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CONGENITAL DISLOCATION OF THE KNEE: PRENATAL DIAGNOSTICS AND TREATMENT AT AN EARLY AGE

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Background. Congenital dislocation of the knee (CDK) is a rare abnormality of the musculoskeletal system, with an incidence of 1 per 100,000 liveborn infants. Timely prenatal diagnostics and treatment during the initial days of life can help avoid the development of disabilities in a child.
Aim of the study. We aimed to study the possible prenatal ultrasound diagnostics and to assess the efficacy of early orthopedic alignment using conservative methods of treatment.
Materials and methods. From January 1988 to February 2016, 37 newborns (50 lower limbs) with CDK were treated. The initial assessment of the affected limbs was performed immediately after birth. To determine the severity of dislocation, the Seringe and Tarek classifications were used. Conservative treatment was performed for all the patients. The age of pediatric patients at the time of treatment onset ranged from 2 hours to 5 days. Various methods were used, such as stage plaster bandages (10 lower limbs) and correction using the von Rosen splint (8 lower limbs). Since 2003, a single treatment protocol, developed by the authors of this study, has been applied.
Results. The prenatal ultrasound screening enabled the detection of CDK before birth in 21% of cases. Long-term results (catamnesis from 3 to 28 years) were evaluated by the Seringe scale and were excellent in 60%, good in 32%, and satisfactory in 8% of cases. Bad results were not registered. All the pediatric patients included in the study began to walk independently at the age of 9–18 months.
Conclusion. Prenatal ultrasound diagnosis enables the detection of CDK. Treatment of newborns, started in the first hours of life, according to the protocol developed by the authors, enables the alignment of the dislocated lower leg in a short time, without using prolonged stage plaster bandages. Long-term results demonstrate the efficiency of the proposed methodology.

Keywords: Congenital dislocation of the knee, prenatal diagnostics.

ВРОЖДЕННЫЙ ПЕРЕДНИЙ ВЫВИХ ГОЛЕНИ: ПРЕНАТАЛЬНАЯ ДИАГНОСТИКА И ЛЕЧЕНИЕ В РАННЕМ ВОЗРАСТЕ

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Актуальность. Врожденный передний вывих голени (ВПВГ) — это редкое заболевание опорно-двигательной системы с частотой встречаемости 1 на 100 000 живых новорожденных. Своевременная пренатальная диагностика и лечение, начатое в первые дни жизни, позволяют избежать инвалидизации ребенка.
Цель исследования: изучить возможности пренатальной ультразвуковой диагностики и оценить эффективность ранней ортопедической коррекции с применением консервативных методов лечения.
Материалы и методы. За период с января 1988 по февраль 2016 г. были пролечены 37 новорожденных (50 нижних конечностей) с ВПВГ. Первичная оценка пораженных конечностей осуществлялась сразу после рождения. Для определения тяжести вывиха использовались классификации Seringe и Tarek. Всем пациентам проводилось консервативное лечение. Возраст детей в момент начала лечения составил от 2 часов до 5 дней. Были использованы различные методики: этапные гипсовые повязки (10 нижних конечностей), коррекция на

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шине Розена (8 нижних конечностей); с 2003 г.применялся единый протокол лечения, разработанный авторами исследования.

Результаты. Пренатальный УЗ-скрининг позволил выявить ВПВГ до рождения ребенка в 21 % случаев. Отдаленные результаты (катамнез от 3 до 28 лет) оценивались по шкале Seringe и были отличными в 60 % случаев, хорошими — в 32 % и удовлетворительными — в 8 % случаев. Плохих результатов не было. Все дети, включенные в исследование, начали самостоятельно ходить в возрасте 9–18 месяцев.

Заключение. Пренатальная УЗИ-диагностика позволяет выявить ВПВГ. Лечение новорожденных, начатое в первые часы жизни по протоколу, разработанному авторами, позволяет в короткие сроки, без длительных этапных гипсовых повязок вправить вывих голени. Отдаленные результаты демонстрируют эффективность предложенной методики.

