ORIGINAL ARTICLE

The Earliest Age Limit for Manipulation and Casting to Avoid Surgical Intervention in Idiopathic Congenital Knee Dislocation

Anisuddin Bhatti, Kiran Maqsood, Maqsood Hassan Shaikh, Pervez Ali, Zulfiqar Ali, Muhammad Yousuf Bhatti

ABSTRACT

Objective: To know the earliest age limit when surgical intervention can be avoided in Congenital Dislocated Knee.

Methods: This Prospective study was carried at Jinnah Postgraduate Medical Centre Karachi, Pakistan from January 2013 to December 2016.The patients with Idiopathic, Type II & III Congenital Knee Dislocations (CDK), in newborn to three months were managed by serial manipulation and Casting with and without per Cutaneous Quadriceps Tenotomy (PCQT).

Results: At 26 months median follow-up, out of 32 knees of 23 patients, 29(90.62%) behaved Excellent and 2(6.25%) behaved good. Average 4 casts required, to achieve >120^0 flexion, stable knees with negative Drawers test in 96.87% Knees. The required number of casts increased from 3 to 5 as age increased from one day to one month & above, moreover 7 knees of 3 patients, who got treatment initiated after 3 weeks of birth also required PQCT.

Conclusion: The CDK is easy to reduce without significant manipulation time, when treatment is started within 24 hours of birth. With increasing age from hours to four weeks, the manipulation takes more time to reduce and required more number of cast to maintain reduction. After three weeks of age, CDK often require an addition procedure of PCQT.

Key words: Casting, Congenital, Dislocation, Knee, Manipulations, Per Cutaneous Tenotomies.

This article may be cited as:

INTRODUCTION

The Idiopathic, Congenital Dislocated Knee (CDK) is a rare congenital disorder, that occur 1 in 100,000 live births, comparatively 1% of Developmental Dysplasia of the Hip (DDH) [1-3]. Both intrinsic [3,4] and extrinsic factors [1-3] has been attributed to its pathogenesis. One of the good evidence for familial, genetic origin have been reported with a mother and her three children from three different fathers, all had CDK [5]. Whereas; non-genetic dysplasia of sporadic occurrence, has also been reported [2,5-8]. CDK is easily recognizable at birth, it is classified in three types / grades, based on Clinico-radiological severity of dislocation [9,10].

Department of Orthopaedic, Trauma & Reconstructive Surgery, Jinnah Postgraduate Medical Centre, Rafiqui Shaheed Road, Karachi 75510, Pakistan
Correspondence: Anisuddin Bhatti
Email: dranisbhatti@gmail.com

Various Non-operative manipulative techniques including methods of additional Percutanous Quadriceps Tenotomies (PCQT) and maintenance casts / splints have been used to achieve satisfactory results in babies aged less than three months [7,11,15]. The methods of PCQT includes: Percutanous Quadriceps Recession (PQR) [10,16], Percutanous Quadriceps Needle Tenotomy (PCQNT) [14] and Mini Open Quadriceps tenotomy (MOQT) [17]. Whereas; few investigators [10,17] suggest, open reduction with Quadriceps lengthening V-Y Plasty (QVYP) [2], in age group over 1 months, Grade III, more rigid CDKs and failed PCQTs.The best results however, have been reported with earliest treatment initiated within hours to days after birth, as the delayed treatment may complicate with poor outcome, premature physeal fusion and flattening of condyles [1,7,8]. Haga [18] on other side, suggest to wait for 1 month as spontaneous reduction of CDK may occur in isolated CDK without associated with Clubfoot, Arthrogryposis or Larsen's syndrome.

There seems to be a significant consensus on open reduction of CDK in age over three months, however; the earliest age for conservative treatment with or without minimal surgical intervention is yet debatable. This report shares an experience and signifies how earliest the conservative treatment provides best result and avoids surgical intervention including simple PCQT.

METHOD

This Prospective, serial case study was carried out at Jinnah Postgraduate Medical Centre (JPMC). The patients included were managed by corresponding author only, at Paediatric Orthopaedic clinics at Jinnah Postgraduate Medical Centre Karachi, Pakistan. The study duration includes from 1stJanuary 2013 to 30thDecember 2016 with onward follow up to 30th June 2017.

