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ARTICLE

Surgical treatment of macrodactyly of the foot in children

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ABSTRACT

The purpose of the study was to produce an algorithm and surgical procedure for optimum treatment of pedal macrodactyly. Surgery was performed on 27 feet of 26 patients with a mean age of 33 months at the time of surgery (range: 7–108 months). A multi-technique procedure based on the involved elements of the foot (soft tissue, phalanx, or metatarsal, or a combination of these) was adopted. The intermetatarsal width ratio, phalanx spread angle, and metatarsal spread angle was used to evaluate the severity of the macrodactyly and the effect of treatment. The Oxford Ankle Foot Questionnaire for Children and the Questionnaire for Foot Macrodactyly were employed to evaluate the clinical results. Under the guidelines of the treatment algorithm, all patients were successfully operated on using the multi-technique procedure; the sizes of the affected feet decreased significantly. After a mean of 33 months (range: 18–42 months) follow-up, the intermetatarsal width ratio decreased from 1.13 to 0.93 ($p < 0.05$), the phalanx spread angle decreased from 31.3° to 17.9° ($p < 0.05$), the metatarsal spread angle decreased from 33.2° to 15.8° ($p < 0.05$), and the mean score for the Oxford Ankle Foot Questionnaire for Children improved from 42 to 47 ($p < 0.05$) after surgery. The mean score for the Questionnaire for Foot Macrodactyly was 9.35 at the time of follow-up. The goal of the treatment of pedal macrodactyly is to obtain a functional and cosmetically acceptable foot. This treatment algorithm and multi-technique procedure could fully satisfy this goal.

Abbreviation: OxAFQ-C: the Oxford Ankle Foot Questionnaire for Children

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KEYWORDS
Macrodactyly; macrodactyly of the foot; pedal macrodactyly; surgical treatment

Introduction

Macrodactyly of the foot is an uncommon congenital malformation characterized by enlargement of all elements of the foot [1]. The condition usually results in a significant increase in the length, width, and height of the affected foot; this may impede the overall development of normal function and gait [2].

To date, surgical treatment is the only option for correction of the deformity [3]; the goal of treatment is to obtain, through a minimum number of operations, a pain-free, functional foot, with cosmetically acceptable digits, that tolerates shoe wear [2,4]. Several surgical interventions are usually needed, owing to the complicated situation and, because of the condition's rarity, the scarcity of surgical experience [5], especially with respect to repeated debulking [6,7]. It is reported that patients operated on undergo an average of 2.5 operations in total (range 1–6) [5]. Repeated surgical operations and anesthesia might cause damage to brain development [8,9], as well as psychological trauma for children [10,11].

Although many treatment options (e.g., soft tissue debulking, skeletal reduction [12], physeal resection or epiphysiodesis [13], reduction syndactyly, assorted osteotomies, toe amputation [14], ray resection [15], and interphalangeal and metatarsophalangeal joint arthrodesis) and procedures (e.g., the Tsuge procedure, a method of excision of the distal phalanx using a dorsal flap while preserving the nail [6,16], and the Barsky procedure, a method of reducing the length of the digits by ablation of the distal interphalangeal joint, sometimes in combination with epiphysiodesis [17]) have been proposed, results have frequently been suboptimal or unpredictable, owing to a lack of guidance for surgical decision-making. Inappropriate selection of surgical treatment according to a surgeon's individual preferences not only increases the number and complexity of operations but is also more distressing for children.

The clinical characteristics of pedal macrodactyly vary extensively, from mild enlargement of the affected toe, with only soft tissue involvement, to gross overgrowth, involving all elements of the foot. There is currently no consensus as to the best procedure for any given type of macrodactyly of the foot, and there is no widely used treatment algorithm. An objective and useful surgical algorithm for surgical treatment of pedal macrodactyly, with indications of corresponding procedures, is urgently needed for this rare condition.

