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MICROSURGICAL TOE-TO-HAND TRANSFER IN CHILDREN WITH MACRODACTYLY OF THE HAND

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Background. At present, there are multiple approaches to the treatment of macrodactylia in children. Additionally, there are no comparative data on the cosmetic and functional aspects of corrective surgery versus microsurgical transplantations performed in children with a marked disproportion of the segment, making this study particularly relevant.
Aim. The aim of this study was to determine the role of microsurgical transplantation of the toes in the treatment of children with isolated macrodactylia, based on a comparative analysis of various surgical interventions.
Materials and methods. Twenty-five children with congenital macrodactylia of the hand were examined and surgically treated in the department of reconstructive microsurgery from 2013 to 2017. Of these, 13 (52%) had macrodactyly of the 2nd and/or 3rd fingers, and of the total number of hyperplastic hand segments (n = 45), 17 rays had hyperplasia greater than 25%.
Results and discussion. Stage-by-stage modeling resections of enlarged segments were performed in all children with ray hyperplasia that was about 25% of the size of intact fingers. In 4 cases, excision of soft tissues was combined with marginal resection of phalanges. When hyperplasia of the segment reached up to 10% of macrodactylia, single isolated modeling resection of soft tissues and bones was performed. Repeated modeling plasties in 16% (n = 4) of the children were accompanied by gross postoperative scars with the development of secondary angular deformities and loss of function of interphalangeal joints.
Nine children (n = 14) underwent amputation of hyperplastic fingers followed by microsurgical reconstruction of rays by autografting of toes. In 4 cases, the first finger was reconstructed, and in the other 5 cases, toes of both feet were transplanted to the positions 2 and 3 (n = 4) or 3 and 4 of fingers (n = 1). It was found that in cases of significant hyperplasia of the affected segments of the hand (hyperplastic segment is 1.5–2 times greater compared to normal ones), microsurgical autografting of toes is more effective and acceptable compared to bone and soft tissue resections.
Conclusions. In cases of segment hyperplasia less than 1.25-times the normal size, the optimal surgical interventions are stage-by-stage modeling resections. An alternative surgical treatment for children with hyperplasia of more than 1.25-times the normal size is microsurgical transplantation of the toes in the position of the involved rays of the hand, which results in good cosmetic and functional outcomes in these patients.

Keywords: microsurgery; toe-to-hand transfer; macrodactyly.

АСПЕКТЫ ИСПОЛЬЗОВАНИЯ МИКРОХИРУРГИЧЕСКОЙ АУТОТРАНСПЛАНТАЦИИ ПАЛЬЦЕВ СТОП НА КИСТЬ У ДЕТЕЙ С МАКРОДАКТИЛИЕЙ ВЕРХНЕЙ КОНЕЧНОСТИ

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Актуальность. В настоящее время не существует единого алгоритма при лечении макродактилии у детей, отсутствуют сравнительные данные о «косметичности» и функциональности проводимых корректирующих операций у детей с выраженной диспропорцией сегмента по сравнению с микрохирургическими трансплантациями, что обусловливает актуальность данного исследования.
Цель исследования — определить значение микрохирургической аутотрансплантации пальцев стоп в лечении детей с изолированной макродактилией на основании сравнительного анализа различных видов оперативных вмешательств.
Материалы и методы. В отделении реконструктивной микрохирургии с 2013 по 2017 г. находилось на обследовании и хирургическом лечении 25 детей с врожденной макродактилией кисти. Из 25 пациентов у 13 имела место макродактилия 2-го, 3-го пальцев, что составило 52 %. При этом из общего количества гиперплазированных сегментов кисти (n = 45) 17 лучей имели гиперплазию больше 25 %.

