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TREATMENT OF CONGENITAL FOOT DEFORMITY IN CHILDREN

DOI 10.25789/YMJ.2020.72.11

Foot deformities in children with no proper correction are often accompanied by pain, functional changes and high risk of developing disability, which determines the high social significance of the nosology. The aim of this study was to evaluate the effectiveness of treatment of children's foot deformities in outpatient and inpatient settings. In the period from 2015 to 2020 109 children were examined and treated at the clinical base of the Moscow City Children's Clinical Hospital named after N.F. Filatov. The results of the study showed that in all children after the comprehensive assessment and the combination of conservative and operative correction techniques, there was complete elimination of congenital foot deformity. To achieve the complete and effective res equino-varus correction, the Ponseti procedure must be carefully followed. The early detection and correction of foot deformity is effective.

Keywords. clubfoot, adducted foot, vertical ram, congenital foot deformity, Ponseti procedure, pes equino-varus, metatarsus varus, vertical talus, pes varus, pes planovalgus, pes cavus.us.

Introduction. Pathology of the foot of congenital etiology is represented by such nosologies as Pes equino-varus (clubfoot), metatarsus varus (reduced foot), vertical talus (vertical RAM), pes varus (varus foot), pes planovalgus (flat foot), pes cavus (hollow foot). According to ICD-10 code Q66.5. The epidemiology of pes equino-varus is 1 per 1000 newborns [4], while vertical talus and metatarsus varus are quite rare [3, 8].

These nosologies are accompanied by a pronounced pain syndrome, functional changes in foot, which forces the patient to use orthopedic shoes. In the absence of proper surgical correction, the risk of disability is high. Functional disorders affect the patient's quality of life and determine the high social significance of these nosologies [5].

To date, there are a number of classifications of congenital foot pathology. According to Zatsepin-Bohm, there are two clinical forms of Pes equino-varus: typical and atypical [6]. Based on the literature available to us, the typical type of deformation accounts for 80% of cases. This type of deformity lends itself well to such treatment methods as bandaging and plaster casting.

There are also three types of soft tissue component involvement - soft tissue and bone (rigid). Belonging to a particular type of pathology is distinguished by the possibility and effectiveness of a conservative method of treatment. A number of soft-tissue types of deformations are described in the literature as the most common [1].

The aim of the study was to improve the results of treatment of pes equino-varus using the Ponseti procedure, as well as vertical talus correction by Dobbs in children in combination with massage, physiotherapy and physical therapy.

Materials and methods. In the period from 2015 to 2020, a double prospective cohort study was conducted at the clinical base of the Moscow state medical UNIVERSITY named after N. F. Filatov. 109 children with congenital deformities of the feet were selected for treatment with the proposed methods.

During the examination, 102 children (93.6%) were diagnosed with a typical and 7 (6.4%) with an atypical form of pes equino-varus. The soft tissue form was found in 51.4% of cases (in 56 children), and in 48.6% - the bone form (53 children). In 22.0% of cases, we found a left - sided type of deformity (24 children), in 18.3% - a right-sided type (20 children), and in 59.6% of cases (65 children), a bilateral lesion.

According to the age at which the deformity was detected, the patients were distributed as follows. In 73.4% of cases, deformity was diagnosed before 3 months (80 children), in 6.4% of cases from 3 to 6 months – (7 children), in 20.2% of cases over the age of 6 months (22 children). The average start time of clinical follow-up was 1.0 (1.0; 3.5) months. The average start time of treatment was 1.0 (1.0; 4.0) months. The average duration of surgical intervention was 3.0 (2.0; 4.25) months.

Surgical correction was performed in 100% of cases (91 children) with pes equino-varus and in 50% of cases (3 children) with vertical talus. Metatarsus varus

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in 100% of cases were subjected to conservative treatment. Surgical treatment was performed in 94 children (achillotomy was performed in 91 children with pes equino-varus and 3 with vertical talus).

All children with PES equino-varus and 11 (91.7%) of 12 children with metatarsus varus used the Ponseti procedure. This is a conservative technique of plastering congenital clubfoot, which consists in gradually removing all components of the deformity to the correction position, based on the biomechanics of the ankle joint and supplemented by percutaneous achillotomy.

All children with vertical talus had the Dobbs technique applied. This is a conservative technique of plaster cast for congenital equinovalgus deformity of the feet, which consists in gradually removing all components of the deformity to the correction position, based on the biomechanics of the ankle joint, supplemented by percutaneous achillotomy and in some cases fixing the 1st leg of the foot with a Kirschner spoke.