The purpose of this study was (1) to introduce a surgical algorithm for treatment of macrodactyly of the foot based on the involved elements of the foot (soft tissue, phalanx, or metatarsal, or a combination of these), and (2) to demonstrate a multi-technique procedure to be applied under the guidance of the

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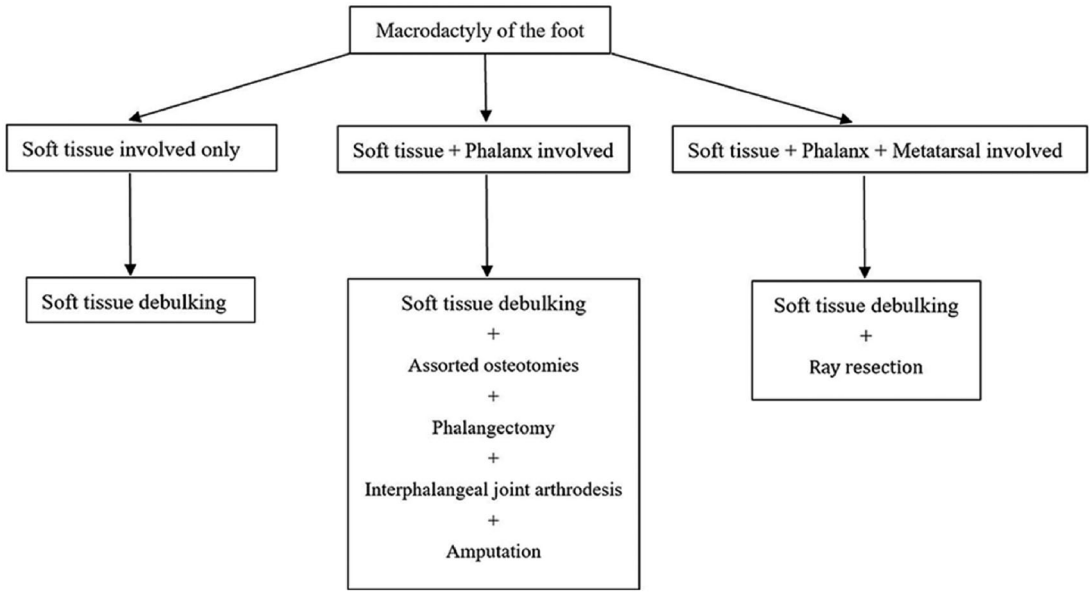

Figure 1. Algorithm of treatment procedures according to the involved elements of the foot: soft tissue, phalanx, metatarsal, or a combination of these.

proposed algorithm. This is a single-stage surgical procedure for achieving the goal of treatment of pedal macrodactyly and dramatically reducing the number of operative interventions.

Methods

The study protocol was approved by the ethics committee of our hospital, and written confirmation of informed consent was obtained from the patients' parents. The study protocol conformed to the ethical guidelines of the 1975 Declaration of Helsinki.

A total of 27 feet of 26 consecutive patients with macrodactyly of the foot, who experienced difficulties in wearing shoes, aesthetic problems, or pain, underwent surgical treatment at our hospital between January 2017 and August 2018.

Data derived from clinical examination and anteroposterior radiographs of both feet were collected. The rationale applied when generating the surgical algorithm was based on the severity of the affected foot and the treatment goals. The more serious the affected foot was, and the more elements of the foot affected, the more complicated the surgical options were. Accordingly, the affected toes were classified into three groups, based on the elements of the feet involved; that is, the soft tissue, the phalanx, or the metatarsal, or a combination of these. This simple and objective classification system could reduce the number of decision-making mistakes derived from varied clinical manifestations and individual surgeon preferences. If only soft tissue was involved (group A), soft tissue debulking was performed; if soft tissue and the phalanx were involved (group B), soft tissue debulking was performed in addition to assorted osteotomies, phalangectomy, interphalangeal joint arthrodesis, or amputation, or a combination of these; if soft tissue, the phalanx, and the metatarsal were involved (group C), soft tissue debulking and ray resection were performed. The surgical algorithm is shown in Figure 1.

If the affected toe was at least twice the length of the toe on the contralateral side, the distal phalanx was amputated; if it was less than twice this length, phalangeal shortening by wedge osteotomy or interphalangeal joint arthrodesis was performed. If both the width and height of the affected phalanges were increased, longitudinal osteotomy of the phalanx was carried out. Ray resection was performed in the following cases: (1) involvement of the metatarsal; (2) gross overgrowth of the forefoot; (3) accompanying syndactyly; (4) the order of the affected toes (more third and fourth rays were resected because the third and fourth toes were shorter than the first and second toes, anatomically); (5) the desire of the patients and parents (older patients had a stronger desire for ray resection). If the first metatarsal and phalanx were involved, osteotomy was preferred, rather than resection of the first ray, as the first toe contributes substantially to weight-bearing and normal gait. Soft tissue debulking was an integral part of the treatment and was performed in all cases.