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Результаты и обсуждение. Этапные моделирующие резекции увеличенных сегментов выполняли всем детям с гиперплазией луча около 25 % от размера интактных пальцев, в четырех случаях иссечение мягких тканей сочеталось с краевой резекцией фаланг. При гиперплазии сегмента до 10 % проводили изолированные однократные моделирующие мягкотканно-костные резекции. Повторные моделирующие пластики у 16 % (n = 4) детей сопровождались образованием грубых послеоперационных рубцов с формированием вторичных угловых деформаций и потерей функций межфаланговых суставов.

Девяти пациентам (n = 14) произвели ампутацию гиперплазированных пальцев с последующей микрохирургической реконструкцией лучей методом аутотрансплантации пальцев стоп. В четырех случаях была выполнена реконструкция первого пальца, еще в 5 — пересадка двух пальцев обеих стоп в позицию 2-го, 3-го (n = 4) и в позицию 3-го, 4-го пальцев (n = 1). Установлено, что при значительной гиперплазии пораженных сегментов кисти (1,5–2 раза) метод микрохирургической аутотрансплантации пальцев стоп более эффективен и лучше подходит по сравнению с методом костных и мягкотканных резекций.

Выводы. При гиперплазии сегмента менее четверти от нормальных размеров оптимальными оперативными вмешательствами являются этапные моделирующие резекции. Альтернативным методом оперативного лечения детей с гиперплазией более 25 % служит микрохирургическая трансплантация пальцев стоп в позицию вовлеченных в процесс лучей кисти, что позволяет получить у данных пациентов хороший косметический и функциональный результат.

Ключевые слова: микрохирургия; аутотрансплантация пальцев стопы на кисть; гигантизм.

Background

Macrodactyly or gigantism is a rare congenital malformation characterized by a significant increase in the longitudinal and transverse dimensions of the upper limb soft tissues and bone structures [1, 2]. According to modern sources, macrodactyly occurrence is 0.5%–1% among all cases of congenital hand pathology [1–4].

Etiologically, it is now customary to distinguish isolated macrodactyly from macrodactyly, which is part of a genetic syndrome (Proteus syndrome, Beckwith-Wiedemann syndrome, CLOVES-syndrome, etc.). According to J. Hardwicke and R. Lester [1], the majority of isolated, non-syndromic macrodactyly do not have a known genetic etiology that is recorded in the oMIM database [1, 5].

Since the first description of this macrodactyly by the English physician John Locke in 1675, there have been many attempts to create a single classification that will comprehensively describe both the clinical and morphological picture and the progression of the defect [1–5]. Nowadays, the most complete classifications are based on the original work of Kelikian (YEAR) in the modifications of Dell, Flatt, and Upton [6–12]. The last classification by Oberg, Manske, and Tonkin was presented in the format of *dysplasia — hypertrophy — macrodactyly or dysplasia — hypertrophy — upper limb* and *macrodactyly*, which enables classification of patients depending on the axis of formation/differentiation and limb segment [1]. Accordingly, it was proposed in 2015 to combine and supplement existing classifications to improve the understanding of the prognosis and the optimization of treatment tactics (Table 1).

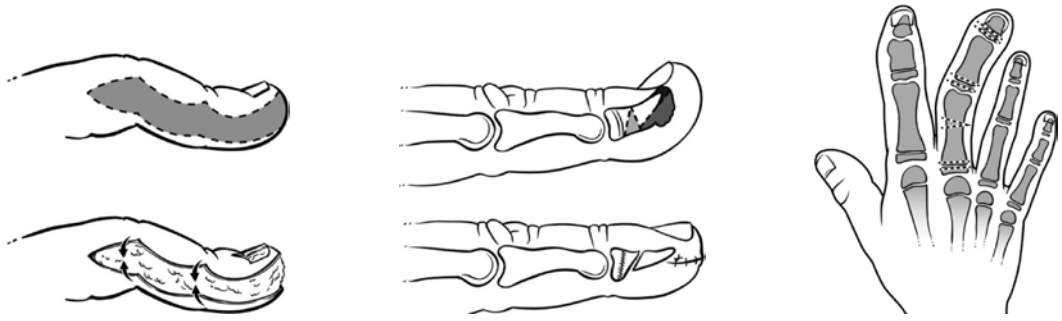
Lipomatous macrodactyly is the most common form of macrodactyly and differs from the nerve-associated macrodactyly by lack of involvement of the digital nerves and vascular bundles. The term "nerve-associated macrodactyly" was first introduced by Kelikian (year) in order to emphasize the connection between the increase in nerve and musculoskeletal structures. This type refers to isolated macrodactyly, and in 90% of cases it is unilateral. In this article, we will consider treatment options for patients with this macrodactyly variant.

