KONAČAN ISHOD U LEČENJU LEG-KALVE-PERTESOVE BOLESTI KORISTEĆI KOMBINACIJU ILIZAROVOG METODA I TUNELIZACIJE – PRIKAZ SLUČAJA

PRIKAZ SLUČAJA

CASE REPORT

THE FINAL OUTCOME IN THE TREATMENT OF LEGG-CALVE-PERTHES DISEASE USING THE COMBINATION OF ILIZAROV METHOD OF TREATMENT AND TUNELISATION (CASE REPORT)

Branislav Krivokapić[1,2], Bojan Bukva[2,3], Danilo Jeremić[1,2], Nikola Bogosavljević[1,2], Slavko Tomić [1,2]

[1] Institut za ortopediju „Banjica", Beograd, Srbija

[2] Univerzitet u Beogradu, Medicinski fakultet, Beograd, Srbija

[3] Univerzitetska dečja klinika „Tiršova", Beograd, Srbija

[1] *Institute for Orthopedic Surgery "Banjica", Belgrade, Serbia*

[2] *Medical Faculty, University of Belgrade, Belgrade, Serbia*

[3] *University Children`s Hospital, Belgrade, Serbia*

SAŽETAK

Uvod: Leg-Kalve-Pertesova bolest (LKPB) je relativno česta bolest kuka dečjeg doba, koja pogađa otprilike 0,8 slučajeva na 100.000 dece. Obično se javlja kod dece uzrasta između 4 i 8 godina i kod dečaka je prisutnija više nego kod devojčica. Primarno je zahvaćen jedan kuk, ali bilateralna lokalizacija je prisutna u 9-15 slučajeva. Primarni tretman u Leg-Kalve-Pertesovoj bolesti je koncept fokusiran na očuvanju glave butne kosti u acetabulumu, čime se sprečava omekšavanje i fragmentacija. Najčešći metod hirurškog tretmana sastoji se od butne ili pelvične osteotomije ili kombinacije oba. Ovaj rad ima za cilj da predstavi minimalno invazivnu alternativnu metodu za lečenje Leg-Kalve-Pertesove bolesti.

Prikaz slučaja: Predstavljamo izveštaj o slučaju 19-godišnjaka sa dugom istorijom bolesti. Njegovo lečenje počelo je u petoj godini života, i ovo je najnoviji izveštaj njegovog stanja 14 godina nakon operacije.

Zaključak: Artrodijastaza kuka korišćenjem Ilizarovljevog cirkularnog okvira u kombinaciji sa tunelizacijom glave i vrata butne kosti je minimalno invazivni hirurški zahvat. Sa tehničke tačke gledišta, ovo je relativno zahtevna procedura koja rezultira kraćim periodom hospitalizacije, smanjenim rizikom od komplikacija i troškovima, u poređenju sa karličnom i butnom osteotomijom.

Ključne reči: Leg-Kalve-Pertesova bolest, karlične kosti, Ilizarov tehnika, ishod lečenja

ABSTRACT

Introduction: Legg-Calve-Perthes disease (LCPD) is a common childhood hip disease, affecting approximately 0.8 in 100,000 children. It usually affects children between 4 and 8 years old, and boys are more affected than girls. One hip is initially affected, but bilateral localization is present in 9-15 of the cases. The primary treatment in LCPD focuses on containing the femoral head in the acetabulum, thus preventing mollification and fragmentation. The most common surgical treatment method consists of either femoral or pelvic osteotomy or a combination of both. This paper aims to present a minimally invasive alternative method for treating LCPD.

Case report: We are presenting a case report of a 19-year-old with a lengthy history of illness. His treatment started at the age of 5, and this is an update on his condition 14 years after the surgery.

Conclusion: Hip arthrodiastasis using the Ilizarov circular frame combined with femoral head and neck tunneling is a minimally invasive surgical procedure. From a technical standpoint, this is a relatively demanding procedure that results in shorter in-patient treatment, reduced risk of complications, and cost compared to pelvic and femoral osteotomy.