Outcome evaluation

Follow-up assessments were conducted by two surgeons, independently of each other. Anteroposterior radiographs of both feet were routinely obtained. The intermetatarsal width ratio, calculated as the intermetatarsal width of the macrodactylic foot divided by that of the normal foot, was used to evaluate the change of width of the foot [18]. The phalanx spread angle measured as the angle between the medial border of the first phalanx and the lateral border of the fifth phalanx, was used to evaluate the changes in the width of the toes and the comfort of shoe wear. In ray resection cases, the metatarsal spread angle, which is measured as the angle between the medial border of the first metatarsal and the lateral border of the fifth metatarsal (Supplementary Figure 1) [1], was also determined, to evaluate the effect of treatment. We administered the Oxford Ankle Foot Questionnaire for Children (OxAFQ-C) [19] and the Questionnaire for Foot Macrodactyly to evaluate the clinical results (Supplementary Table 1) [18]. Shoe wear and cosmetic appearance are the two most important concerns to parents, and each foot was graded according to these two concerns. The foot was graded as 'good' if the shoe on the affected foot was equal to that on the opposite shoe and all toes matched without residual soft tissue bulkiness; it was graded as 'fair' if it was necessary to wear a shoe that was at least one or one-half size larger than the opposite shore, or there was residual soft tissue bulkiness without additional treatment and all toes presented overall coordination simultaneously; and it was graded as 'poor' if it was necessary to wear a shoe that was at

Table 1. Clinical data of 26 patients (Patient No.12 case is bilateral involvement).

Patient (*n*)	Age (months)[a]	Sex	Side	Involved structures				
				Toes	Phalanx	Forefoot	Metatarsal	Syndactyly
1	9	F	R	1,2,3	2,3	Yes	2,3	
2	22	M	L	1,2,3	2,3	Yes	2	
3	60	F	L	1,2	1,2	Yes		
4	66	F	R	1,2,3	2	Yes	2	
5	10	M	R	1,2,3,4	2,3	Yes	2,3	
6	15	F	L	2,3,4	2,3	Yes	3	
7	36	F	L	1,2	2	Yes		
8	36	M	L	1	1	Yes		
9	28	M	L	5		Yes		
10	48	F	R	2	2	Yes		
11	7	M	R	1,2,3,4	2,3,4	Yes	2,3,4	2,3
12	13	M	L	3,4	3	Yes		
			R	3,4,5	4	Yes		
13	108	F	R	2,3,4	3	Yes		
14	48	F	L	1,2,3,4	1,2,3	Yes	2,3	
15	11	M	R	1,2,3	2	Yes	2	
16	48	F	L	2	2	Yes		
17	14	M	L	2,3,4	2,3	Yes		2,3
18	82	M	L	2,3	2,3	Yes		
19	41	F	L	3,4	3	Yes		
20	17	M	L	2	2	Yes		
21	21	M	L	2	2	Yes		
22	13	F	L	2,3,4	2,3	Yes		2,3,4
23	60	M	R	1,2	1	Yes		
24	9	M	L	2,3,4	2,3,4	Yes		2,3,4
25	13	M	L	1,2	1	Yes		
26	34	F	L	1,2	1	Yes		

[a]Age at the time of operation.

Table 2. Surgical techniques used in the management of macrodactyly of the affected toes.

Patient (*n*)	The affected toes				
	1st	2nd	3rd	4th	5th
1	A	F	E		
2	A	F	C		
3	B[a]	F			
4	A	F	A		
5	E	F	F	A	
6		E	F	E	
7	A	B[b]			
8	B[a]				
9					A
10		B[b]			
11	A	F	F	E	
12 L			F	A	
12 R			A	B[b]	A
13		A	E	A	
14	A	F	E	A	
15	A	F	A		
16		C			
17		E	F	A	
18		C	D		
19			C	A	
20		B[b]			
21		B[b]			
22		A	F	A	
23	B[a]	A			
24		E	F	E	
25	A	F			
26	B[ab]	A			

A: Soft tissue debulking only. B: Soft tissue debulking + Assorted osteotomies. C: Soft tissue debulking + Phalangectomy. D: Soft tissue debulking + Interphalangeal joint arthrodesis. E: Soft tissue debulking + Amputation. F: Soft tissue debulking + Ray resection.
[a]Longitudinal osteotomy; [b]wedge osteotomy.

least two sizes larger than the opposite shoe, or a customized shoe was required, there was residual soft tissue bulkiness needing additional treatment, or the toes did not match.