Ключевые слова: врожденный передний вывих голени, пренатальная диагностика.

Introduction

Congenital dislocation of the knee (CDK) is an extremely rare disease of the musculoskeletal system. It was first described by Chanssier in 1812 and Chatelaine in 1822 [1, 2].

CDK occurs in about 1 per 100,000 liveborn infants, which is approximately 1% of the incidence of congenital hip dislocation [3] and is characterized by anterior displacement of the tibial condyles relative to the hip condyles (Figs. 1 and 2). It either presents as an isolated deformation or combined with various musculoskeletal abnormalities, such as congenital hip dislocation and congenital clubfoot. Often CDK is observed in genetic syndromes (such as arthrogryposis and Larssen's syndrome) and lesions of the nervous system (such as a cerebrospinal hernia and diastematomyelia) [4, 5].

Although the etiology of the disease is not entirely understood, evidently CDK does not belong to the so-called "congenital abnormalities of aniage," since it manifests in the second half of pregnancy, when all elements of the musculoskeletal system have already been formed. Various studies suggest that hereditary predisposition is not traced, although there are separate observations of familial cases of CDK [6-8]. Hypoplasia of the anterior crucial ligament and hypoplasia or contracture of the quadriceps femoris cause "instability of the knee joint" [4, 9, 10]. The combination of these internal factors with external factors (such as the lack of amniotic fluid, pelvic presentation, and lack of intrauterine space), acts as a precursor for CDK [9-11].

Of various classifications of CDK, based on anatomical and functional changes, present in the literature, the most common classification is submitted by J. Leveuf (1946) [3, 4, 11-13].

The stages of the emergence of CDK are:

Stage I (recurvation): the articular surface of the tibia is displaced anteriorly regarding the epiphysis of the femur and with its upper edge extending into the area of articulation of the hip with the kneecap (Fig. 3a).

Stage II (subluxation): the posterior edge of the tibia rests against the anterior part of the articular surface of the condyles of the hip (Fig. 3b).

Stage III (dislocation): displacement of the tibia occurs under the influence of the load both anteriorly and in an upward direction (Fig. 3c).

Currently, the diagnosis of CDK remains disputable, and many researchers believe that

Fig. 1. Patient M., 6 hours after birth, with congenital anterior dislocation of the right knee

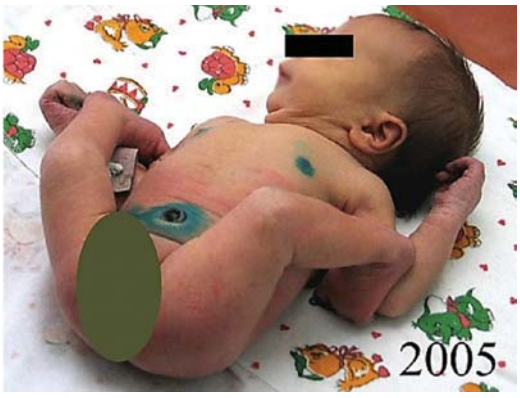
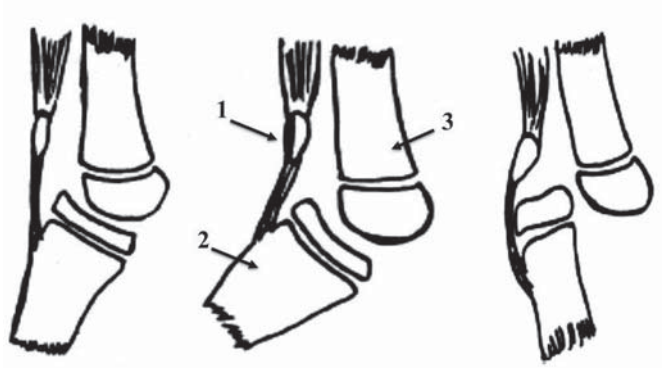


Fig. 2. Patient S., 6 days, with bilateral CDK



a *b* *c*

Fig. 3. The stages of CDK: *a* — recurvation (stage I); *b* — subluxation (stage II); *c* — dislocation (stage III). 1, Kneecap; 2, tibia; 3, femur. [Source: Leveuf (1946)]