The patients included were having Idiopathic CDK Type II & III, in age range of newborn to three months. The patient with type I CDK, age over three months and associated with Teratologic, Paralytic, Infective and arthrogrypotic features were excluded from the study.

All the patients were managed by serial Manipulation and Casting method of J.Y. Ko (1999) cited by Chun Chien Cheng & JihYangKo [7]. The steps include, gradual, sustained traction by pulling the leg in the direction of femur, for 5 minutes or more and gentle manipulation of Tibial condyles down over Femur and Femur pushed upward to realign normal Tibio-Femoral relationship [7]. Once realignment and easy knee flexion achieved, the leg was immobilized in cast. The cast was changed with fortnightly regimen at Sukkur clinic and with weekly regimen at Karachi clinics. The Tibio-Femoral realignment was confirmed clinically by palpation of femoral condyles in popliteal fossa on each visit and on radiographic examination, usually after 2-3 cast. The manipulation and casting continued until knee flexion was achieved to >120^0. The last cast in all patients was maintained for two weeks duration; to decrease the swelling, irritability and pain free post cast mobilization. Following each cast the circulation in Toes were checked with blanching and refilling within 2 seconds that was demonstrated to parents as well, with directions to check every 6 hours and inform for any unexpected squeale or complications. In rigid Knees an additional PCQT of Quadriceps was made after 4th- 5thserial casts. Tenotomy performed by a single stab from lateral side, just under Quadriceps tendon made tense by holding knee in forced flexion, with a surgical blade no.11, under local anaesthetic infiltration. In the older age patient with bulky thighs and with associated DDH cases a Pavlik Harness was used for additional 6-8 weeks. The parents were taught, at home exercises during and after weaning period.

The followup protocol includes clinic-radiological assessment, every fortnightly for 2 months, then every month for six months and thereafter every 3 months until last follow up in June 2017. The Modified Scoring system[10] for knee function described by Ferris and Aichron [9], was adopted to evaluate the results at each quarterly visit. The study was carried out with prior approval from the Institutional Review Board JPMC, NCCI (NMI) Hospital and Bhatti Hospital Sukkur. An Informed written consent was taken from parents of each patient before embarking to treatment.

Statistical Analysis

A database was developed on SPSS Version 19. The descriptive statistics of age (months) were represented by their range and median values due to their extreme values. The categorical data were presented by their frequencies along with percentages. Association of outcome with gender, side of knees, idiopathic types, number of casts and duration of cast were found by using Chi-square test of dependency along with probability of significance.

RESULTS

The pertinent demographic data and outcomes of individual patients are given in table- 1-2. There were 8(34.8%) males and 15(65.2%) Females, with Ratio of 2:1. The age ranges from 1 to 150 days with median age of 18.5 days. There were 09 (39.1%) bilateral knees, 08 (34.8%) left knees and 06 (26.1%) right knees. Overall 32 knees, 15 (46.9%) Right Knees and 17 53.1% left knees were manipulated and casted for the purpose of study. Only 02 (6.2%) were of Idiopathic Type II and remaining 30 (93.8%) were of Idiopathic Type III (Table:1). Two patients had associated bilateral DDH (Tonnis level II), other one had associated Clubfoot and Knee flexion contracture of 45^0 on other side, whereas; one other patient also had Knee flexion contracture of 45^0.

Number of Casts ranges from 3 to 9 (Table:2). Out of 32 knees, 21 (65.62%) who achieved excellent to good results required 04 Casts, 05 (15.62%) who achieved excellent results required 03 casts. The among remaining 5 (15.6%), 04 knees who behaved

good required 05 cast and 01 knee failed to achieve desirable results even with PCQT and was subsequently got open reduction with V-Y plasty. The remaining one patient with bilateral knees, who initially got 5 cast somewhere else, was further managed with PCQT and 4 more casts (total 09 casts) to achieve excellent results.

Table 1: Descriptive Statistics of 32 Congenital Knee Dislocations in New borns.