Keywords: Legg-Calve-Perthes disease, pelvic bones, Ilizarov technique, treatment outcome

Autor za korespondenciju:
Branislav Krivokapić
Institut za ortopediju „Banjica"
Mihajla Avramovića 28, 11000 Beograd, Srbija
Elektronska adresa: branislav.krivokapic@iohbb.edu.rs

Corresponding author:
Branislav Krivokapić
Institute for Orthopedic Surgery "Banjica"
28 Mihajla Avramovića Street, 11000 Belgrade, Serbia
E-mail: branislav.krivokapic@iohbb.edu.rs

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UVOD

Leg-Kalve-Pertersova bolest (LKPB) je relativno česta bolest kuka dečjeg doba, koja pogađa otprilike 0,8 slučajeva na 100.000 dece [1]. Bolest su 1910. godine prvi put opisali tri naučnika iz tri različite zemlje, ali su njena etiologija i načini lečenja još uvek predmet rasprave [2]. Obično se javlja kod dece uzrasta od 4 do 8 godina i češće se javlja kod dečaka nego kod devojčica [1,2]. Primarno je zahvaćen jedan kuk, ali bilateralna lokalizacija je prisutna u 9-15 slučajeva [1,2].

Leg-Kalve-Pertesova bolest je avaskularno stanje koje utiče na epifizu kapitela budne kosti (glava) [1,3]. Avaskularno stanje je praćeno usporavanjem rasta jezgra kostiju. Kostno jezgro se naknadno resorbuje, zamenjuje novom i preuređuje u skladu sa mehaničkim svojstvima. Glava butne kosti postaje sravnjena i uvećana nakon preuređenja. Proksimalni butni deo i metafiza takođe mogu biti zahvaćeni, što može dovesti do sekundarnih promena u acetabulumu. Tretman LKPB-a i dalje ostaje kontroverzan uprkos opsežnoj literaturi o ovoj temi [3].

Princip LKPB tretmana je koncept zadržanja glave butne kosti u acetabulumu, kao zaštite od omekšane i fragmentovane glave butne kosti [4,5]. Koncept suzbijanja se može postići korišćenjem konzervativne metode (kao što su odmor ili ortoza) ili hirurške metode lečenja. Ovaj princip lečenja u ranoj fazi LKPB-a u uzrastu mlađih od 8 godina je široko prihvaćen.

Lečenje kasnog LKPB-a (preko 8 godina) je još uvek izazovno. Lečenje kasnog LKPB-a butnom ili karličnom osteotomijom ili kombinacijom oba, imao je loš konačni ishod [6-9].

Mnogi autori preporučuju koncept distrakcije kuka (artrodijastaza) kao početni metod lečenja [10-16].

Artrodijastaza nema uticaja na anatomske promene u zglobu kuka. Smanjuje opterećenje zglobne površine kuka i podspešuje zarastanje glave butne kosti [17]. Takođe, može se kombinovati sa tuneliranjem vrata i glave butne kosti kako bi se poboljšalo izlečenje [13].

Ova studija ima za cilj da predstavi slučaj minimalno invazivne alternativne metode u lečenju LKPB-a kod 5-godišnjeg dečaka i praćenje početne procedure 14 godina kasnije.

PRIKAZ SLUČAJA

Predstavićemo izveštaj o slučaju 5-godišnjeg dečaka sa dugom LKPB istorijom. Žalio se na bolove u levom kuku i levom kolenu. Takođe je šepao na levu nogu. Nije imao opšte znake infekcije, noćne bolove ili prateće bolesti. U kliničkim nalazima, imao je ograničenje abdukcije (30 stepeni) i unutrašnje rotacije (5 stepeni) u levom kuku. Takođe, imao je pozitivan Trendelenburgov znak sa leve strane.

INTRODUCTION

Legg-Calve-Perthes disease (LCPD) is a common childhood hip disease, affecting approximately 0.8 in 100,000 children [1]. The disease was first described in 1910 by three scientists from three different countries, but its etiology and treatment modalities are still a matter of debate [2]. It usually occurs in children aged 4 to 8 years, and it is more common in boys than girls [1,2]. One hip is initially affected, but bilateral localization is present in 9-15 cases [1,2].

Legg-Calve-Perthes disease is an avascular condition that affects the capital femoral epiphysis (head) [1,3]. The avascular condition is followed by growth retardation of the ossific nucleus. The ossific nucleus is subsequently resorbed, replaced by a new bone, and remodeled according to mechanical properties. After remodeling the femoral head becomes flattened and enlarged. The proximal femoral physis and metaphysis could also be affected, leading to secondary acetabulum changes. The treatment of LSPD remains controversial despite the extensive literature on this topic [3].