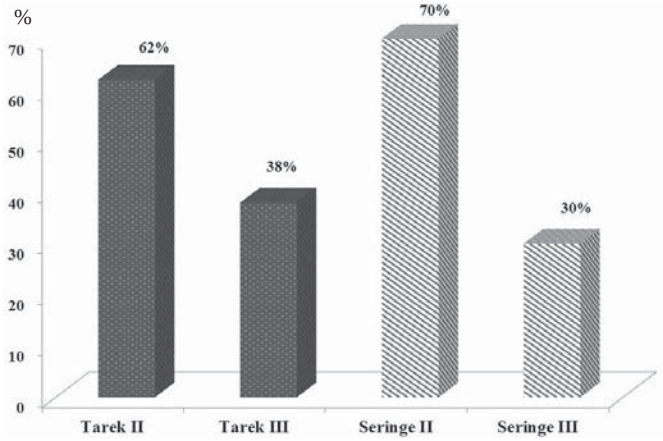


Fig. 4. Classification based on the severity of CDK (n = 50; p = 0.01)

radiography after the birth of a child offers the possibility of accurate diagnosis. Although there are studies in which ultrasound was used to diagnose and monitor the treatment, very few papers on prenatal diagnostics have been published [14-19].

Perhaps, determination of the onset time and selection of the treatment modality remains an urgent issue. Reportedly, while initiating a conservative treatment right after the birth is suggested by some, observation during the first month when spontaneous repositioning of CDK is possible is recommended by others [20-24].

The aim of this study was to investigate the results of prenatal ultrasound diagnostics and early orthopedic correction of CDK with conservative methods of treatment.

Materials and methods

From January 1988 to February 2016, we examined and treated 37 newborns with CDK (50 lower extremities), from the age of 5 minutes to 5 days. Isolated pathology was observed in 29 children, CDK combined with congenital dislocation of the hips was identified in 5 patients, and equinus foot deformity was present in 3 patients (Table 1). All parents of patients voluntarily signed informed consent to participate in this study.

We used the following two systems to assess the severity of CDK during the initial examination and according to records in the case history (retrospectively) [22, 25]:

1. The Tarek system (2011) is based on the assessment of the level of passive bending of the tibia:
- Degree I: the value of passive bending is more than 90° and the X-ray pattern reveals simple hyperextension;
- Degree II: the value of passive bending is 30°–90° and the X-ray pattern reveals subluxation/dislocation; and
- Degree III: the value of passive bending is less than 30° and the X-ray pattern reveals dislocation.

2. The Seringe system (1992) is based on the assessment of the possibility of eliminating a tibial dislocation at the one-stage correction:
- Type I: the result of the one-stage correction of the tibia is removable dislocation;
- Type II: the result of the one-stage correction of the tibia is a persistent or incompletely removable dislocation; and
- Type III: the result of the one-stage correction of the tibia is an irreducible dislocation.

In this study, two-third of treated patients presented with moderate CDK. Based on the

Table 1

Distribution of children by gender and the presence of CDK (n = 37; P = 0.01)

Patients	Left-sided CDK	Right-sided CDK	Bilateral CDK	Total
Boys	8	6	8	22
Girls	6	4	5	15
Total	14	10	13	37

CDK — congenital dislocation of the knee

Fig. 5. Location of the hands of the surgeon with manual correction of CDK

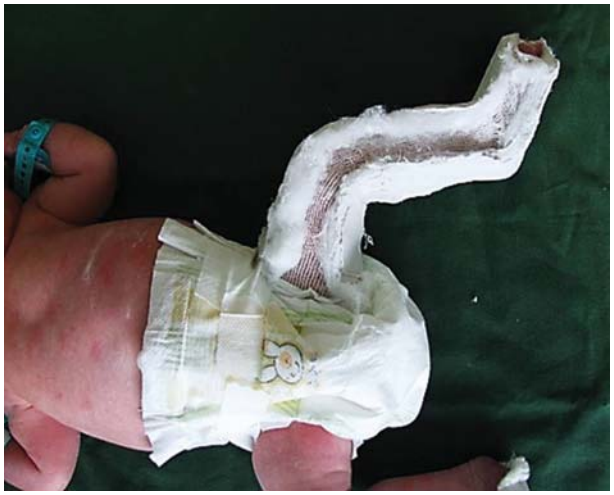


Fig. 6. Fixation of the lower limb with a three-quarter splint in the position of the correction achieved