Statistical analysis

The intermetatarsal width ratio and OxAFQ-C scores were compared pre- and post-operatively using the Wilcoxon signed-rank test, while the metatarsal spread angle and phalanx spread angle were compared pre- and post-operatively using the paired *t*-test. Statistical significance was considered for $p < 0.05$. The statistical analyses were performed using SPSS 20.0 (SPSS Inc., Chicago, IL, USA).

Results

A total of 27 feet of 26 patients with macrodactyly of the foot were evaluated, with a mean patient age of 33 months at the time of surgery (range: 7–108 months). Macrodactyly involved the right foot in eight patients, the left foot in seventeen patients, and both feet in one patient. The first, second, third, fourth, and fifth toes were involved in 13, 22, 16, 11 and 2 feet, respectively. All feet exhibited various degrees of hypertrophy of the soft tissues in the forefoot. One foot had only soft tissue involvement, eighteen feet had soft tissue and phalanx involvement, and eight feet had soft tissue, phalanx, and metatarsal involvement. Syndactyly presented in four feet, with two feet showing syndactyly of the second and third toes and two feet showing syndactyly of the second to fourth toes. Table 1 gives the clinical data of the patients.

The surgical techniques used in the treatment of macrodactyly of the affected toes were as follows. In group A, eight first toes, four second toes, three third toes, seven fourth toes, and two fifth toes were treated with soft tissue debulking only. In group B, four first toes, four second toes, and one fourth toe were treated with soft tissue debulking combined with assorted osteotomies; two second toes and two third toes were treated with soft tissue debulking combined with phalangectomy; one third toe was treated with soft tissue debulking combined with interphalangeal joint arthrodesis; and one first toe, three second toes, three third toes, and three fourth toes underwent soft tissue debulking combined with distal phalanx amputation. In group C, nine second toes and seven third toes underwent soft tissue debulking combined with ray resection (Table 2).

No early complications, such as hematoma, infection, wound breakdown, or skin necrosis, were encountered. Wound healing of three feet was postponed for postoperative swelling, and secondary healing was achieved after the dressings were changed.

At follow-up, the sizes of the affected feet were found to have decreased significantly (mean: 33 months; range: 18–42 months), at a mean age of 65 months (range: 36–126 months). The mean preoperative intermetatarsal width ratio was 1.13; this decreased to 0.93 postoperatively ($p < 0.05$). In 14 feet undergoing ray resection, the mean metatarsal spread angle was 33.2° in the affected feet preoperatively ($p < 0.05$); this decreased to 15.8° postoperatively ($p < 0.05$). The mean phalanx spread angle was 31.3° in the affected feet preoperatively ($p < 0.05$); this decreased to 17.9° postoperatively ($p < 0.05$; Figure 2).

In terms of clinical results, all patients were observed to develop a normal gait, with no dysfunction on walking and running (Supplementary Video). Chronic pain and skin ulcers were non-existent, except for calluses in six feet. Local mild recurrence in soft tissue presented in two cases, but without necessitating additional treatment. A total of 15 patients could wear shoes sized to the unaffected foot, while 11 patients wore ready-made