Number of Patients	23
Gender of Patients	
Male	08 (34.8 %)
Female	15 (65.2 %)
Age of Patients (Days)	
Median Age (Days)	18.5
Age Range (Days)	1 – 150
Side of Knees	
Right Knees	06 (26.1 %)
Left Knees	08 (34.8 %)
Bilateral Knees	09 (39.1 %)
Number of Knees	32
Right Knees	15 (46.9 %)
Left Knees	17 (53.1 %)
Types of Knees	
Type-II	02 (6.2 %)
Type-III	30 (93.8 %)
Number of Casts among 32 knees	
Three Casts	05 (15.6 %)
Four Casts	21 (65.6 %)
Five Casts	05 (15.6 %)
Nine Casts	01 (3.1 %)
Median casts	04

Reference to over al results (Table:1-2) at median follow up duration were of 26 months, 29 (90.7%) out of 32 knees achieved Excellent, 02 (6.2%) knees were of Good outcome and only 1 (3.1%) knee had Poor outcome. The majority (81.25%) of excellent to good behaved knees, required 3-4 cast, minimal duration of manipulations and no mini-surgical intervention (Table:2).

There were no association found between Gender, Age, side, idiopathic types, number of casts with the outcome ((Table-2). The cast duration had no association with outcome of knee as well (Chi-square=0.219, p=0.12). However; significant association was noticed between age at initiation of treatment with duration of manipulation and number cast.

The patients who got their first cast on the day of birth (Group I) required 3 casts, the patients who seeks treatment within 15 days after birth (group II) required 4 casts, patients who were casted within 16 days to 28 weeks (Group III) required 4 casts, while patients in which treatment was commenced after 29 days (Group IV) required 5 casts except two, who required 4 casts with soft reduction. The group I patients required gentle traction and manipulation for 3-5 minutes to achieve 90^0 flexion on first visit and second cast to achieve $>120^0$ knees flexion. Compared to group I, another groups II-IV required more than 5-7 minutes & up to 5 cast. Whereas; one patient from group II required over 15 minutes stretching and manipulations to bring Tibio-Femoral condyles in alignment. Five patients (7 Knees) from group III & IV, required additional PCQT. While one of them who initiated treatment after four weeks of birth (groupIV) failed to achieve satisfactory reduction, was later operated with QVYP.

All four patients with associated DDH, Club foot and other side Knee flexion deformity resulted to e excellent outcome with simultaneous casting and follow-up Splints [19]. Two patients developed plastic deformation [1,17,18] (anterior Bow) of tibia that resolved spontaneously over next 6-8 weeks without further intervention. A single patient of 5 months age was included in study, as she had treatment initiated at the age of 1 month, the surgeon could not achieve satisfactory reduction even after 9 casts, on seeking second opinion we found, femoral condyles palpable in popliteal fossa, despite having 10^o knee flexion. Her X-ray revealed Type III CDK, she responded well to serial casting initiated with PCQT.

Table 2: Association of Outcome with independent variables

	Outcome			
Variables	**Excellent n=29 (90.7%)**	**Good n=2 (6.2%)**	**Poor n=1 (3.1%)**	**Total n=32 (100 %)**
Gender				

Male	9 (90.0%)	0 (0%)	1 (10.0 %)	10 (100%)
Female	20 (91.0 %)	2 (9.0%)	0 (0%)	22 (100%)
Median age (days)	15	21	90	18
Side of Congenital Knee Dislocation				
Right knee	14 (93.3%)	1 (6.7%)	0 (0%)	15 (100%)
Left knee	15 (88.2%)	1 (5.9%)	1 (5.9%)	17 (100%)
Types of Congenital Knee Dislocation				
Type-II	2 (100%).0	0 (0.0%)	0 (0.0%)	2 (100%)
Type-III	27 (90.0%)	2 (6.7%)	1 (3.3%)	30(100%)
Number of Casts				
3 Casts	5 (100.0%)	0 (0.0%)	0 (0.0%)	5 (100%)
4 Casts	19 (90.5%)	2 (9.5%)	0 (0.0%)	21 (100%)
5 Casts	4(80.0%)	0 (0.0%)	1 (20.0%)	5 (100%)
9 Casts	1 (100.0%)	0 (0.0%)	0 (0.0%)	0 (100%)
PCT	4 (57.1%)	2 (28.6%)	1 (14.3%)	7 (100%)
Harness	9 (75.0%)	2 (16.7%)	1 (8.3%)	12 (100%)
Follow-up Duration (Months)	27	25	14	26
Cast Duration				
Weekly	15(93.8%)	0 (0.0%)	1 (6.2%)	16 (100%)
Fortnightly	14 (87.5%)	2 (12.5%)	0 (0)%)	16 (100%)

Fig. 1: Case 19 (A) The characteristic appearance of bilateral CDK at birth. (B) Radiograph before reduction demonstrating anterior subluxation in the bilateral knees. (C) The closed reduction starts with gentle, persistent manual traction. (D) During traction, an anteriorly directed force is applied to the distal femur, and a posteriorly directed force to the proximal tibia. (F) Follow-up radiograph showing normal alignment of both knees at 9 months of age.