Tarek system, 31 knee joints (KJ) were evaluated as Tarek II and 19 as Tarek III. Furthermore, per the Seringe system, 35 KJs were persistent, i.e., type II, and 15 KJs were irreducible, i.e., type III (Fig. 4). This study did not include patients with simple hyperextension, easily reducible cases (i.e., Seringe 1 and Tarek 1 with bending > 90°), and stiff arthrogryposis KJs.

Prenatal diagnosis

All pregnant women underwent the standard screening ultrasound examination on the decreed dates (on 10–14, 18–22, and 30–34 weeks of pregnancy). For pathological advice, an expert from a medical genetic center and an orthopedist were appointed. The ultrasound examinations were conducted on diagnostic devices of the middle and expert classes, Logiq 500PRO (General Electric Medical System, Germany) and Accuvix XQ (Medison, South Korea), with the use of multifrequency abdominal convex transducers with a working frequency of 3 to 7 MHz. Moreover, in the early stages of pregnancy, transvaginal sensors with an operating frequency up to 7.5 MHz were used. In one case, a magnetic resonance imaging (MRI) scan of the fetus was also performed at a term of 30 weeks.

Thus, upon detection of CDK, pregnant women were informed about the presence of pathology of the lower limbs in the fetus. An orthopedist assessed the case immediately after the delivery.

In cases where prenatal diagnostics did not detect CDK, the data of ultrasound studies were retrospectively evaluated. The so-called "missed" pathology was not revealed.

Treatment

Based on the methods of treatment used, patients were categorized into three groups:

- Group 1 (10 patients in the period from 1988 to 2000): a method of correction with stepped cylinder casts was used;
- Group 2 (5 patients in the period from 1991 to 2002): included patients who received treatment by using the Rosen splint; and
- Group 3 (22 patients in the period from 2003 to 2015): included children whose treatment was carried out according to the CDK correction protocol developed by us.

We devised the following treatment protocol:

Stage 1: Treatment with manipulation.

Continuous traction over the tibia and foot was performed, with simultaneous counterpressure on the condyles of the hip and tibia and subsequent bending of the tibia (Fig. 5).

If after the manipulation, the repositioning was achieved with bending in the KJ > 90°, the limb was fixed with a three-quarter splint or a cylinder cast for 7–10 days in the position of the tibia bending by 90° (Fig. 6).

Stage 2

In the absence of repositioning after the initial manipulation, the limb was fixed in the position of the bending obtained for 2–4 days. After removal of the primary fixation, traction and bending were repeatedly continued, and the additional bending achieved was fixed with a subsequent dressing.

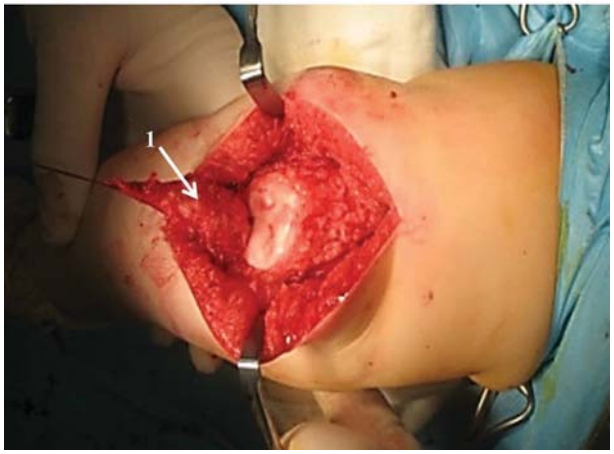


Fig. 7. Stage of the surgical correction: *1* — the tendon of the quadriceps femoris

