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Long-term results of pediatric treatment of congenital vertical talus

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Цель. Изучение отдаленных результатов лечения стоп с врожденным вертикальным положением таранной кости **Objective** To review long-term results of congenital vertical talus treated with classical techniques and a minimally invasive treatment approach offered by M. Dobbs. **Material and methods** Review of 30 pediatric cases (54 feet) with severe congenital convex pes valgus was performed over the period of 11 years. Patient ages ranged from 1 month to 13 years at the start of treatment. The 54 primary operations performed included triple arthrodesis (n = 2), open talus reduction according to S.J. Kumar, K.R. Cowell, D.L. Ramsey (n = 5), Coleman open reduction of the talus and screw fixation (n = 6), serial casting and percutaneous fixation of the talonavicular joint with a Kirschner wire and a percutaneous Achilles tendon tenotomy (n = 41). Reduction of the talus was produced under vision and followed by capsuloplasty of 23 feet. C-arm was used for closed correction in the rest of the cases. **Results** A minimally invasive Dobbs approach that consisted in serial casting, minimal surgery followed by bracing was shown to provide favorable outcomes in 21 patients (41 feet) treated at early childhood. Recurrent deformity with the oblique talus developed in 3 cases (4 feet) during growth of an average 6.1 years despite satisfactory primary correction and required further interventions. However, open reduction has become a rare procedure in the patient cohort with introduction of Dobbs practice at our hospital. There are more reports of successful primary treatment with Dobbs method in the national literature with no long-term follow-ups available in our country. **Conclusions** Kumar and Coleman operative interventions are reserved for unrecognized or recurrent cases following Dobbs manipulations. Triple arthrodesis as a definitive procedure can be used in a late detection of the condition or poor outcomes.

Keywords: congenital convex pes valgus, vertical talus, Dobbs method, foot deformity

INTRODUCTION

Vertical talus is a congenital abnormality characterized by a rigid flat and valgus foot [1]. The estimated prevalence of vertical talus is 1 in 10,000 live births [2, 3]. Approximately 50 % of the cases occur in conjunction with neurologic disorders, chromosomal abnormalities and genetic syndromes, neuromuscular conditions including arthrogryposis, neurofibromatosis, myelodysplasia and other anomalies [4, 5, 6]. The condition is inherited in an autosomal dominant fashion with incomplete penetrance [7]. Mutations in the HOXD10 M319K genes have been identified in patients with isolated vertical talus by M. Dobbs in 2006 [8]. Early conservative treatment with serial casting is recommended as a method of improving the deformity. The principles of cast correction of vertical talus are similar to those used in the Ponseti method of clubfoot correction. Dr. Matthew Dobbs, an orthopaedic surgeon and geneticist (Philadelphia, USA), developed the author's method. The Dobbs technique of cast correction of a vertical talus is based on a specific way of manipulating the foot using elastic soft tissues of the foot. Children up to the age of 3 years can avoid major surgical interventions due to fibrous changes in soft tissues of hind- and midfoot that are not resistant to restoration of anatomical relationship in the joints [3, 6, 9, 10, 11]. However, successful outcomes of congenital flat and valgus foot are reported less commonly than those of clubfoot [4, 12, 13, 14–17]. An observation of the group of non-idiopathic vertical talus has shown loss of correction during growth that may require surgical interventions.

MATERIAL AND METHODS

Dr.M.Dobbs attended our hospital in 2006 and reported short-term follow-up in patients with vertical talus treated with the author's method. We normally used traditional approaches in treatment of this cohort of patients prior to his arrival. The serial casting was used for the vertical talus to improve the

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deformity and prepare the patient for surgery that was an analogous to V.Ya. Vilensky and T.S. Zatsepin casting applied for clubfoot. At that time all patients with vertical talus underwent open reduction of the talus. Procedures were performed for children older than 18 months and consisted of soft-tissue release followed by fibular grafting of sinus tarsi after 5 years and triple arthrodesis at the age of 12 years.

Review of 30 pediatric cases (54 feet) with severe congenital vertical talus was performed over the period of 11 years. Patient ages ranged from 1 month to 13 years at the start of treatment. There were 54 primary surgeries performed with one procedure per foot. The patients were subdivided into 2 groups.