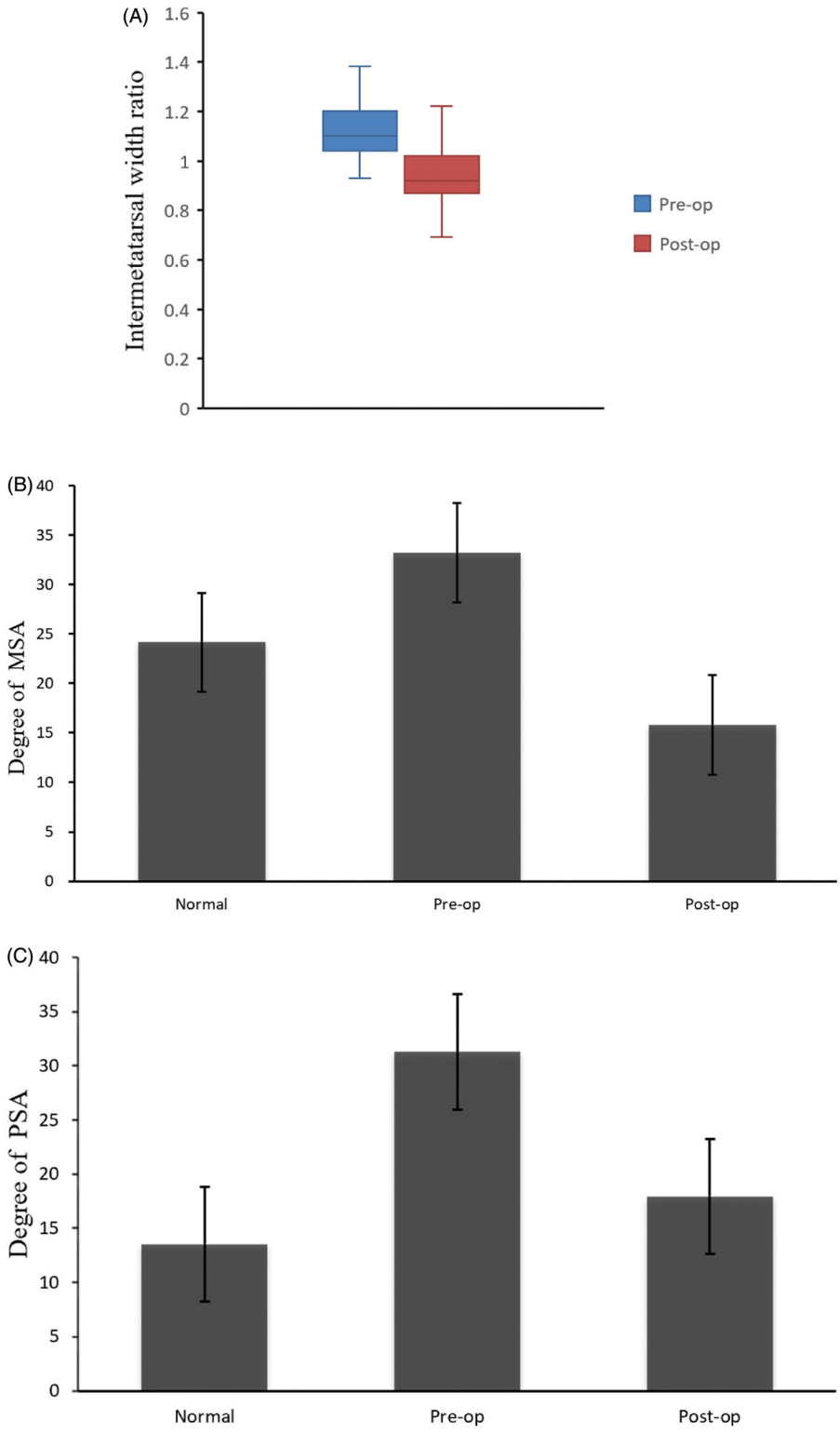

Figure 2. The affected foot size decreased significantly: (a) intermetatarsal width ratio; (b) metatarsal spread angle; and (c) phalanx spread angle are closer to those of the normal foot after the multi-technique procedure.

shoes that were one or one-half size larger than the contralateral side and used insoles. All parents were satisfied with the current operation and did not express any regrets (Figure 3). Of the 27 feet, 16 feet were with good results and 11 feet were with fair results. Although the 11 feet with fair results, which had residual soft tissue bulkiness, did not need additional treatment, six parents expressed a desire for further improvement in appearance. Figure 4 shows an example of the clinical results and follow-up observations.