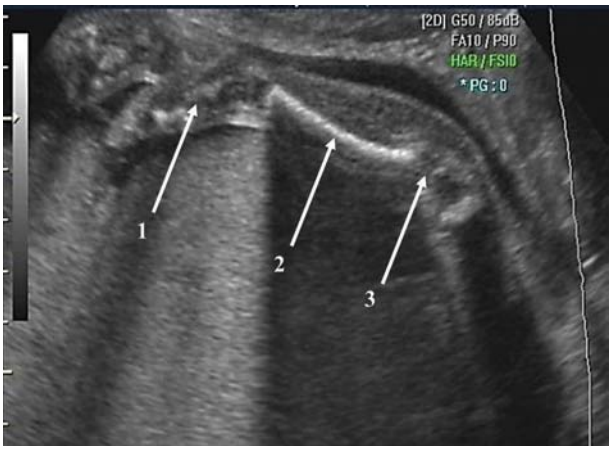


Fig. 8. Ultrasonogram of the lower limb of the fetus with CDK at the gestation period of 20 weeks. *1* — Foot; *2* — tibia; *3* — KJ

The total number of dressings until the tibia repositioning was 2 to 4, and the total fixation time was up to 1 month.

Stage 3

During a conservative treatment, all KJs were clinically monitored (by measuring the angle of bending of the tibia and palpation for false correction) by the ultrasound examinations. In control, an increase in the bending volume in the KJ was determined in a number of patients. However, it was not because of repositioning but due to anterior displacement of the proximal end of the tibia, which we regarded as a false correction. In this case, if no repositioning was achieved, subcutaneous tenotomy of the tendon of the quadriceps femoris was performed, followed by plaster corrections.

Stage 4

If, after the stage 3, the tibia repositioning was not achieved, a patient underwent V–Y-shaped quadriceps grafting and the front release of the KJ (Fig. 7). These procedures were performed at the age of under 1 year.

For successful traction, bending, and repositioning of the tibia, the following conditions were necessary: during traction, the foot and the epimalleolar region were captured, the traction was performed with one or both hands, the surgeon's elbows were fixed on the table, and additional pressure was applied to the pelvis to enhance the effect. The minimum traction time was 20 minutes. Then, counterpressure was applied to the condyles of the hip and the tibia. Simultaneously, the distal end of the hip was displaced ventrally (anteriorly) and the proximal end of the tibia was displaced dorsally and posteriorly. Pressure on the distal end of the tibia was not applied to avoid angulation of the tibia.

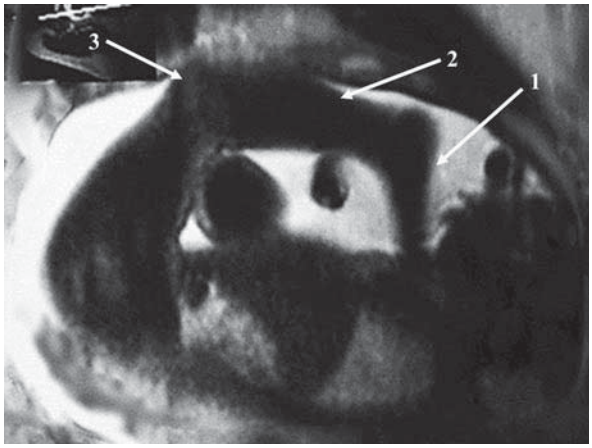
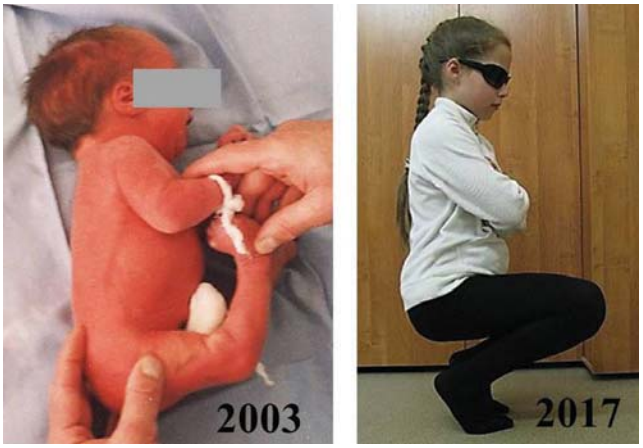