Figure 1: Courtesy: Reference no: 7
Chun-Chen et al, Chang Gung Med j Vol 33 No.3, 2010.

DISCUSSION

The early recognition and non-operative treatment of Congenital Dislocated Knee (CDK), has been of paramount importance to achieve satisfactory functional results with anatomical reduction and well contoured condyles [10,19,20]. Though mainstay of treatment with closed serial manipulation and casting is to initiate soon after birth [7,17,19,21], the best results however, could only be achieved when treatment is initiated within 8 hours of birth [7]. The current study & others reports [7,10,12] find significant difficulties to achieve absolute reduction as the age increases from hours to days and thereafter. The difficulties include increased duration of traction / manipulation from 5 t0 15 minutes, more number of casts, PCQT in age over 28 days. In age group over 1 month, we find simple PCQT [releasing only Rectus Femoris tendon & fascia, without dividing peripheral retinaculum] under local anaesthesia, easiest and fruitful techniques. This also reduces risk of complications associated with General Anaesthesia (GA) used in PCQR [10,16,23], PCQNT [14] & MOQT [17], QVYP [2,13,20]. The complications include risk of anaesthesia, hospitalization cost, and a great source of anxiety for parents [7,8,10,11,17].

Compared to initiation of CDK treatment within day of birth and thereafter, we found comparable results to earlier reported in multiple studies, that ranged in 95.65%-100% successes [4,7,11,14,15,21,23]. However; the success rate decline from 95% to 88%, as the age increases from 2 weeks to 3 months, despite having done additional PCQT [7], PCQR [16], PQR [10,16], PCQNT [14] and MOQT [17] that too with increasing percentage from 21.87% to 81.25%, details referred in Table 4 [1,7,10,12,13,14,16,17,19,25,16]. The success rate further declines from 80% - 33% when treatment is initiated after age of 3 months with open reduction and QVYP [12,27]. More poor results with QVYP are reported due to complications of increased morbidity, stiffness, instability, long incision, scarring, adhesions, wound dehiscence, blood loss and significant parent's anxiety [1,8,9,11,12,13,17].

Tarek & Shady [10,17], suggest different criteria for PQR that: if a range of knee flexion is achieved >90°, serial casting is continued; if range of flexion remains <90°, proceed for PQR and cast. In CDK III dislocations Tarek [10] reports, many failures even with addition of PQR and recommend PQR from the start, in the age over one month, in Grade I & II and QVYP in Grade III and recurrent CDK as well. Similar to Bansahel [13] in current study there was no significant failures in Grade III dislocations even in age over one month with & without PCT. It was mostly the matter of time paid for manipulation, gentleness and patience at first and subsequent visits especially when treatment begun day after birth. Cheng [7] on other side consider a dislocation as "teratogenic", when closed reduction within 24 hours of birth failed to achieve desirable results and recommend PCQT. The four patients in this series having associated non-syndromic DDH, Club foot & knee flexion deformity on other side, fortunately responded well without difficulty with casting and PCQT followed by Pavlik Harness and Foot abduction splint, respectively.

Our previous experience [8] and the current study indicates manipulation, casting, PCQT and QVYP techniques are significant skilled work that needs a steep learning curve to achieve proper Tibio-Femoral realignment and flexion >130 degrees [10]. Otherwise it deems to failure as has been observed in case no. 22, who had 9 cast somewhere else and ultimate advice for surgery but we managed that with PCQT and casting at the age of 5 months which ends up excellently. The forced manipulation even in rigid knees, is absolutely contraindicated due to risk of physeal damage, separation of cartilaginous epiphysis, premature physeal fusion, impaired circulation, fracture and skin necrosis [1,4,8,10,18]. The only preventable complication, alarming for parent's anxiety in this study was plastic deformation (anterior bowing) of tibia that too resolved in due course of time, with added Vitamin-D drops therapy. Nirav R Shah [17] and others suggest to prevent fracture tibia or procurvatum [1,18] by not to use distal tibia or tibial shaft as lever to reduce, but pressure should be applied on proximal tibia & femur in posterior to anterior direction [1,17,18]. Till date follow up, we do not have seen any other complications.