We further compared feet in which only the lesser toes were affected (14 feet) and feet in which the great toe was affected (13 feet). No significant difference in the two groups between intermetatarsal width ratio and phalanx spread angle was found. However, of 11 patients who needed shoes one or a one-half size

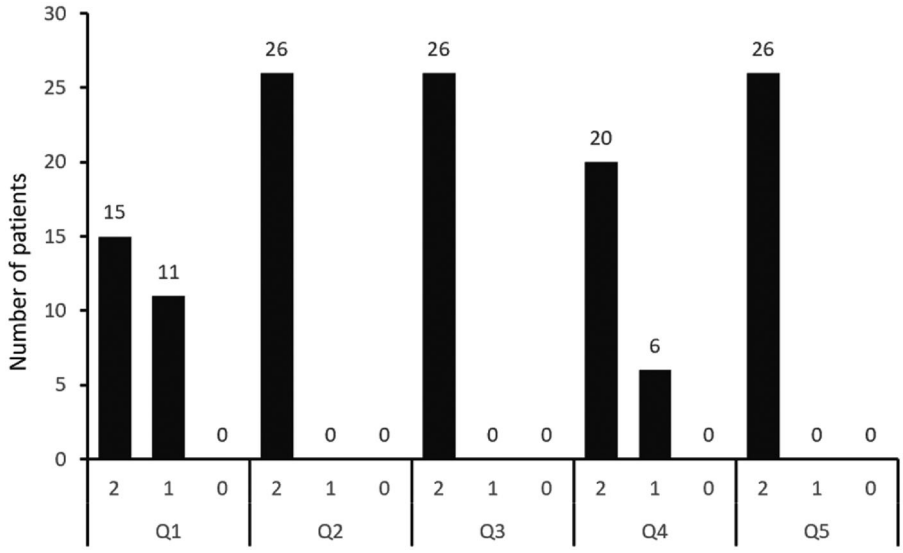

Figure 3. Postoperative scores obtained for the Questionnaire for Foot Macrodactyly (Q1, postoperative footwear; Q2, ambulatory ability; Q3, presence of pain; Q4, presence of skin lesions on the foot; Q5, postoperative satisfaction. Scores 0, 1, and 2 are explained in Supplementary Table 1).

larger in the affected foot than in the contralateral side, six cases involved the great toe; the surgery for these patients produced fair results.

The mean score for the Questionnaire for Foot Macrodactyly at the time of follow-up was 9.35 (Figure 3). The mean preoperative OxAFQ-C score was 42; this score increased to 47 ($p < 0.05$) after surgery. The score for the 'physical' category improved from 16 to 21; that for 'school and play' from 20 to 28, that for 'emotional' from 10 to 14, and that for 'footwear and clothing' from 4 to 12.

Discussion

Macrodactyly of the foot is an exceptionally uncommon deformity that has led to a paucity of descriptive literature on its treatment. Surgical treatment is mostly determined by the extent of the macrodactyly, the rate of overgrowth, and the surgeon's individual preferences. Few treatment algorithms for any given type of macrodactyly of the foot have been introduced. Chang et al. [1] introduced a simple guideline according to whether the great toe was involved; this guideline only applied to some cases and played a limited role in the clinical characterization of pedal macrodactyly, which varies extensively, from the involvement of only soft tissue to the involvement of all elements of the foot. Our algorithm was determined according to the involved elements of the foot, including soft tissue, the phalanx, or the metatarsal, or a combination of these elements. In practice, this method gives a classification of the severity of the deformity. In fact, the more elements involved, the more serious the deformity, and the more treatment procedures must be employed. Moreover, the location, extent, and degree of growth in the affected foot vary among individuals [12]. This means that it is more useful and objective to make a surgical decision on the basis of which elements of the foot are involved and that the treatment process is simplified.

Static or progressive types of macrodactyly have been considered in some treatment strategies, in order to decide between earlier and later surgical intervention [20], but this point was not considered in our algorithm. Any degree of toe enlargement would have an effect on normal function, shoe wear, evenness of gait, or aesthetics, or a combination of these, and surgical intervention, in theory, inescapable. Early surgical intervention, which could minimize the development of deformities, related to the size, stiffness, and deviation of the phalanges, that could impede the development of a normal gait, is beneficial for patients with macrodactyly of the foot [21,22], Our algorithm, which is based on the involved elements of the foot, is not subject to the type of macrodactyly, and can be used to guide operation as early as possible.