Complex treatment of children with metatarsus varus included massage procedures. Also, 5 out of 6 children (83.3%) with vertical talus had massage. Children with pes equino-varus were not given massage treatments. Courses of physiotherapy procedures were used in 16.7% of children with metatarsus varus (2 out of 12). Complex physical therapy sessions with metatarsus varus were conducted in 70.3% of cases (64 children out of 91) and in 33.3% of cases with vertical talus (4 out of 6 children). Children with pes equino-varus did not receive comprehensive physical therapy classes.

Research results. Criteria of effectiveness of treatment was: emptiness of the heel, the degree of rigidity of the Cavus the medial folds form the lateral bending of the arch of the foot, its aquinos and degree of dorsiflexion. Changes of the foot were determined according to the classification Pirani:

1. the condition of the posterior part of the foot according to Pirani before correction had more pronounced statistical differences than after correction (according to the Wilcoxon criterion = -8.955, p<0.001);

2. the degree of Cavus rigidity according to the piranha classification before correction had more pronounced statistical differences than after correction (Wilcoxon criterion = - 9.125; p<0.001);

3. Assessment of the medial fold of the foot before correction had more pronounced statistical differences than after correction (Wilcoxon criterion = - 9.105; p<0.001);

4. the Bending of the outer edge of the foot before correction had more pronounced statistical differences than after correction (Wilcoxon criterion = - 9.364; p<0.001);

5. the Equinus of the foot before correction had more pronounced characteristics than after correction (Wilcoxon criterion = - 8.879; p<0.001);

6. The evaluation of the posterior heel fold according to the Pirani system before correction had more pronounced statistical differences than after correction (Wilcoxon criterion = - 8.791; p<0.001).

The total number of points according to the piranha criteria before the correction was 4.5 (3.0; 6.0), after the correction 0 (0;0) points. The obtained differences are statistically significant (we used Friedman's analysis of variance for related samples, p<0.001).

In our study, 61 patients received outpatient surgical treatment and 45 patients received inpatient treatment (n = 106).

The results of outpatient surgical treatment. Changes in the foot according to the piranha classification were distributed as follows:

1. heel Emptiness according to the Pirani classification before correction revealed statistically significant differences than after correction (Wilcoxon criterion = -6.705, p<0.001, 1.0 (0.5; 1.0) before correction versus 0.0 (0.0; 0.0) after correction);

2. the rigidity of the Cavus according to the Pirani classification before correction had more pronounced statistical differences than after correction (Wilcoxon criterion = - 6.628; p<0.001, 1.0 (0.5, 1.0) before correction versus 0.0 (0.0; 0.0) after correction);

3. Assessment of the medial fold of the foot before correction had more pronounced statistical differences than after correction (Wilcoxon criterion = - 6.628; p<0.001, 1.0 (0.5, 1.0) before correction vs. 0.0 (0.0; 0.0) after correction);

4. the Bending of the outer edge of the foot before correction had more pronounced statistical differences than after correction (Wilcoxon criterion = - 6.683; p<0.001, 1.0 (0.5, 1.0) before correction vs. 0.0 (0.0; 0.0) after correction);

5. Equinus of the foot before correction had more pronounced statistical differences than after correction (Wilcoxon criterion = - 6.753; p<0.001, 1.0 (0.5, 1.0) before correction vs. 0.0 (0.0; 0.0) after correction);

6. The assessment of the back heel fold according to the Pirani classification before correction had more pronounced statistical differences than after correction (Wilcoxon criterion = - 6.662; p<0.001, 1.0 (0.5, 1.0) before correction versus 0.0 (0.0; 0.0) after correction).

Thus, the total score for the piranha classification before correction was 5.0 (4.0; 6.0), after correction 0 (0;0) points. The obtained differences are statistically significant (we used Friedman's analysis of variance for related samples, p<0.001).

The results of hospital surgical treatment. Changes in the foot according to the piranha classification were distributed as follows:

1. heel Emptiness according to the Pirani classification before correction had more pronounced statistical differences than after correction (Wilcoxon criterion = -5.665, p<0.001, 1.0 (0.5;1.0) before correction versus 0.0 (0.0; 0.0) after correction);

2. the rigidity of the Cavus according to the Pirani classification before correction had more pronounced statistical differences than after correction (Wilcoxon criterion = - 5.557; p<0.001, 1.0 (0.5, 1.0) before correction versus 0.0 (0.0; 0.0) after correction);

3. Assessment of the medial fold of the foot before correction had more pronounced statistical differences than after correction (Wilcoxon criterion = - 5.516; p<0.001, 1.0 (0.5, 1.0) before correction vs. 0.0 (0.0; 0.0) after correction);

4. the Bending of the outer edge of the foot before correction had more pronounced statistical differences than after correction (Wilcoxon criterion = - 5.631; p<0.001, 1.0 (0.5, 1.0) before correction vs. 0.0 (0.0; 0.0) after correction);

5. Equinus of the foot before correction had more pronounced statistical differences than after correction (Wilcoxon criterion = - 5.674; p<0.001, 1.0 (0.5, 1.0) before correction vs. 0.0 (0.0; 0.0) after correction);

6. the evaluation of the posterior heel fold according to the Pirani classification before correction had more pronounced statistical differences than after correction (Wilcoxon criterion = - 5.631; p<0.001, 1.0 (0.5, 1.0) before treatment versus 0.0 (0.0; 0.0) after treatment).