Group I (n = 9, 13 feet) included patients treated with traditional methods of open reduction of the talus. Preoperative casting applied in the group mostly failed. The mean age of the patients was 6.1 years. Open reduction of the talus was based on the age of the patient at presentation. Kumar, Cowell, Ramsey procedure was carried out in children younger than 4 years (5 feet), Coleman open reduction of the talus and screw fixation produced up to the age of 12 years (6 feet) and triple arthrodesis performed after 12 years of age (2 feet). Screw fixation of the foot with triple arthrodesis was traumatic but allowed the patients ambulate with full weight-bearing on the operated limb on week 4 postsurgery.

Group II (n = 21, 41 feet) included patients with vertical talus who received treatment at our hospital after introduction of Dobbs method into our practice. The mean age of the patients was 15 months. Serial casting and percutaneous fixation with a Kirschner wire and Achilles tendon tenotomy was produced for 41 feet. Talonavicular joint was reduced with consecutive manipulations of the forefoot and counterpressure to the plantar medial hemisphere of the talus head with still persisting equinus that was simultaneously corrected with Achilles tenotomy and fixation of the reduced talonavicular joint with Kirschner wire. Closed manual correction failed under C-arm control in 23 feet, and medial incision was produced for reduction, the head of the talus elevated under vision and capsulotomy carried out. Plaster cast was applied for 8 weeks postsurgery. Children had short leg casts on and longitudinal arch mold after pin removal without fixation of the knee joint to allow full weight-bearing on the leg within one month. Bracing period with the foot in neutral was 3 months 23 hours per day and further on during any sleep until the age of 4 years to prevent recurrence. The children were recommended to wear orthopaedic shoes with longitudinal arch mold. Despite satisfactory correction of the deformity with the Dobbs method recurrent deformity with the oblique and vertical talus developed in 3 cases (4 feet) and became an indication to repeat surgery.

All children of groups I and II were referred to a geneticist but not of them were seen by the specialist and received test results for various reasons. From 30 children, 24 (80 %) had vertical talus associated with either neuromuscular diseases or verified genetic syndromes: Freeman-Sheldon syndrome (n = 2), Patau syndrome (n = 1), arthrogryposis (n = 4), Marfan syndrome (n = 1), CP (n = 2). The majority of children had non-verified genetic syndromes: 14 patients with multiple (more than 5) minor malformations combined with severe weight deficiency, hypermobile joints of the limbs and presence of unilateral or bilateral vertical talus.

In addition to genetic tests all children underwent radiographic evaluation as an objective, simple and inexpensive method to assess severity of the deformity and the outcome. Radiographs were produced prior to non-operative treatment with suspected vertical talus, at the end of the course, prior to surgical treatment, intraoperatively and at the end of the treatment. Radiographs were also made with child growth in questionable clinical scenario Standard radiographic evaluation of vertical talus deformity included an anteroposterior view (AP) in neutral and lateral views (L) of the foot in maximal dorsiflexion and maximal plantar flexion. Patients with reduced talus at maximal plantar flexion that radiologically appeared as realigned longitudinal axis of the ossification center and the first metatarsal were excluded from the study because the oblique talus was not meant to be discussed in the article. The angles measured were: anteroposterior talocalcaneal (AP1), anteroposterior talar axis - first metatarsal base (AP2), lateral talocalcaneal (L1), lateral tibiocalcaneal (L2), lateral talar axis - first metatarsal base (L3). AP1 angle indicated to divergence of the hindfoot; AP2 angle tended to decrease during treatment and revealed abduction of the forefoot; L1 progressively decreased with increasing deformity reaching 0° in some cases

but demonstrated greater values with correction of the hindfoot; L2 indicated to severity of the hindfoot equinus; L3 tended to zero due to the treatment. Radiographic values are presented in Table 1.

Group I included patients treated with traditional techniques of open reduction of the talus. Group II was subdivided into IIA and IIB due to considerable differences in radiographic values. Subgroup IIA consisted of patients with vertical talus as the only clinical feature and without genetic syndromes (n = 6, 6 feet). Subgroup IIB included patients with vertical talus and verified and non-verified genetic syndromes (Fig. 1). Total 164 radiographs were evaluated pre-, intra- and postoperative treatment over the period of 6.8 years on average following the primary surgical treatment.

