considering acetabular component placement. However, methods for predicting the degree of change are unclear in clinical practice. Konishi formulated an equation for calculating pelvic retroversion on a pelvic AP radiograph: pelvic angle = (vertical extension of obturator foramen/distance between the teardrops) * x − y, where x = 137.4 and y = 23.1 in males, and x = 207.0 and y = 32.0 in females.[11] This parameter was positive for retroversion. Muir et al suggested a model in which the vertical distance between the superior margin of the pubic symphysis and the trans-ischial line was determined as the most reliable parameter on AP radiographs for the calculation of pelvic tilt and developed sex-specific nomograms.[12] By calculating the pelvic tilt using these two methods, the results of the pelvic tilt changes in our case were identical (Table 2). Previous studies have shown that the ratio of cup anteversion changes to pelvic tilt change was approximately 0.7.[14] In this case, the ratio was 0.7 with Muir's nomogram and 2.0 with Konishi's formula. Therefore, Muir's nomogram was more practical than Konishi's formula. If accurate prediction of pelvic tilt changes after THA was possible, the appropriate intraoperative angle for cup implantation can be calculated.

CONCLUSION

We present a case of QJA for bilateral hip and knee ankylosis in a patient with JIA. QJA for ankylosis dramatically improved the mobility and quality of life of this patient. Twelve years after THAs and 11.5 years after TKAs, her joint function was good with no evidence of radiological loosening, and dislocation did not occur. QJA may be an effective treatment for severe joint dysfunction in JIA patients with ankylosed joints. It is essential to select implants with excellent long-term survival and appropriate surgical strategies by considering postoperative pelvic tilt changes and rehabilitation.

ACKNOWLEDGEMENT

We are grateful to the Rehabilitation Center of Kyoto Katsura Hospital for the rehabilitation

of the patient. We would like to thank Editage (www.editage.com) for English language editing.

CONFLICT OF INTEREST STATEMENT

The authors did not receive and will not receive any benefits or funding from any commercial party related directly or indirectly to this article.

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