Thus, the total score for the piranha classification before correction was 5.5 (4.0; 6.0), after correction 0 (0;0) points. The differences are statistically significant (two-factor Friedman analysis for related samples, p<0.001).

Comparison of inpatient and outpatient treatment groups. Prior to treatment, the inpatient and outpatient treatment groups were comparable in all piranha classification criteria:

1. The emptiness of the heel (the Mann-Whitney test, p=0,466);

2. Cavus Rigidity (Mann-Whitney test, p=0.611);

3. Medial fold of the foot (Mann-Whitney test, p=0.986);

4. Bending of the outer edge of the foot (Mann-Whitney test, p=0.978);

5. Equinus of the foot (Mann-Whitney test, p=0.663);

6. Back heel folds (Mann-Whitney test, p=0.671).

By total score (Mann-Whitney test, p=0.917). Thus, the groups are comparable to each other in terms of these indicators. Based on the criteria for the effectiveness of treatment according to the Pirani classification, it is possible to compare clinical comparison groups by the degree of dorsiflexia achieved.

Achieved dorsiflexia greater than 15 degrees was observed in 51 cases of surgical treatment (83.6%) in outpatient settings and in 39 cases (86.6%) of surgical treatment in inpatient settings (table 1).

Table 1 – Achieved dorsiflexia in comparison groups.

The table shows that the differences between the groups are statistically insignificant (Fisher's exact test, exact significance (2-sided) = 0.139). By total score (Mann-Whitney test, p=0.917). Thus, the groups are comparable to each other in terms of these indicators.

Adverse outcomes of inpatient and outpatient treatment. After surgical correction in a hospital setting, one child required repeated surgery due to a relapse (an additional achillotomy was performed). Based on our experience and the literature we have studied, early detection of relapses of pathology is the key to successful elimination of secondary deformity. The cause of secondary deformity is usually a violation of the rules for using rehabilitation correctors, braces, and orthopedic shoes after the main stage of surgical correction is completed. Relapse is usually detected during the period of intensive foot growth-up to 10-13 years of age. Therefore, at the beginning of adolescence, such children should be regularly monitored by an orthopedist [10].

In 9.1% of outpatient cases (5 out of 55 children), children had limited movement in the distal part of the lower leg, while the same complication in the hospital was observed in 2.6% (1 out of 39 children). There are no statistical differences in the compared groups (Fisher's criterion, exact significance (2-sided) = 0.395).

Thus, both outpatient and inpatient treatment options for children with foot pathology had an equally significant impact on the evaluation criteria for treatment effectiveness. In 100% of cases of operative correction, satisfactory results were achieved. When choosing a treatment method (outpatient or inpatient), the principal criteria should be considered not only the degree of social adaptation of the patient, but also economic factors, since the clinical effectiveness of these treatment approaches was the same.

Discussion. In modern pediatric orthopedic practice, PES equino-Varus correction using the ponseti method is the "gold standard" of treatment. To achieve complete successful correction of PES equino-Varus with the prevention of relapses or other deformities, careful compliance with the ponseti Protocol is necessary. Initially, the ponseti procedure was used only in children under two years of age, but current research on the results of PES equino-Varus correction is already focused on older age groups of children [9].

Our research results are consistent with the data obtained by other authors. The Ponseti procedure is successful and relapse-free in 94-96% of cases [7].

We believe that the most preferable age for correction of deformities is early age and adhere to the position that it is necessary to start correction of deformities early (immediately after diagnosis). Based on the literature available to us, late initiation of treatment is directly proportional to the frequency of relapses and duration of treatment [2].

Conclusion. Based on the data obtained, we recommend treating PES equino-Varus as early as possible after birth (3-5 months), to prevent relapses and ensure complete correction of the deformity. Strict compliance with the Ponset Protocol is required to prevent relapses.

When correcting vertical scree, conservative correction in combination with minimally invasive surgical techniques can prevent the development of complications that were previously observed during extensive surgical procedures.

The method of Dobbs correction used by us is simpler and more effective in young children. Our data are consistent with reports of excellent results from other authors. The Dobbs correction method is less invasive and avoids the risks associated with more extensive operations [11].