Although many treatment options and surgical procedures have been reported [6], each has its respective indications and limitations. Soft tissue debulking on its own is insufficient in reducing the size of the foot [1]. First ray resection can not be performed, as the first toe contributes to weight-bearing and normal gait. Forefoot enlargement cannot be corrected by toe amputation [14]. Epiphysiodesis restricts longitudinal rather than appositional growth [13]. In the face of the diverse clinical manifestations of pedal macrodactyly, numerous treatment options, with various combinations of surgical procedures, are needed to achieve better results, as well as to reduce the number of operations. The single-stage procedure in our treatment algorithm is an individualized multi-technique procedure, incorporating numerous treatment option combinations. Using this method, foot size was effectively reduced, and good clinical outcomes were achieved.

Of the varied treatment options, we particularly focused on debulking and ray resection. Although it is recommended to stage an operation when two sides of the toe are involved, we performed debulking of both sides of the affected toe simultaneously, as this could reduce the number of surgical interventions; however, digital arteries needed to be protected carefully. Moreover, soft tissue debulking of the dorsal and plantar aspects of the forefoot, which is usually ignored [14], is very important, in order to reduce the height of the affected forefoot and improve shoe comfort. Kim et al. [18] introduced ray resection in patients with metatarsal involvement, a motionless toe, or the involvement of more than one toes as indicators for surgery. More items should be added to this list to account for the diverse clinical characteristics of pedal macrodactyly and enable cautious consideration of ray resection. In addition to the location of the involved metatarsal, the severity of the deformity, the location of the

Figure 4. A 13-month-old girl with macrodactyly of the left foot, before and after surgery, and at 6 months and 3 years after surgery. (a, b) The second to fourth toes and the forefoot are enlarged, and there is enlargement of the distal and middle phalanges. The deformity is accompanied with syndactyly of the second to fourth toes. (c) The third ray was resected and soft tissue debulking was performed on the affected toes and forefoot. (d) Wearing shoes the same size as each other, 6 months after the operation. (e, f) There is no scar hypertrophy. (g, h) Calluses and local mild recurrence in soft tissue on the foot were observed at follow-up 3 years postoperatively.

affected elements, and any additional pathology, as well as expectations of the patients or parents, must be taken into account. In the foot where the first metatarsal is involved, osteotomy, rather than resection of the first metatarsal, was performed, as the first toe substantially contributes to weight-bearing and normal gait.

Our single-stage surgical procedure presented better results under the guidance of our new algorithm (Supplementary Table 2). Moreover, none of the patients experienced a recurrence of the deformity that warranted additional treatment during follow-up (mean: 33 months, range: 18–42 months), perhaps for the following reasons.

1. Macrodactyly appears to be a condition driven by the nerves themselves; nerve excision could prevent recurrence [3]. We excised the nerve branches and retained the digital nerve trunk, avoiding sensory disturbances or toe growth.
2. The affected adipose tissue might be an initiating factor in the pathogenesis of macrodactyly [23]. We excised as much affected adipose tissue as possible when debulking.
3. Ray resection dramatically reduces the probability of recurrence in metatarsal-involved cases [22]. However, the patients were all growing children and the possibility of recurrence remained; continued follow-up until skeletal maturity is still recommended.
4. The treatment algorithm could reduce decision-making mistakes derived from varied clinical manifestations and surgeon's individual preferences, and improved the administration of our single-stage multi-technique procedure.

Although, to our knowledge, we present the largest case series of macrodactyly of the foot and, to date, the most diverse range of clinical phenotypes, we also acknowledge several limitations of our study.

1. Standard anteroposterior foot radiographs could not be obtained in a few patients because the patients were too young to be able to cooperate.
2. The multi-technique procedure that we used is complicated and radical and had to be performed by experienced pediatric orthopedic surgeons.
3. The validity of the OxAFQ-C in patients younger than 5 years has not yet been demonstrated; neither has that of the Questionnaire for Foot Macrodactyly.
4. Owing to the rareness of the deformity, the results and statistical significance are limited; more cases will be included in further studies.
5. Owing to the progressive nature of the deformity, our follow-up period was limited; continued follow-up until skeletal maturity or after adolescence is still recommended.

The purpose of treatment is to obtain a functional and cosmetically acceptable foot that permits the wearing of regular shoes through a minimum number of operations; our new treatment algorithm, which is based on the involved elements of the foot, and comprehensive multi-technique procedure could fully satisfy this goal.

Disclosure statement

No potential conflict of interest was reported by the author(s).

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