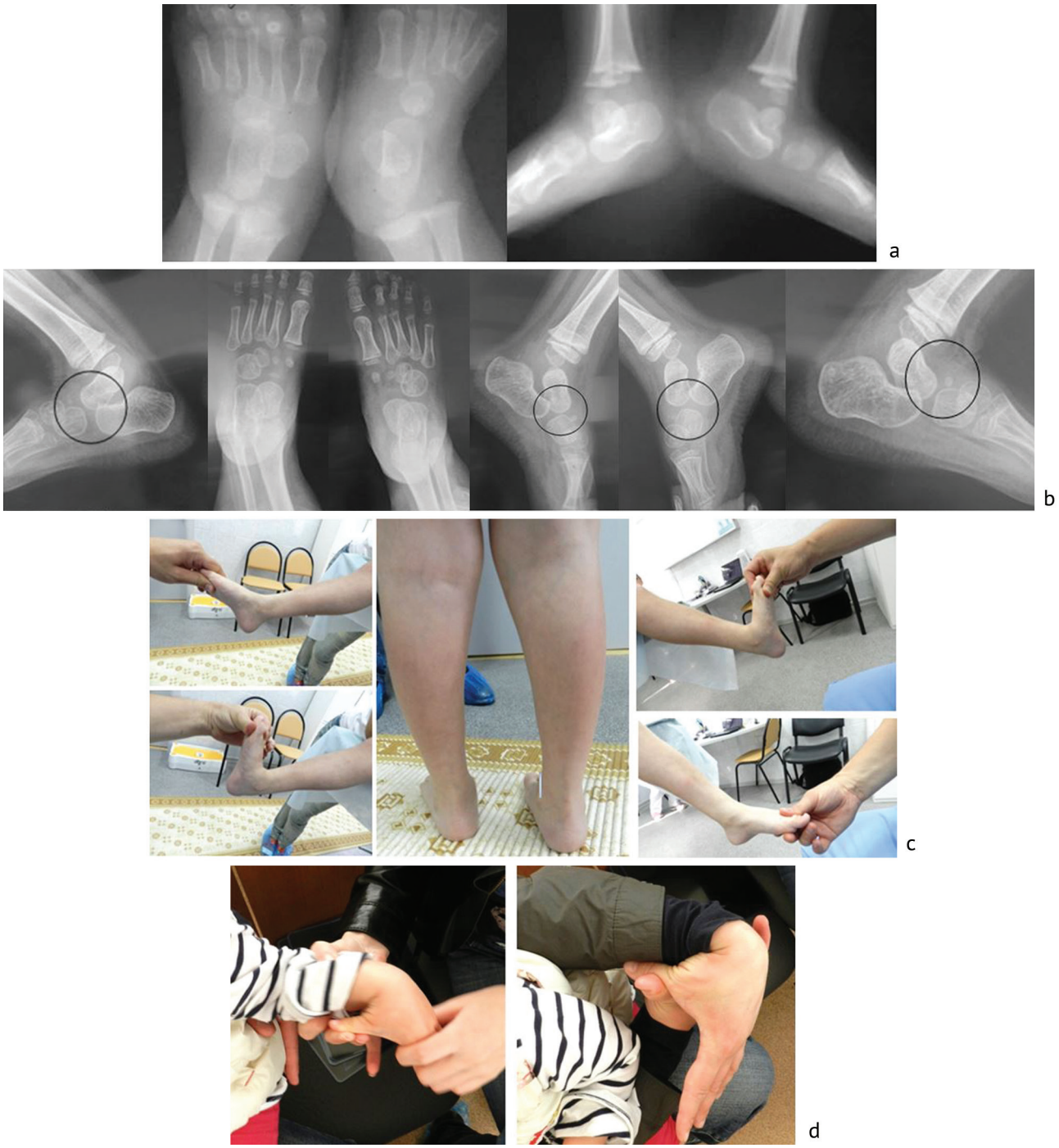

Fig. 1 Patient from subgroup IIB, 25 months old underwent reduction of the talus with Dobbs method in an open fashion on the right and closed manner on the left: ***a*** preoperative AP and lateral views of the feet at plantar flexion showing marked rigid deformity of the vertical talus; ***b*** AP standing and lateral views of the patient aged 5 years at maximal dorsiflexion and maximal plantar flexion showing navicular ossification center developing in the anterior-dorsal hemisphere of the head of the talus at dorsiflexion. Centering of the navicular bone is seen as complete on the right and incomplete on the left; ***c*** photograph of tibiae and feet of the child showing lowered longitudinal arch and weak triceps strength due to generalized dysplasia of connective tissue; ***d*** one of the tests demonstrating enhanced elasticity of connective tissue in father and the child