The principle of LCPD treatment is the concept of containment of the femoral head in acetabulum, as a protection against a mollified and fragmented femoral head [4,5]. The containment concept could be attained using conservative methods (like rest or orthosis) or surgical treatment. This principle of treatment in the early stages of LCPD at the age of under 8 is widely accepted.

The treatment of late LCPD (over the age of 8) is still challenging. Late LCPD treated by femoral or pelvic osteotomy, or a combination of both has had poor outcomes [6-9].

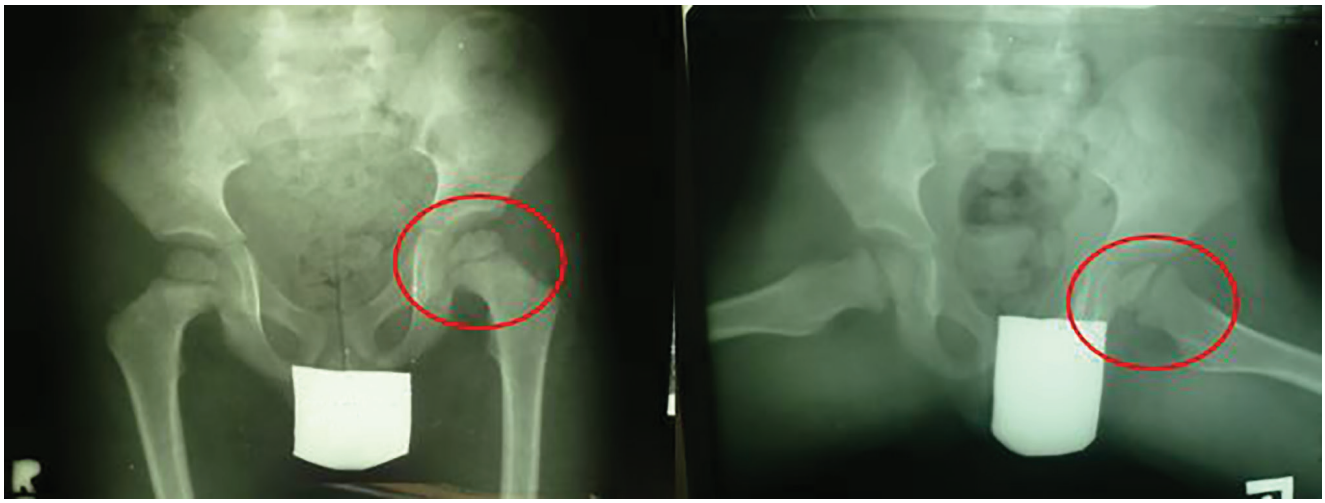
Many authors recommend the concept of hip distraction (arthrodiastasis) as the initial treatment method [10-16].

Arthrodiastasis does not have an impact on anatomic changes in the hip joint. It decreases the stress on the hip articular surface and enhances the healing of the femoral head [17]. It could also be combined with the femoral neck and head tunneling to improve healing [13].

This study aims to present a case of a minimally invasive alternative method in treating LCPD in a 5-year-old boy and a follow-up on the initial procedure 14 years later.

CASE REPORT

We will present the case report on a 5-year-old boy with long LCPD history. He complained of pain in the left hip and left knee. He had also been limping on the left leg. He had no general signs of infection, night pain, or concomitant diseases. In the clinical findings,



Slika 1. Inicijalni rendgenski snimak levog kuka u AP i u „žabljem" položaju Pertesove bolesti.

Figure 1. Initial X-ray in AP and in "frog-like" view of left hip Perthes disease

Nakon analize krvi i rendgenskih nalaza (u AP i „žabljem" položaju) LKPB je potvrđen. Prema Katarel i Hering klasifikaciji, on je klasifikovan je kao Katarel 3 i Hering B grupa.