Surgical treatment of macrodactyly depends on the need for correction of the tissues involved,

Table 1

Integrated system of macrodactylia classification

Characteristics	Classification			
Height	*Static*		*Progressive*	
Form	Isolated		Syndromic	
Cause	Lipomatosis	Nerve hyperplasia	Hyperostosis	Vascular malformations



a *b* *c*

Fig. 1 J.S. Gluck, M. Ezaki. (YEAR) Scheme of lateral resection of soft tissue (*a*), corrective osteotomy (*b*), and epiphysiodesis (*c*) with finger macrodactyly [3]

namely the excision of soft tissues, corrective osteotomies, epiphysiodesis, etc. In an article by J.S. Gluck, M. Ezaki (YEAR), the degree of increase in the segment is proposed to be evaluated relative to the similar finger on the parent of the same sex as the child [3]. Accordingly, the choice in surgical treatment option depends on the degree of the segment increase. If the child's fingers are smaller or the same size as the parent's fingers, it is recommended to perform stepwise soft tissue and bone resections, but only if segment function is preserved. If the fingers of the child are significantly larger than the fingers of the parent, i.e., if they are disproportional and do not have adequate function, then it is necessary to perform amputation without subsequent reconstructions. (CITATION)

This surgical technique also depends on the severity of the hyperplastic segment. With an increase in subcutaneous fatty tissue predominantly on the palmar surface of the nail phalanx, the authors recommend excision of the skin and soft tissues with an ellipsoid incision in the center of the palmar surface of the phalanx, which enables the maintenance of innervation and sensitivity at the fingertip. It is also noted that the ellipsoid incision should not reach the level of the interphalangeal joint so as to avoid secondary cicatricial contractures. This surgical intervention is a good method for prevention of ingrowth of the nail plate in the hyperplastic periungual folds.

In the presence of severe hyperplasia of soft tissues and bone structures, or the spread and asymmetry of the process, M. Ezaki et al. (year) in their study performed excision of soft tissues on the side most increased. To do this, a shaped incision on the mediolateral surface was performed with the selection and excision of the digital nerve and branches, which can slow down the process of continued growth according to the etiology of the development of the process. It was noted that the restoration of sensitivity of most of the remaining soft tissues and skin is restored due to hyperplasia of the contralateral nerve. However, removal of nerves from both sides is impractical due to the subsequent deterioration of its functionality. As soon as the size of the child's finger approaches that of the same finger of the parent, epiphysiodesis is recommended to halt longitudinal growth. Epiphysiodesis is performed with a midline incision along the lateral surface with a Kirschner wire or with a drill equal to the width of the growth zone and, as a rule, is combined with resection of soft tissues according to the procedure described above. In the presence of pronounced angular deformities, surgical treatment is supplemented with corrective osteotomies (Fig. 1).

These surgical techniques, which include partial or complete removal of the distal and middle phalanges and central resection of the nail phalanx, consistently lead to joint stiffness and severe loss of function, which is an unsatisfactory treatment outcome (CITATION). If the hyperplastic finger (at an early age) exceeds the size of an adult, or if it markedly limits the use of the other healthy fingers and consequently reduces the function of the hand, segment amputation without subsequent reconstruction is performed. The authors do not exclude the possibility of reconstruction of the first finger using microsurgical autografting of the toe, but consider it impractical, because segment hyperplasia is possible due to the preservation of activity of diffuse growth factors according to disease pathogenesis. In these cases, preservation of the first metacarpal bone with subsequent lengthening and reconstruction of the interdigital space is suggested.