RESULTS

Long-term results were evaluated in 23 children (76.7 %). Patients were followed from 1 year to 14 years with the mean term of 6.8 years. The American Orthopaedic Foot and Ankle Society (AOFAS) scoring system was used as a clinical outcome measure evaluating hind- and midfoot. Midfoot Scale AO and Hindfoot Scale AO scores were 51.3 and 50.8 for arthrodesis group (n = 3), respectively. The mean scores for mid- and hindfoot were higher in soft-release group (n = 7) measuring 56.2 and 55.4, respectively. The Dobbs patients (n = 13) showed the highest values being 82.9 for midfoot and 85.5 for hindfoot. Statistical data analysis was problematic due to a small number of patients identified in both groups and but there was an evident tendency of increase in scores observed with less aggressive procedure. Further studies of the rare condition are necessary. Preoperative evaluation with the above scale systems could not be performed for some children with vertical talus because Dobbs treatment was performed before the children could walk.

Preoperative AP1 and AP2 angles indicated to marked abduction of the forefoot in both groups. Preoperative L1, L2 and L3 angles were characteristic of the severity of the deformity and presence of 'true' anomalies of the vertical talus. Postoperative radiographic values were better on the lateral views with the evidence of disturbed ossification and osteoarthritis in the joints of the hind- and midfoot. Lateral views of Group II showed inferior radiographic values with no signs of circulation disorders observed (Table 1). Radiometric values allowed evaluation of dynamics in changes of the foot during the treatment of the vertical talus in both study groups. Retrospective analysis of intraoperative radiographs showed that incomplete reduction of the talus and pin fixation (recurrence developed in two feet within a year) could be a major cause of failure (Fig. 2). Now we choose to make a medial incision to achieve complete correction under vision if there are doubts about complete reduction of the talonavicular joint.

Table 1

Preoperative and postoperative radiographic values of hind- and mid-foot by groups

Angle	Normative value	Term					
		Pre-op		Post-op		6-year follow-up	
		Group I (º)	Group II (º)	Group I (º)	Group II (º)	Group I (º)	Group II (º)
AP1	from +30º to +50º	53.2	56.4	31.1	37.2*	30.8	49.5*
AP2	from 0º to +20º	46.9	45.1	15.6*	18.5	13.4*	10.3*
L1	from +25º to +55º	2.2	2.5	45.1	39.2	33.4	19.2
L2	from +10º to +40º	-3.1	-7.7	19.9*	18.4	15.6	10.7
L3	from 0º to -45º	55.4	76.2	-20.1	-11.5	-15.9	-9.5*

* – P < 0.05 in relation to baseline value.