The fortnightly casting regimen [7] compared to weekly serial casting [4,10,17] reproduce similar results. Moreover, the accelerated weekly regimen produced significant psycho-social benefits and relieved parent's anxiety regarding outcome. On other side the babies do have erythematic and swollen knees on weekly casting, compared to fortnightly casting, that too resolved after last maintenance cast for two weeks. The redness and swelling got subside, without deleterious effect on outcome.

The laterality, gender and associating Non-Syndromic Clubfoot / DDH have no significant influence on overall outcome of treatment. They however, require a lengthy period of foot abduction brace and a Pavlik Harness respectively. CDK responds well to at home exercises, to maintain the outcome achieved at the weaning of cast, hence post weaning splints for long duration is not usually required [10,22,23].

This study has certain short falls including lack of investigation regarding Pre-& natal predisposition and a need for long term follow up to know incidence of complication of premature physis fusion with resultant Tibial Procurvatum [1,18].

CONCLUSION

The CDK is easy to reduce without significant manipulation time, when treatment is started within 24 hours of birth. With increasing age from hours to four weeks, the manipulation takes more time to reduce with traction / manipulation and require more number of cast to maintain reduction. After four weeks age CDK often require an addition procedure of PCQT. However, after three months age the CDK mandatorily require an open reduction with QVYP. Age less than one month therefore shall be considered as a GOLDEN PERIOD for a conservative treatment without even minimal invasive surgical intervention. We conclude that the duration from birth to start of treatment is directly proportional to duration of traction / manipulation and required number of casts, with a significant financial and psychosocial burden on the family and surgeon as well, to achieve best functional outcome.

It is further recommended that the antenatal care in our community must include an antenatal Sonologic study to detect musculoskeletal defects, especially in high risk patients with family history, breach presentation and Oligo-hydromnios. The Obstetrician hence to take responsibility to advice family for earliest (within 24 hours) Orthopaedic consultation of new borns with antenatal Ultrasonography impression of CDK & other congenital deformities as well.

REFERENCES

1. Jacobsen K, Vopalecky F. Congenital dislocation of the knee. Acta Orthop Scand. 1985;56:1
2. Curtis BH, Fisher RL. Congenital hyperextension with anterior subluxation of the knee. J Bone Joint Surg (Am), 1969 Mar,51:255–269.
3. Niebauer JJ, King DE. Congenital dislocation of the knee. J Bone Joint Surg Am. 1960 Mar;42:207.
4. Dobbs MB, Boehm S, Grange DK, Gurrnet CA. Case Report, Congenital knee dislocation in a patient with Larsen syndrome and a novel filament B mutation. Clin orthoprelat res 2008 Jun,466(6):1503-1509.
5. Mac Farland B. Congenital dislocation of the knee. J Bone Joint Surg 1929;11:281-5.
6. Katz MP, GrogonoBJS, Soper KC. The etiology and treatment of congenital dislocation of the knee. J Bone Joint Surg (B),1967 Feb;49:112-120.
7. Cheng CC, KoJY. Early reduction for congenital dislocation of the knee within twenty-four hours of birth. Chang Gung Med J. 2010 May-Jun,33(3):266-73.
8. Bhatti A, Lakho, MT, Azfar, M, Mengal, MA. Functional Outcome of Open Relocation of the Congenital Dislocated Knee with and without Distal Arthrogryposis. J Pak Orthop Assoc. 2015, 27 (3):11-16.
9. Ferris B, Aichroth P. The treatment of congenital knee dislocation. A review of nineteen knees. Clin Orthop Relat Res. 1987 Mar;(216):135-140.
10. Abdelaziz TH, Samir S. Congenital dislocation of the knee: a protocol for management based on degree of knee flexion. J Child Orthop. 2011 Apr;5(2):143-149.
11. Alurkar S, Walter S. Congenital Dislocation of the knee joint. Infant. 2008; 4(2):50.
12. Mayer L. Congenital anterior subluxation of the knee: Description of a new specimen, summary of the pathology of the deformity and discussion of its treatment. Am J Orthop Surg 1913 Feb;210(3):411-37.
13. Bensahel H, Dal Monte A, Hjelmstedt A, et al. Congenital dislocation of the knee. J Pediatr Orthop.1989 Mar;9(2):174–177.
14. Patwardhan S, Shyam A. Use of Percutaneous Needle Tenotomy for Treatment of Congenital Knee dislocation. J Orthop case Rep. 2012 Jul-Sep;2(3): 25–27.
15. Farhad Tajdar, Jan Victor. Unilateral congenital dislocation of the knee and hip: a case report. *Acta Orthop. Belg.*, 2012, 78,134-138.
16. Roy DR, Crawford AH. Percutaneous quadriceps recession: a technique for management of congenital hyperextension deformities of the knee in the neonate. J Pediatr Orthop. 1989 Nov-Dec;9:717–719.
17. Shah, Nirav R; Limpaphayom, Noppachart; Dobbs, Mathew. A Minimally Invasive Treatment protocol