We did not find any significant differences in the choice of outpatient or inpatient treatment. Taking into account the economic factor, in conditions of statistically reliable identical clinical outcomes, outpatient treatment is most preferable.

Conflict of interest. The authors declare that there is no conflict of interest.

Achieved dorsiflexia in comparison groups

			The comparison group	
			Outpatient n=61	Stationary n=45
Achieved dorsiflexia	<15°	Number, people	4	6
		Frequency, %	6.4	3.4
	> 15°	Number, people	57	39
		Frequency, %	83.6	86.6

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CLINICAL – AUDIOLOGICAL AND CLINICAL - GENEALOGICAL ANALYSIS OF CASES OF HEARING LOSS IN THE REPUBIC OF BURYATIA

DOI 10.25789/YMJ.2020.72.12

In this paper we presented for the first time the results of the clinical-audiological and clinical-genealogical research of hearing impairments in the Republic of Buryatia. The sample included Buryats (47.9%), Russians (46.1%) and representatives of other ethnicity (6%), amounting 165 people. As the result of the clinical-audiological analysis, 70.3% (n=116) of individuals had bilateral deafness of the sensorineural type, and 29.7% (n=49) had bilateral hearing loss of varying severity. The segregation analysis was carried out in 17 Buryat and 18 Russian families made it possible to assume the hereditary nature of cases of hearing loss, segregating according to the autosomal recessive mode of inheritance only in Russian families (segregation frequency SF = 0.25, at t = 0.64). The frequency rate of segregation (SF = 35, at t = 0.38) of the pathological trait in Buryat families turned out to be higher than theoretically expected for the autosomal recessive type of inheritance (SF_0 = 0.25), which indicates the presence of other types of inheritance and other forms of hearing impairments caused by non-hereditary reasons. The results of this study and the expeditionary material will be the basis for further study of the molecular genetic etiology of deafness/hearing loss in Buryatia.

Keywords: clinical-audiological analysis, clinical-genealogical analysis, hearing impairment, hereditary burden, Republic of Buryatia

Introduction. For the majority of hereditary diseases associated with organs of hearing, a large number of genes have been identified with a significant variety of mutations contributing to their development [6, 9-11; 13, 14, 18, 22, 25-29, 31], also regional and ethnic differences in the spectrum and frequencies of identified mutations have been manifested [3, 7, 15, 19-23, 30, 36]. Hereditary hearing impairments (HI) are genetically heterogeneous and manifest with different penetrance, which requires a special approach to the development of molecular diagnostics methods for genetically different forms of deafness [12, 24]. Recently, a significant number of works have been published on the successful identification (using various strategies of WES - exome sequencing) of genetic factors leading to hearing loss (HL) and the list of genes associated with hearing loss is constantly expanding (Hereditary Hearing Loss Homepage: http://hereditaryhearingloss.org). To search for the molecular genetic causes of rare forms of deafness in humans at the first stage of research, a thorough clinical and genealogical analysis of families of deaf people with large pedigrees is required.

It is known that the accumulation of a rare genetic disease due to the founder effect can occur in small isolated human populations. Most of the genes associated with one or another rare genetic disease, including those associated with hearing impairment, were first identified in families with large branched pedigrees with numerous affected individuals from isolated populations with a high endogamy index (Ashkenazi Jews, Finns, Sami, as well as inbred families from the Middle East and South Asia) [4, 8, 11]. In such populations, there is high probability of detecting new genes for human mendelian diseases. In Russia, the study of the fundamental foundations of rare (monogenic) human diseases can be carried out using the example of endogamous populations of the peoples of the Caucasus, the Volga-Ural region, Siberia and indigenous peoples of the North.

As a result, the studies of congenital forms of deafness (as one of the most frequent mendelian human diseases) in poorly studied regions of the world, such as the territory of Siberia, are especially relevant. Earlier, according to the contribution of *GJB2* gene mutations (Cx26) among samples of patients with hearing impairments from Siberian regions, the following were described in detail: the Altai Republic [16], the Sakha Republic (Yakutia) [5, 33] and the Tyva Republic [2, 34, 37]. The study of hereditary deafness in the Republic of Buryatia is a logical continuation of research among the populations of Siberia, which makes it possible to close many “blank spots” concerning the issues of genetic epidemiology of hereditary forms of deafness.

The aim of this study is to conduct audiological and clinical-genealogical analysis of the families with hearing impairment in the Republic of Buryatia, which will serve as the basis for further study of the molecular genetic etiology of HI in peoples of Eastern Siberia.

Materials and methods.

The patients

During the expedition work of the Yakutsk Scientific Center for Complex Medical Problems (Yakutsk) in the Republic of Buryatia 165 (n=160 unrelated) deaf

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