Fig. 9. MRI of a pregnant woman at 30 weeks. Congenital anterior dislocation of knee in the is observed. 1, Foot; 2, tibia; 3, KJ



a *b*

Fig. 10. Patient Sh. *a* — the first day after birth; *b* — 14 years after the conservative repositioning of a bilateral severe congenital anterior dislocation of the knee

Surgical treatment

For patients resistant to conservative treatment, surgical correction was used. In 8% of patients (3 patients, 4 KJ) after conservative treatment and tenotomy, the full anterior displacement of the tibia relative to the hip condyles was preserved, which was an indication for surgical intervention. The V–Y-shaped quadriceps grafting and the anterior release of the KJ was performed. The elongated tendon of the quadriceps femoris was sutured with bending of the tibia at 45°, and a three-quarter plaster cast was applied in the same position for 3 weeks. A special opening window was created for performing dressings and checking the postoperative area. In addition, stage plaster corrections with additional bending were performed neatly, starting from day 20 after the surgery to 2 months.

Evaluation of treatment results

To evaluate the results of the treatment at different times of observation, we used the Seringe criteria, which considers the volume of movements and stability of the KJ (Table 2) [22].

The excellent results include normal KJs; good results include joints with normal mobility, having slight anteroposterior instability, or stable KJs with slight decrease in bending; satisfactory results include stable KJs with a tibial bending from 50° to 90° or unstable KJs with a tibial bending greater than 90°; and poor results include unstable and/or stiff KJs.

Results

In this study, the sample was representative, and the probability of error was $p = 0.01$. In all comparison groups, the χ^2 compliance index was calculated, including the corrections of Yates and Fisher. In all patients, the indices were higher than the tabulated ones, which confirms the statistical reliability of the results of this study.

In 8 children (21.6%), CDK was diagnosed during the second trimester of pregnancy, gestational age of 18–24 weeks, by an ultrasound examination (Fig. 8). In remaining children, the disorder was not revealed in an ultrasound examination.

In one of the fetuses with CDK, at week 12 of gestation, normally formed KJs were noted, which was confirmed by a videotaped fetal study; however, at week 24, CDK appeared. Furthermore, in 1 patient, the diagnosis of CDK was confirmed at week 30 of pregnancy with an MRI scan (Fig. 9), which indicated suspected multiple malformations.

The catamnesis ranged from 3 to 28 years, and the average follow-up period was 13.5 years (Fig. 10).

In our study, the results of treatment of 30 KJs (60%) were assessed as excellent, 16 (32%) were good, and 4 (8%) were satisfactory. There were no poor results (Table 3).

Notably, the time to achieve repositioning of the tibia was different for patients in different groups. In group 1, using gypsum corrections, the duration of treatment was maximal, which on average was

Table 2

The Seringe criteria for evaluation of treatment results

Result	Range of mobility	Instability
Excellent	Complete	Absent
Good	Complete	Anteroposterior
	Bending limitation (90–140°)	Absent
Satisfactory	Bending limitation (50–90°)	Absent
	Complete	Complete
Poor	Stiff	Complete

Table 3

Distribution of results by treatment methods ($n = 50$; $p = 0.01$)

Method	Result (%)			
	Excellent	Good	Satisfactory	Poor
Plaster corrections	53,8	30,8	15,4	0
Rosen splint	57,1	28,6	14,3	0
Author's method	66,7	30	3,3	0

Table 4

The average time to achieve repositioning of the tibia with different methods of treatment ($n = 50$, $p = 0.01$)

Method	Term of repositioning	Average term of repositioning
Plaster corrections	21–28 days	25 days
Rosen splint	5–10 days	8 days
Author's method	40 minutes — 6 days	3 days

Table 5

Term of the tibia repositioning and frequency of extensive surgical interventions in children with CDK ($n = 37$, $p = 0.01$)

Number of children (number of KJ)	Treatment onset	Mean time to achieve the tibia repositioning	Necessity in extensive surgical interventions
28 (38)	1 day of life	6 days	5 %
9 (12)	2 days of life and older	23 days	17 %

25 days; in groups 2 and 3, it was 8 and 3 days, respectively. Thus, selection of the method of treatment of CDK determines the time of achieving repositioning (Table 4).