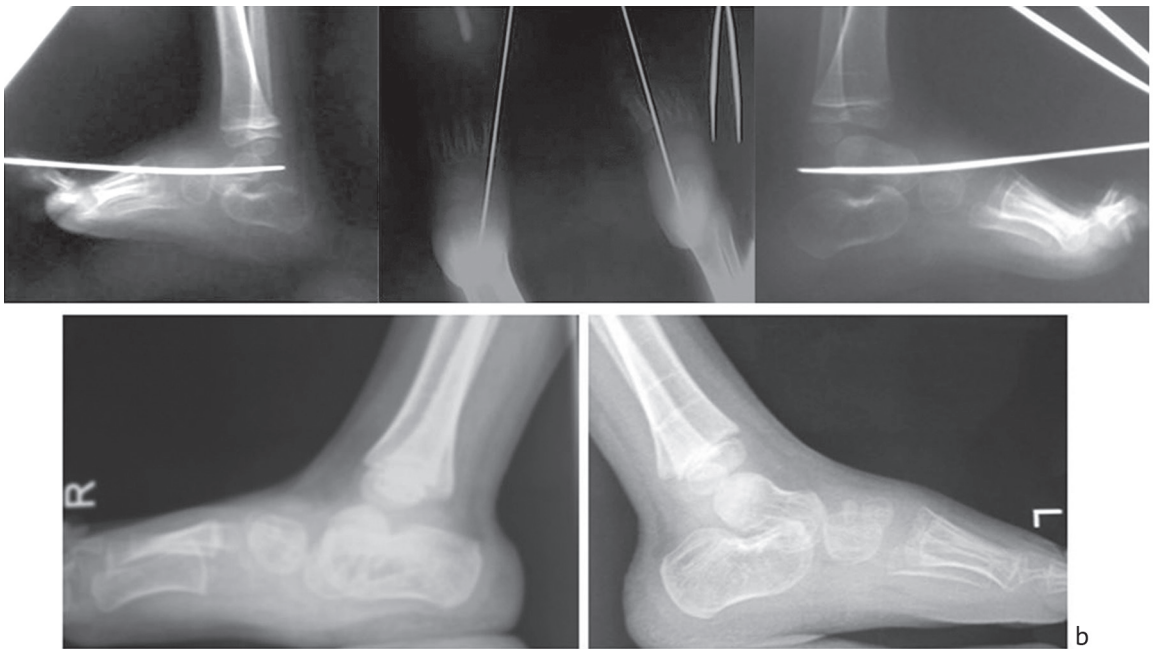

Fig. 2 Patient 2 with vertical talus was treated with Dobbs method at the age of 2 years: ***a*** intraoperative radiographs showing incomplete correction of the right foot; ***b*** recurrence of the vertical talus on the right developed at one year

We experienced difficulties in reduction in children younger than 4 to 5 years with absent ossification center of the navicular bone when applying Dobbs method. But the use of the axis of the first metatarsal and longitudinal axis of the talus ossification center was helpful in reduction maneuver and pin fixation. Complete reduction of idiopathic vertical talus in a 'key into lock' manner maintained with braces up to 3 years allowed prevention of recurrence. Loss of primary correction occurred in presence of genetic syndromes and neuromuscular diseases accompanied by pathological traction of tendons during growth. But flat foot and the oblique talus appeared to be pain free and mobile after the Dobbs treatment. Open reduction resulted in less AOFAS scores (by 28, on average) following open reduction. Arthroereisis procedure performed on larger anatomical structures and more mature bone was shown to provide good stable outcomes in children with partial recurrence and presence of syndromes at the age of 5-6 years due to more safe and reliable performance (Fig. 3).