Nakon prijema, počeli smo sa konzervativnim tretmanom koristeći perkutanu trakciju u blagoj abdukciji na nedelju dana (otprilike 10% ukupne težine pacijenta). Nakon vuče usledio je odmor bez opterećenja još nedelju dana. Nakon toga, pristupili smo protokolu zatezanja Atlanta ortozom, u položaju blage abdukcije i unutrašnje rotacije sa punim opterećenjem na obolelu nogu. Položaj glave butne kosti (suzbijanje) potvrđen je rendgenskim snimkom (u AP prikazu). Posle 4 meseca konzervativnog lečenja, ishod je bio loš i hirurško konsultantsko telo (troje specijalista ortopedske hirurgije) odlučilo je da obavi hirurški tretman.

Roditeljima su predstavljeni svi modaliteti hirurškog lečenja i oni su odobrili ovaj retko korišćeni hirurški tretman. Ova metoda je izvedena kao kombinacija artrodijastaze levog kuka pomoću Ilizarov okvira (kružni spoljni fiksator) i tuneliranja glave i vrata butne kosti korišćenjem Kiršnerovih žica od 2,0 mm (K žice).

Ova metoda lečenja je minimalno invazivna hirurška metoda. Glavni cilj artrodijastaze pomoću Ilizarovskog kružnog fiksatora je odvlačenje pažnje sa zgloba kuka i smanjenje pritiska na fragmentovanu glavu butne kosti.

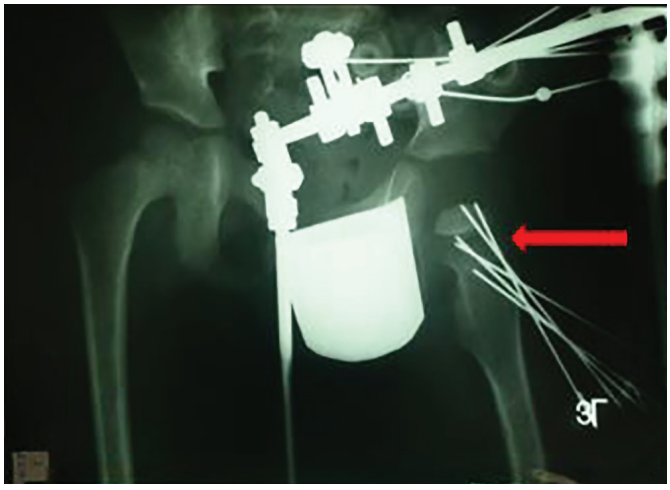
S druge strane, glavni cilj tuneliranja pomoću K žica je povećanje dotoka krvi u vrat i glavu butne kosti (Slika 2). Proces izlečenja glave butne kosti biće poboljšan u ishodu.

Tokom hirurškog zahvata i postoperativno nije došlo do komplikacija. Dva dana posle operacije pacijent je počeo sa fizikalnom terapijom, pasivnom kinezioterapijom sa delimičnim nošenjem težine na levoj nozi koristeći štake. Fizikalna terapija je bila obezbeđena u

he had the limitation of abduction (30 degrees) and internal rotation (5 degrees) in the left hip. He also had a left-side positive Trendelenburg sign.

Legg-Calve-Perthes disease was confirmed after blood analysis and X-ray findings (in AP and 'frog-like' position). According to Catarell and Herring classification he was classified as Catarell 3 and Herring B group.

After admission, we started with conservative treatment using percutaneous traction in slight abduction for one week (approximately 10% of the patient's total weight). The traction was followed by non-weight-bearing rest for one week more. After that, we started the bracing protocol using Atlanta orthosis, in the position of a slight abduction and internal rotation with full-weight bearing on the affected leg. The posi-



Slika 2. Intraoperativni rendgenski snimak u AP položaju (operativni postupak: Aplicatio aparatus pro distractio fixatione ad pelvis et femoris sinistri sec. Ilizarov. Transfixatio capitis femoris sinistri cum filli Kirshneri No V)

Figure 2. Intraoperative X-ray in AP position (Operative procedure: Aplicatio aparatus pro distractio fixatione ad pelvis et femoris sinistri sec. Ilizarov. Transfixatio capitis femoris sinistri cum filli Kirshneri No V)

Slika 3. Rendgen u AP položaju nakon uklanjanja K-žica sa glave i vrata butne kosti.