In addition, we identified that the age of patients undergoing the treatment influenced the term of the tibia repositioning. Thus, 28 children were treated on the first day of life (38 KJs). Overall, the CDK repositioning was achieved for 6 days, whereas extensive surgical interventions were required in 5% of patients. In patients who started the treatment at the age of 2 or more days (9 children, 12 KJs), the average time to achieve the tibia repositioning was 23 days, and extensive surgical interventions were required in 17% of patients. Thus, an orthopedic correction of CDK in the first day of life was much more efficient than in later periods (Table 5).

A conservative treatment without any surgical intervention (including without tenotomy) was successful in 81% of patients (30 of 37 children). Tenotomy was performed on the remaining 7 patients (11 KJs), of whom 4 responded positively (7 KJs) and 3 needed extensive surgical interventions to obtain repositioning (4 KJs).

Complications were noted in 6 patients (12% of the overall KJs). In 1 patient (2% of all KJs), maceration of the skin was observed at the points of application of pressure along the posterior surface of the condyles of the femur. In another patient (2% of all KJs), there was a minimal fracture of the cartilaginous model of the inner condyle of the hip due to excessive pressure during correction. In 4 patients (8% of all KJs), the angulation of the tibia was detected and bony cartilaginous complications were confirmed by an MRI scan. Later, the complications were resolved independently and did not manifest in the long-term results.

Discussion

Prenatal diagnostics. To date, a single study has reported prenatal diagnostics of CDK. Another study presented random findings during a radiographic examination of a pregnant woman about another pathology [14, 16]. Furthermore, there is only one report on prenatal ultrasound diagnostics of CDK without any systemic character. However, studies defining CDK in an ultrasound examination demonstrate the detection of this congenital abnormality during 20–24 weeks of gestation [17-19]. All studies mentioned above do not report on the timing of the treatment onset, the severity of CDK after delivery, and do not explain the treatment protocol. Notably, mandatory screening of all pregnant women is not applied in foreign health systems, and a small number of published observations indicate that the researchers did not undertake the factor of prenatal diagnostics [14, 16-19].

In our study, while conducting an ultrasound examination in the decreed period, CDK was diagnosed in 21.6% of patients (8 patients), which indicates a purposeful and non-accidental detection of such a rare pathology. In contrast, such a low percentage of CDK detection, even at week 32 demonstrates, in our opinion, the lack of orthopedic alertness, rather than, first of all, the difficulty in diagnosing this congenital abnormality. In all patients of prenatal detection

of CDK, a preliminary treatment plan was devised before delivery, and the orthopedic correction began in the first hours of life in the absence of somatic contraindications.

Terms of the treatment onset. There is no consensus in the literature on the timing of the initiation of CDK treatment. Many believe that treatment should be initiated in the first few days after birth. Thus, Laurence (1967) argues that the prognosis of CDK deteriorates due to delay in treatment and the presence of other deformations of the musculoskeletal system. Similarly, Chun-Chien Cheng (2010) recommends early, accurate repositioning of CDK in the first 24 hours of life, as, in his opinion, the repositioning of CDK deteriorates with each subsequent hour of the child's life. Furthermore, he suggests that it is especially easy to set the KJ, if treatment is started before 8 hours of life because in the case of any delay, the likelihood of surgical intervention increases. Hence, CDK is a diagnosis that requires immediate treatment [12, 21, 24, 26, 27].