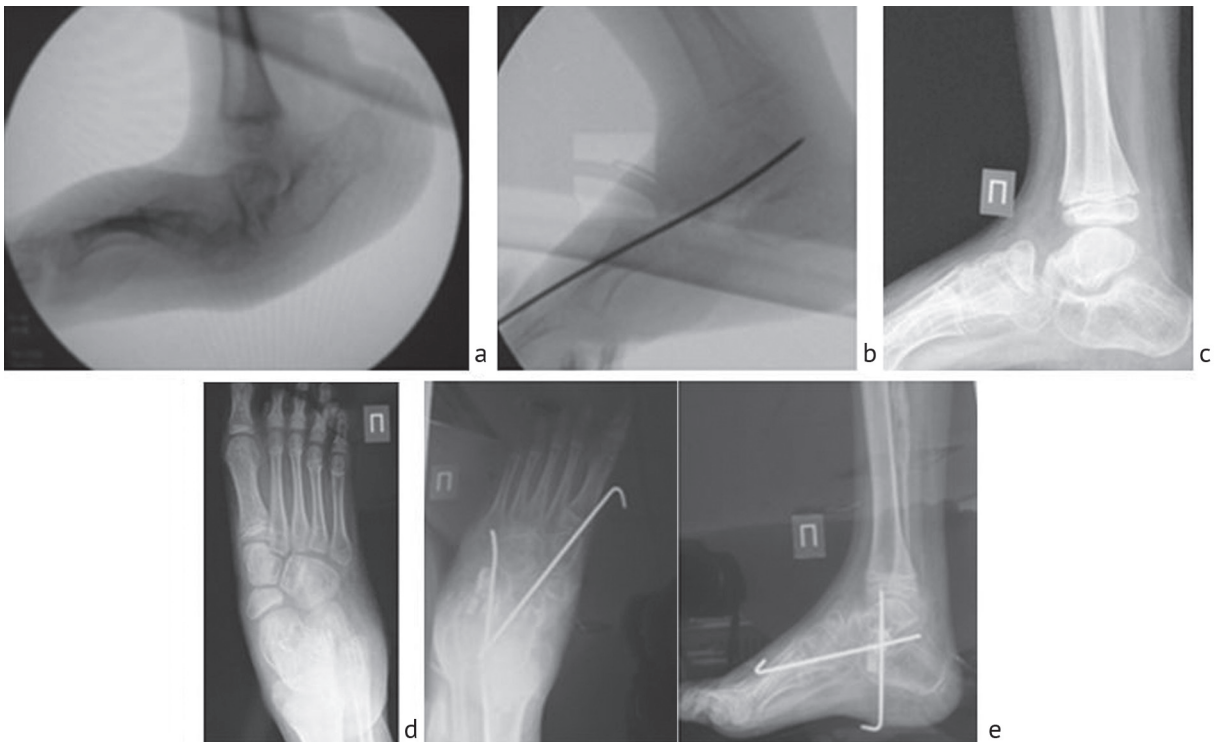

Fig. 3 ***a*** radiograph with plantar flexion showing vertical talus; ***b*** position of the talus after Kumar, Cowell, Ramsey procedure performed at the age of 3 years; ***c, d*** recurrence and the oblique talus developed during growth with non-verified genetic syndrome at the age of 6 years. Radiological signs of disturbed circulation in the hind- and midfoot developed after peritalar soft-tissue release. The hindfoot appeared to pull up, talar block lowered, specific features of ossification center of the head of the talus seen; ***e*** position of the talus after the Grice procedure at 8 weeks prior to pin removal

DISCUSSION

Radiographs in dynamics showed more stable variant of deformity correction in Group I with open reduction of the talus through aggressive intervention on soft tissues and foot bones. A cautious surgical approach is required due to unique pattern of the blood supply of talus (Fig. 4).

Signs of disturbed blood supply are normally seen after open reduction of the talus at a long-term follow-up with decreased height of the talar block, subchondral osteosclerosis and marginal osteophytes in the hind- and midfoot. Outcomes of open procedures measured with AOFAS scale for hind- and midfoot are inferior to those achieved with Dobbs treatment. The outcome measures are not expected to improve with weight gain during growth. However, radiographic values are close to normative ones and demonstrate more stable results during growth as compared to those seen with Dobbs treatment. Introduction of Dobbs method in treatment of congenital vertical talus in children younger than 3 years allowed us to avoid open reduction in 38 cases out of 41 (92.7 %). Loss of correction after primary treatment in subgroup IIB required repeat surgery (Fig. 3) in three patients (4 feet) with the oblique and vertical talus that resulted in repeat procedure. The Dobbs methods is likely to be relatively young and we found no reports on long-term outcomes of pediatric treatment of vertical talus in Russian and foreign literature.

Numerous techniques for treatment of congenital vertical talus have been proposed depending on clinical features and availability of foreign literature on primary correction of vertical talus in children. Multiple variants of operative techniques used for treatment of patients with vertical talus emphasize the absence of unified system of treatment. Nearly 50 % of all the cases are associated with neurologic disorders, chromosomal abnormalities and genetic syndromes, neuromuscular conditions including arthrogryposis, neurofibromatosis, myelodysplasia and other anomalies. The possibility and accessibility of syndrome grouping are very low in our country. An orthopaedic surgeon can often treat a child with vertical talus having no knowledge of underlying disease. So, the child is an enigma. The orthopaedic surgeon is unable to answer the question about the stability and perspectives of foot position with child's growth that indicates to the importance of continuous observation and preventive long-standing fixation of the foot to maintain the correction. Furthermore, because the congenital vertical talus is rare with the estimated prevalence of 1:10000 the condition remains unrecognized and treatment delayed. Congenital vertical talus occurs with the frequency of 1–4 births per year in a medium-sized city of the Russian Federation and a pediatric orthopaedic surgeon may never see a case throughout the career which makes diagnosis and treatment more difficult. Apart from that, long-standing stable correction is observed in subgroup IIA. No physical retardation is noted in the children, and bilateral involvement does not reveal 'faint traces' of the disorder. They can use regular footwear and participate in physical activity classes. Publications in Russian and foreign literature report short-term follow-up within a year when ossification is not complete and we cannot radiographically judge aseptic iatrogenic osteonecrosis. Longer-term studies are necessary for the patient cohort.

CONCLUSION

Peritalar soft-tissue release and arthrotomy often result in poor circulation of the talus due to specific features of blood supply to the bone and can cause development of deforming osteoarthritis. The condition is likely to lead to lower health-related quality of life scores with weight gain during growth. This can reveal in a delayed manner due to evolution of ossification centers in the hind- and midfoot. That is why the Dobbs method like the Ponceti method of clubfoot care is clinically important. Good outcomes achieved with the practice require a thorough observation of children with genetic and neuromuscular disorders, in particular, to avoid additional interventions with lost correction. Children require observation after the treatment and it is evident that the rate of primary open reduction of the talus has dramatically reduced with AOFAS scores improved.

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