According to I.V. Shvedovchenko, O.N. Sosnenko and others, the main modeling surgeries used in

macrodactyly are surgeries aimed at reducing the length/volume of the affected segment. Modeling resections are aimed at the simultaneous reduction of both length and volume of the finger, and surgeries have aimed to eliminate concomitant deformities. When the segment is enlarged by more than 300% relative to normally formed rays, microsurgical autografting of the toe to the position of the removed hyperplastic segment is indicated, which can significantly improve hand function and the possibility of self-service in the future [13–16].

P. Cavadas, J. Hardwickeand, R. Lester et al. (YEAR) in their study also believe that amputation does not sufficiently improve the functional and cosmetic condition of the hand, and thus have suggested performing microsurgical grafting of the toe to the position of the resected finger. Accordingly, only one toe was transposed in these clinical studies regardless of the number of affected segments on the hand [1, 17].

Thus, we can conclude that, at present, there is no unified consensus for treating macrodactyly in pediatric patients. Accordingly, there are no comparative data on the "cosmesis" and functionality of corrective surgeries in pediatric patients with severe segment disproportion as compared with microsurgical grafting.

Therefore, **this study**, based on a comparative analysis of various types of surgical interventions, aimed to determine the value of microsurgical autografting of toes in the treatment of children with isolated macrodactyly.

Materials and methods

We examined and surgically treated 25 pediatric patients (12 boys and 18 girls) with congenital macrodactylia of the upper limbs from 2013 to 2017 in the department of reconstructive microsurgery. All patients voluntarily signed an informed consent to participate in the study. The patients' ages at the time of first admission to the clinic were 16 ± 2 months and were directly dependent on the degree of deformity; the greater the deformity, the earlier it was necessary to perform a surgical intervention. Out of 25 patients with partial gigantism, 13 patients had macrodactyly of fingers two and three, accounting for 52% of cases. In another four pediatric patients, macrodactylia of fingers one and two was clinically determined (16%), and three patients had gigantism of fingers three and four. Isolated lesions of fingers two and five were noted in isolated cases. Hyperplasia of fingers four and five was detected in 8% ($n = 2$) of cases. Out of 45 hyperplastic hand segments, 17 rays had hyperplasia of more than 25%. With a pronounced imbalance of the ray, we clinically noted gross changes in the interphalangeal joints (IPJ) in the form of their extensor position and noted also a sharp restriction of passive and active movements with a total volume of up to 15°.

Results and discussion

Staged modeling resections of soft tissues of enlarged segments were performed for all pediatric patients with ray hyperplasia, which constitutes about a quarter of the size of intact fingers. In four cases, the excision of soft tissues was combined with marginal resection of the phalanges. Repeated modeling grafting in 16% ($n = 4$) of pediatric patients was subsequently accompanied by the formation of coarse postoperative scars with the formation of secondary angular deformities and loss of function of the IPJ. In this connection, at the first state, resection of the nail phalanx in the distal section was performed to improve the cosmetic and functional result. Also, shortening and correcting resections of the middle and main phalanges were performed at the level of the middle third finger. In subsequent surgical interventions, phased resections of bone and soft tissues were performed mainly along the lateral and palmar surfaces of the fingers. However, performance of subsequent corrective surgeries had negative aspects as they were performed on scar-modified soft tissues in which neurovascular bundles were sealed; this later caused vascular disorders and partial necrosis of the soft tissue segments in eight cases (32%). The lateral resections of the phalanges of the hyperplastic fingers in turn affected the ligament-capsule apparatus, which led to the deterioration in the IPJ function in 11 patients.

Isolated single modeling soft tissue and bone resections were performed in the case of hyperplasia of the segment at up to 10% in stable-type macrodactyly. Monitoring at various times after surgical treatment showed good cosmetic and functional results (7.25%).