Conversely, Haga et al. (1997) believe that it is necessary to wait for 1 month before the spontaneous repositioning of CDK to rule out its association with clubfoot, arthrogryposis, and Larssen syndrome. They study 6 patients with CDK, 4 of whom did not receive any treatment, and 2 received a conservative treatment in the form of a posterior plaster splint with fixed bending. When assessing the volume of movements in the long-term results, which was 2 years, they noted the hyperextension of the KJs in 2 patients from the group without treatment, to 20° and 10°, and in 2 patients who received treatment, up to 10° and 10°. Iwaya et al. (1983) examined 5 patients (6 CDK) who started receiving the treatment from the age of 20 days to 8 months with plaster corrections and the Pavlik Harness. In their study, the total volume of bending was reached only in 4 KJs, as in the remaining 2 KJs, the bending was 120° and 145°. In 2 patients with CDK, the hyperextension of the tibia was observed at an angle of 30° [20, 28].

In our study, in 76% of patients, treatment was started on the first day of life. On average, the CDK repositioning was achieved in 6 days, and extensive surgical treatment was required in 5% of patients. In patients who started receiving the treatment at the age of 2 or more days (9 children, 12 KJs), the average time to achieve the tibia repositioning was 23 days, and extensive surgical treatment was required in 17% of patients. Thus, we state that an orthopedic correction of CDK, started in the first hours of life, enables a significant reduction in the percentage of patients who need a surgical treatment, to shorten the period of treatment, which positively affects not only the well-being of the newborn but also helps them to develop without delay. Although there is no data on the percentage of patients undergoing a surgical treatment, depending on the timing of the onset of conservative correction, many authors describe methods of surgical intervention with CDK [3-5, 11, 21, 25, 26].

Assessment of treatment results and catamnesis. In the evaluation of treatment results, most authors indicate a rather short period of long-term follow-up (less than 5 years); however, only a few report on the terms for more than 8 years. Thus, Haga et al. (1997) describe results with a follow-up period of up to 9.8 years (an average of 3 years 4 months), evaluating them according to the Johnson classification. Curtis and Fisher (1969) indicate a catamnesis of up to 25 years (an average of 12 years), evaluating the results as excellent, good, satisfactory, and poor. Jih-Yang Ko (1999), who observed patients for 9 years, categorized the results as excellent, good, satisfactory, and poor. Rampal (2016) reports on the time of a long-term follow-up period of up to 26 years (an average term is 9.3 years), using the Seringe system to evaluate the results. In all these systems, the range of movements and stability of the KJ is estimated. Some authors include pain, satisfaction with the result, the need for orthopedic devices, and rapid fatigue in the assessment scale [4, 5, 13, 20, 22, 23, 25].

In our study, catamnesis ranged from 3 to 28 years, with an average of 13.5 years. The literature does not describe such a specified duration of a patient's follow-up. We evaluated the outcome of treatment on the Seringe scale as the simplest and most convenient. In 92% of patients, we obtained excellent and good results, and there were no poor results. Most studies suggest the use of staged plaster corrections [3, 11, 26] but only a few recommend manipulations in combination with retractors, such as the Pavlik Harness [21, 28]. The proposed method enhanced the number of good and excellent results by 12% compared to treatment with plaster corrections and Rosen splint. It should be noted that this technique

ensured the repositioning of the tibial dislocation on average for 3 days, while the average correction period with the Rosen splint was 8 days, and with the staged plaster bandages it was 25 days. Thus, the method developed by us has enabled to shorten the duration of treatment significantly [3, 5, 11, 13, 21, 25, 26, 28].

Conclusion

For the first time, an assessment of the potential for prenatal diagnostics of CDK was carried out in a large clinical study with the longest catamnesis, and a treatment protocol was developed. The obtained results do not contradict the published results, but only confirm the opinion of the majority of authors in the developed observations. The prenatal diagnostics of CDK is effective in more than 20% of patients, which enables treatment initiation in the first hours of the life of an infant. Early conservative correction enables a significant reduction in the duration of treatment (on average up to 3 days) and reduces the number of surgical interventions by 12%. A high percentage (92%) of good and excellent long-term results recommends our method for a wide clinical application in the treatment of newborns with CDK.

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