Figure 3. X-ray in AP position after K-wires removal from femoral head and neck

Slika 4. Rendgen u AP položaju nakon uklanjanja Ilizarovog cirkularnog okvira i K žica

Figure 4. X-ray in AP position after Ilizarov circular frame and K wires removal

bolnici u trajanju od dva meseca. Šest nedelja nakon operacije K žice su uklonjene sa vrata i glave butne kosti. Tri meseca posle operacije pacijent je primljen na ponovnu operaciju kada je izvađen Ilizarov kružni fiksator. Hirurški ishod je prikazan na slici 4.

Posle drugog hirurškog zahvata (uklanjanje Ilizarovljevog okvira) u bolnici je obezbeđen kratak tretman fizikalne terapije sa punim težinom; tokom prve dve nedelje oslonac je bio na štakama, a zatim sa nošenjem pune težine.

Pacijent je kontrolisan u intervalima od 6 nedelja, tri meseca i šest meseci posle druge operacije. Na slici 5 predstavljeni su 6-mesečni i 18-mesečni rendgenski nalazi posle druge operacije (uklanjanje hardvera).

U kliničkom ishodu, 18 meseci nakon uklanjanja hardvera pacijent je imao pun spektar kretanja u levom kuku i levom kolenu, bezbolno hodajući bez ikakve

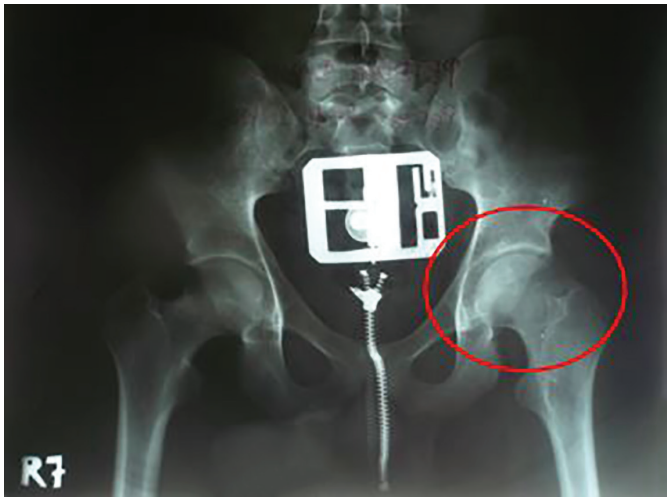
tion of the femoral head (containment) was confirmed by the X-ray (in AP view). After 4 months of conservative treatment, the outcome was poor and the surgical consulting body (3 specialists in orthopedic surgery) decided to perform a surgical treatment.

All modalities of the surgical treatment were presented to the parents, and they approved of this rarely used surgical treatment. This method was performed as a combination of left hip arthrodiastasis using the Ilizarov frame (circular external fixator) and the femoral head and neck tunneling using 2.0 mm Kirschner wires (K wires).

This method of treatment is a minimally invasive surgical method. The main goal of arthrodiastasis using the Ilizarov circular fixator is to distract the hip joint

Slika 5. Rendgen u AP položaju 6 meseci (A) i 18 meseci (B) nakon uklanjanja hardvera

Figure 5. X-ray in AP position 6 months (A) and 18 months (B) after hardware removal



Slika 2. Rendgen u AP položaju 14 godina nakon prve operacije

Figure 6. X-ray in AP position 14 years after the first surgery

podrške i obavljajući normalne fizičke aktivnosti osim skakanja.

Poslednje praćenje urađeno je kada je pacijent napunio 19 godina (14 godina posle prve operacije). Rendgenski snimak u AP položaju predstavio je kongruentnost glave butne kosti bez ikakvih znakova avaskularne nekroze. Estetski razlozi su takođe bili od suštinskog značaja; butni i ilijački ožiljci su bili skoro nevidljivi.

DISKUSIJA

Modaliteti lečenja LKPB-a su različiti, bilo konzervativni ili hirurški. Pacijenti mlađi od 6 godina imaju bolje ishode i prognoze uprkos metodi lečenja za razliku od pacijenata starijih od 6 godina [15,18]. Konzervativne metode lečenja (fizikalna terapija i upotreba ortoze) imaju mnoge prednosti kao neinvazivne metode: nema formiranja ožiljaka, nema rizika od infekcije i nema psiholoških efekata na dete i roditelje. Sa druge strane, potrebno je duže lečenje i dobra usaglašenost između lekara, deteta i roditelja. Da zaključimo, svi ovi faktori se moraju razmotri u donošenju konačne odluke [19].