Based on the above experience, it can be concluded that a number of staged surgeries are necessary for

hyperplasia of a segment of more than a quarter of normal size, but are impractical due to multiple concomitant complications and unsatisfactory cosmetic and functional results. Therefore, an amputation without subsequent reconstruction was performed on one child with macrodactylia of the ray 5 and on two pediatric patients with ulnar form of macrodactyly (rays 4 and 5), due to the minimal functional load of these segments.

In the remaining cases ($n = 9$, $\Sigma = 14$), we performed the amputation of hyperplastic fingers followed by microsurgical reconstruction of the rays using the autografting of the toes. In four cases, the first finger was reconstructed, and in five more cases, two toes of both feet were transplanted to the position of the second, third ($n = 4$) fingers, and the third, fourth fingers ($n = 1$). Two pediatric patients underwent microsurgical grafting to restore the second and third fingers, respectively.

These surgeries had technical aspects compared with standard microsurgical grafting in case of other congenital and acquired defects. Thus, in all patients there were hypoplastic common palmar digital arteries, as well as their branched type of structure as compared with the normal segment anatomy. This necessitated the connecting of the grafted vessels more proximally, namely at the level of the wrist joint. A positive factor of the technical aspects was the possibility of preserving the flexor tendon ligaments during reconstruction, which enabled us to further reduce the amount of tenolysis and improve the functional result. In all cases, during the revision, we noted significant hyperplasia and tortuosity of the median nerve, which was also left intact.

During autografting of toe 1, selection of all the necessary structures did not differ from the standard technique adopted in the department. The incision was made on the dorsum of the left foot in the projection of the intermetatarsal space 1, with a continuation on the plantar surface through interdigital spaces 1 and 2. The great saphenous vein was isolated and all branches were coagulated to toes 1 and 3. Then, the dorsal artery of the foot and its continuation were isolated in the form of the first dorsal and one or two of the plantar metatarsal arteries, depending on the variant of the vascular structure of the foot. The flexor tendon, extensor tendon, and plantar digital nerves were also isolated and transected. After osteotomy of metatarsal bone 2 under the epiphysial plate (or capsulotomy of the metatarsophalangeal articulation 2), the toe was separated from the foot. The nutrient artery and vein were not cut off, and after removing the tourniquet, restoration of blood circulation in the isolated graft was evaluated. Microcirculation of blood in the graft was maintained for 20–30 min to minimize time of ischemia.

On the hand, the second team of surgeons made fringing incisions at the level of the main phalanx of the hyperplastic finger. After isolating the vessels, nerves, and tendons of the finger, it was resected (usually at the level of the proximal or middle third of the main phalanx). On the dorsum of the hand, the extensor tendon and the venous branch were isolated. On the palmar surface of the hand, the digital nerves were isolated and were always greatly enlarged in length and thickness, as well as the flexor tendon of the removed finger. Considering the diameter and the loose type of the structure of the peripheral vessels in macrodactyly, we always attempted to proximally isolate the recipient artery at the level of the wrist joint or even the forearm. Usually, a radial or ulnar artery was used, but in three cases, the palmar superficial arterial arch was used. After removing the tourniquet from the shoulder, hemostasis was performed.

After transecting the nutrient vessels, the toe was transferred to the hand and osteosynthesis was performed with a wire along the axis. Then, anastomoses were placed on the flexor tendons, extensor tendons, and the digital nerves. After the clips were applied to the recipient vessels and their intersections, an anastomosis was applied end-to-end between the graft artery and the proximal part of the recipient artery as well as to the veins of the graft and hand. After removing the clips from the vessels, the rate of blood circulation restoration in the transplanted graft was assessed. Excess flaps of skin were then dissected, and sutures were placed on the wounds in layers (Figs. 2–4). Both limbs were immobilized in a plaster cast, which was left for four to six weeks, the optimal time to stabilize bone consolidation.