for the congenital Dislocation of the Knee. J. Peditar Orthop 2009 Oct-Nov;29 (7):720-725.
18. Haga N, Nakamura S, Sakaguchi R, Yanagisako Y, Taniguchi K, Iwaya T. Congenital dislocation of the knee reduced spontaneously or with minimal treatment. J Pediatr Orthop 1997 Jan-Feb;17:59-62.
19. Roth S, Sestan B, Gruber B, Ledić D, Ostojić Z, Rakovac I. Bilateral congenital dislocation of the knee with ipsilateral developmental dysplasia of the hip - report of three patients. Coll Antropol. 2010 Mar;34(Sp1):299-305.
20. Sud A, Chaudhry A, Mehtani A, Tiwari A, Sharma D. Functional outcome following quadriceps tendon lengthening in congenital dislocation of the knee, with special reference to extensor weakness. Strat Trauma Limb Recon, 2009 Dec;(4): 123–127.
21. Tiwari M, Sharma N. Unilateral Congenital Knee and Hip Dislocation with Bilateral Clubfeet - A rare Packaging disorder. J. Orthop case Report. 2013 Apr-Jun;3(2):21-24.
22. Elmadag M, Ceylan HH, Erdil M, Imren Y, Bilsel K, Tuncay I. *Congenital Dislocation of Knee. Eur J Gen Med 2013;10(3):164-166.*
23. Shah KR, Shipra C, Abul K, Prakash S, Ranjiv J, Sanjaya S. Congenital genu recurvatum with dislocation of knee: a case report. J. Nobel med col. 2011(1):68-70.
24. Mehrafshan M, Wicart P, Ramanoudjame M, Seringe R, Glorion C, Rampal V. Congenital dislocation of the knee at birth- Part I: Clinical signs and Classification. Orthop Traumatol Surg. Res. 2016 Sep;102(5):631-633. Dot:10.1016/J. Ostr.2016.04.008. Epub 2016 June 3.
25. Bell M.J, Atkins RM, Sharrard WJ. Irreducible congenital dislocation of the knee. Aetiology and management. J Bone Joint Surg Br 1987 May;**69**(3): 403-06.
26. Carlson DH, O'Connor J(1976) Congenital Dislocation of the Knee. Am J Roentgenol 127:465-468.
27. Stern MB. Congenital dislocation of the knee. Clin Orthop. Relat Res 1968;61:261-8.

AUTHORSHIP AND CONTRIBUTION DECLARATION

No	Author Name	Contribution to paper	Signature
1	Principal author Anisuddin Bhatti	Managed all cases. Follow till date. Written script.	
2	Second Author Kiran Maqsood	Assisted patients casting. Helped in followup cases. Collected references, helped and written manuscript, re checked manuscript	
3	Third Author Maqsood Hassan Shaikh	Assisted patients caseting, helped in followup, written results and evaluation and followup cases	
4	Pervez Ali	Data Collection & Interpretation	
5	Zulfiqar Ali	Data Interpretation	
6	Muhammad Yousuf Bhatti	Final re-checked study for publication	