Najvažniji faktori u LKPB lečenju su starosni rendgenski nalaz pacijenta u AP-u i pozicija „nalik žabi" (prema katarelovoj i Heringovoj klasifikaciji).

Pacijenti starosti preko 6 godina imaju bolje ishode koristeći hirurški pristup u LKPB lečenju [16]. Prema njihovim studijama, neki autori su zaključili da je rengenski nalaz glavni faktor u prognozi konačnog ishoda LKPB-a, odnosno nivo aseptičke narkoze glave butne kosti [20,21].

Pokazalo se da bušenje tunela utiče na manje od 10% ukupne površine hrskavice rasta, ne narušava rast ploče i ne smanjuje rast u predelu kuka [22]. Nekoliko autora lečilo je pacijente sa artrodijastazom koristeći monolateralni fiksator; međutim, koristili su biomeha-

and to reduce the pressure on the fragmented femoral head. On the other hand, the main goal of tunneling using K wires is to increase the blood supply to the femoral neck and head (Figure 2). The healing process of the femoral head will be improved in the outcome.

No complications occurred during the surgical procedure and postoperatively. Two days after the surgery, the patient started with physical therapy, passive kinesiotherapy with partial weight bearing on the left leg using crutches. All physical therapy treatments were provided in the hospital during the two months. Six weeks after the surgery K wires were removed from the femoral neck and head. Three months after the surgery, the patient was admitted for a redo-surgery procedure when the Ilizarov circular fixator was extracted. The surgical outcome is presented in Figure 4.

After the second surgical procedure (the Ilizarov frame removal), a short physical therapy treatment with full-weight bearing was provided during the stay; at first, it was supported with crutches during the first two weeks and afterward with full-weight bearing.

The patient was controlled at 6 weeks, three-month, and six-month intervals after the second surgery. Figure 5 presents 6-month and 18-month X-ray findings after the second surgery (hardware removal).

In the clinical outcome, 18 months after the hardware removal the patient had the full range of motion in the left hip and left knee, painlessly walked without any support, and carried out normal physical activities besides jumping.

The last follow-up was done when the patient turned 19 (14 years after the first surgery). The X-ray in the AP position presented the congruency of the femoral head without any signs of avascular necrosis. Aesthetic reasons were also essential; the femoral and iliac scars were almost invisible.

DISCUSSION

The treatment modalities of LCPD are various, whether conservative or surgical. Patients under the age of 6 have better outcomes and prognoses despite the method of treatment than patients over the age of 6 [15,18]. Conservative methods of treatment (physical therapy and orthotics usage) have many advantages as non-invasive methods: no scar formation, no infection risk, and no psychological effects on a child and parents. It takes, on the other hand, longer treatment and good compliance between a doctor, a child, and parents. To conclude, all these factors have to be considered in making a final decision [19].

The most important factors in LCPD treatment are the patient's age X-ray findings in AP and the "frog-like" position (according to Catarell and Herring classification).

nički slabiju metodu fiksatora i igle velikog prečnika za tuneliranje i njihovi pacijenti nisu nosili težinu tokom cele dužine lečenja [23,24]. Kočaoglu je predstavio i objavio rezultate upotrebe Ilizarovljevog aparata kod Pertesove bolesti. Koristio je snažan i složen Ilizarov okvir u ranoj fazi fragmentacije glave butne kosti, a njegova stopa uspeha nije bila impresivna. Da bi se održalo zadržavanje glave butne kosti neophodno je započeti tretman u fazi nekroze ili ranog stadijuma fragmentacije, odnosno pre kolapsa epifize butne kosti [25]. Uprkos pregledu literature o zlatnom standardu u hirurškom pristupu (osteotomiji karlice poput Saltera, Kjarija ili trostruke osteotomija karlice), artrodijazu kuka sa tuneliranjem treba smatrati alternativnom hirurškom tehnikom. Artrodijastaza kuka i tuneliranje glave i vrata butne kosti treba razmotriti u ranoj fazi LKPB-a i kasnom početku bolesti kada standarne hirurške procedure ne daju dobre ishode [26].