When two fingers were transplanted simultaneously from both feet, the toe transplant was isolated by the method described above. On the hand, the recipient bed was prepared similarly, but only for two toes (Figs. 5–8). However, there were some special aspects in the preparation of receptive vessels. Veins were more often prepared from different

a *b* *c*

Fig. 2. Macrodactyly of fingers 2–4 of the right hand in a child of two years: *a*, *b* — appearance before the surgery; *c* — radiograph of the hand

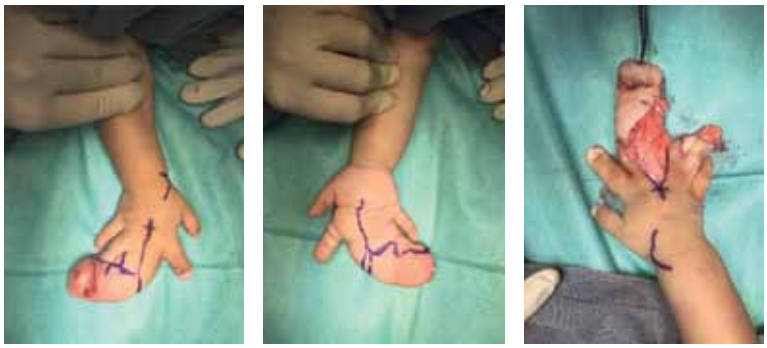


Fig. 3. Stages of surgical intervention

Fig. 4. The appearance of the hand and radiograph at the end of surgery

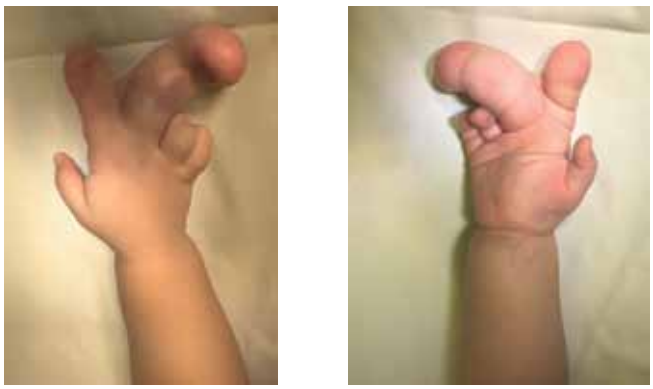


Fig. 5. Appearance of the hand of a child of 1.5 years of age having macrodactyly of the right hand

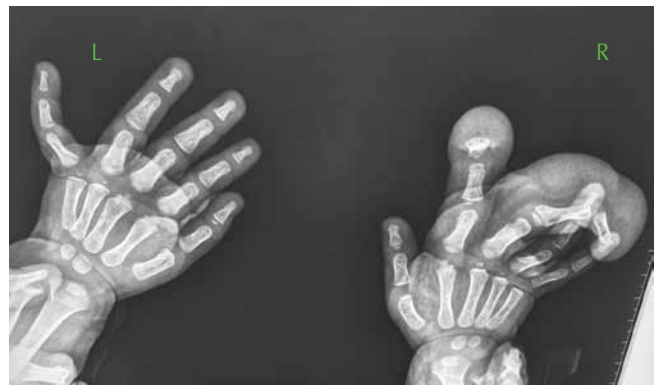


Fig. 6. Radiograph of the hand of a child of 1.5 years of age with macrodactyly of the right hand

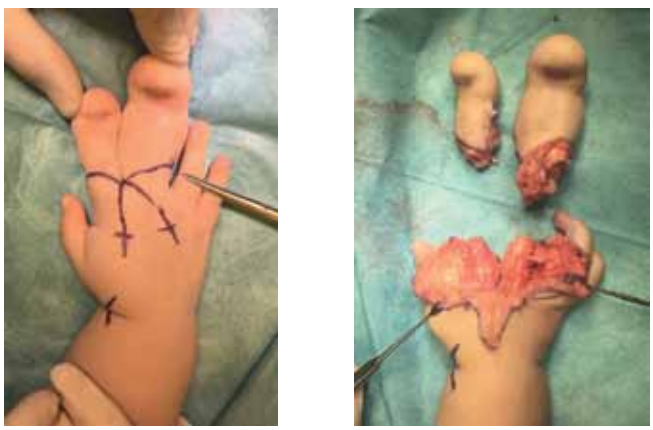


Fig. 7. Stages of surgery

Fig. 8. Appearance and radiograph by the end of the surgery

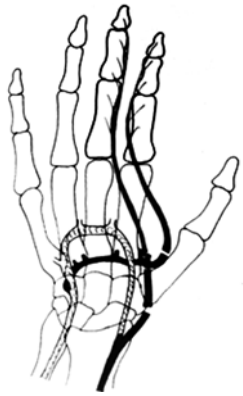


Fig. 9. Scheme of the donor arteries' connection of both grafts to the transected radial artery

venous systems, namely *v. cephalica* and *v. basilica*. Similarly, for the formation of graft nutrient arteries, one main artery (radial or ulnar) was usually used, and after transaction, two blood circulation sources were obtained for both grafts. Anastomoses were applied both with the proximal segment of the artery and with the distal segment of the artery (Fig. 9).

Despite the special aspects of the vascular structure of the hand, we observed no secondary vascular complications, and engraftment of the transposed segments was 100% successful. All the children in this group subsequently underwent corrective surgeries, namely modeling of soft tissues of the grafted fingers (elimination of syndactylia, excision of excess soft tissues) and tenolysis of flexor tendons.

The long-term results of finger grafting were analyzed by measuring the size of fingers and hand, assessing the appearance of the hand of parents or a patient, measuring passive and active finger movements in the metacarpophalangeal and IPJ. The capabilities of the hand were also determined by the main types of capture and also by using the ABILHAND questionnaire (in three older patients 9–13 years old). Previous work has established that with significant hyperplasia of the affected segments of the hand (more than a quarter as compared with normal finger sizes) of pediatric patients with congenital gigantism, the method of microsurgical "replacement" of fingers with toes is more effective and is better suited as compared with traditional methods of bone and soft tissue resections of hyperplastic segments. After grafting of toes, in 89% of cases parents were satisfied with both cosmetic and functional results of treatment. This was especially true of pediatric patients whose affected fingers were enlarged more than 1.5–2 times as compared with healthy fingers.

Therefore, this type of surgery improved the results of surgical treatment of this category of patients, and is an excellent and promising alternative for the method of stepwise resection of the affected fingers, especially in permanent removal of hyperplastic rays.

Conclusion

1) In the case of segment hyperplasia that is less than a quarter of normal size, optimal surgical interventions are stepwise modeling resections that are accompanied by bone resections and corrective osteotomies, where necessary.
2) Stepwise surgical interventions should be performed in the optimal ratio of result and number of surgeries, since each subsequent surgery increases the risk of development of secondary complications and deformities.
3) An alternative method of surgical treatment of pediatric patients with hyperplasia of more than 25% is microsurgical grafting of the toes to the position of the hand rays that are involved in the process. This enables good cosmetic and functional results in these patients.

Additional information

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Conflict of interest. The authors declare no obvious and potential conflicts of interest related to the publication of this article.

Ethical review. Patients (their representatives) gave their consent for the processing and publication of personal data.

Contribution of the authors

S.I. Golyana — development of the concept and design of the study, analysis of the data obtained, preparation of the text, and editing.

T.I. Tikhonenko — analysis of literary sources, material collection, analysis of the data obtained, statistical data processing, and text preparation.

A.V. Govorov — analysis of the data obtained and preparation of the text.

N.V. Zaitseva — analysis of the data obtained and editing.

All authors were directly involved in the implementation of surgical interventions.

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