Primenom ove tehnike dobija se pun opseg bezbolnih pokreta, bez sekundarnih poremećaja ili smetnji u hodu. Rizik od lateralne subluksacije glave butne kosti smanjen je na rendgenskim snimcima, postignuta je kongruencija glave butne kosti bez anatomskih povreda u zglobu kuka. Takođe, ova tehnika se može primeniti kod starije dece i adolescenata sa lošim prognozama (prema rendgenskim nalazima) i u kontraindikacijama za osteotomiju karlice.

Ovaj izveštaj LKPB slučaja predstavlja najvažnije vrednosti u konačnom ishodu: starost, ocenu prema rendgenskim nalazima (prema Heringu i Katarelu) i adekvatnu strategiju u pristupu lečenja.

Odabrali smo manje invazivnu i retko korišćenu tehniku: artrodijastu kuka korišćenjem Ilizarov cirkularnog okvira u kombinaciji sa tuneliranjem vrata i glave butne kosti. To je minimalno invazivna hirurška procedura i tehnički manje zahtevna: kraće lečenje pacijenta, manji rizik od komplikacija i niži troškovi u poređenju sa osteotomijom karlice i butne kosti. Ova tehnika bi mogla pomoći u lečenju manje neusaglašenosti nogu, kao jednog od kliničkih i radioloških znakova u evoluciji Pertesove bolesti.

Želimo da naglasimo da artrodijastazu sa tuneliranjem, kao minimalno invazivnu operaciju, treba smatrati jednom od prvih opcija lečenja u ranoj fazi LKPB-a kada glava butne kosti još uvek nema nikakve strukturne promene i nakon neuspelog konzervativnog tretmana.

IZJAVE ZAHVALNOSTI

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Sukob interesa: Nije prijavljen.

Patients over the age of 6 have better outcomes using the surgical approach in LCPD treatment [16]. According to their studies, some authors concluded that the major factor in LCPD prognosis of the outcome is an X-ray finding, precisely the level of aseptic narcosis of the femoral head [20,21].

It has been shown that drilling a tunnel affects less than 10% of the total area of the growth cartilage, it does not impair the growth plate and it does not reduce growth in the hip region [22]. Several authors treated patients with arthrodiastasis using monolateral fixator; however, they used a biomechanically weaker fixator method, and large diameter pins for tunneling and their patients did not weight bear during the whole length of treatment [23,24]. Kocaoglu presented and published results of the Ilizarov apparatus usage in Perthes disease. He used a robust and complex Ilizarov frame at the early stage of the femoral head fragmentation, and his success rate was not very impressive. To maintain the containment of the femoral head it is mandatory to initiate the treatment during the stage of necrosis or early fragmentation stage, thus before the collapse of the femoral epiphysis [25]. Despite the literature review on the gold standard in surgical approach (pelvic osteotomy like Salter, Chiari, or triple pelvic osteotomy), hip arthrodiastasis with tunneling should be considered as an alternative surgical technique. Hip arthrodiastasis and femoral head and neck tunneling should be considered at an early stage of LCPD and the late onset of disease when standard surgical procedures do not give great outcomes [26].

The usage of this technique has gained a full range of painless motion, without secondary disorders or gait disturbances. The risk of lateral subluxation of the femoral head was decreased in X-ray findings, the femoral head congruency was reached without anatomic violation in the hip joint. This technique might also be used in older children and adolescents with poor prognosis (according to X-ray findings) and in contraindications to do pelvic osteotomy.

This LCPD case report presents the most important values in the outcome: age, rating according to X-ray findings (according to Herring and Catarell), and adequate strategy in the treatment approach.

We chose the less invasive and rarely used technique: hip arthrodiastasis using the Ilizarov circular frame in combination with femoral neck and head tunneling. It is a minimally invasive surgery procedure and technically less demanding: shorter patient–in treatment, minor risk of complications, and lower costs in comparison to pelvic and femoral osteotomy. This technique might help treat a minor leg discrepancy, as one of the clinical and radiological signs in the evolution of Perthes disease.

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We want to emphasize that arthrodiastasis with tunneling, as a minimally invasive surgery should be considered as one of the first treatment options at an early stage of LCPD when the femoral head still does not have any structural changes and conservative treatment failed.

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