Macrodactyly is a rare congenital deformity characterized by hypertrophy of the bones and surrounding soft tissues in one or more digits. The terms “macrodactyly”, “megalodactyly”, and “digital” or “localized gigantism” are often used synonymously [1,2]. The condition seems to affect slightly more boys than girls, and it may occur only on one foot or on both feet, and it may be symmetrical or asymmetrical. Macrodactyly may occur as an isolated defect in either the hand or the foot, or it may present as an element of conditions, such as Proteus syndrome, Klippel-Trenaunay syndrome, Bannayan-Riley-Ruvalcaba syndrome, Maffucci, Ollier’s disease and Miley-Rodriguez’s disease.

The enlargement includes skin, subcuteaneous tissue, nerve, joint, and bone. The bony involvement is usually limited to phalanges [3]. There are two types of macrodactyly: static and progressive. The progressive variant is more frequently seen in the foot than in the hand. Radiologically, X-ray, computed tomography (CT) and magnetic resonance imaging (MRI) are used to determine the size of lesions, involvement...
of soft tissue and bone, which are important for surgical
treatment planning. In macrodactyly, reconstruction is
aimed at decreasing the size of the foot in normal size
and shape as much as possible. Many surgical options are
available for the treatment of macrodactyly of the foot:
amputation, ray amputation, epiphyseal ablation, trans-
verse and longitudinal osteotomies, nerve stripping, and
extensive defatting. None of the available methods, how-
ever, give ideal functional and cosmetic results [4].

Most patients are diagnosed and treated from child-
hood; however, macrodactyly might become more se-
vere if diagnosis and treatment are delayed for a long
time. In this case, we report a rare case of long-term
(23 years) macrodactyly of a toe resulting from delayed
treatment.

**Case Report**

A 23-year-old man was admitted to our clinic with
a giant deformed great toe of the right foot which had
been present since birth. The walking and shoe wear
were difficult for him and even caused the size of the
shoes of both feet to differ from each other to such a
large extent that the right one is six sizes larger than

![Figure 1](image1.png)  ![Figure 2](image2.png)  ![Figure 3](image3.png)

**Figure 1.** On physical examination, the right great toe and forefoot
showed extreme enlargement and incredible abnormality.

**Figure 2.** A plain radiograph of the right foot showed remarkable
overgrowth of the first phalanx bone with significant deformity.

**Figure 3.** Computed tomography and magnetic resonance imaging
scans of the right foot showed that the adipose layer was apparently
thickened.
Adult foot macrodactyly

Figure 4. At the latest follow-up 8 months postoperatively the patient was very satisfied with his foot.

Figure 5. The appearance of the plain radiography of the right foot at 8 months follow-up.

the left one. There were no positive family history and other congenital malformations. Moreover, we couldn’t find any sign of a systemic disease, e.g. neurofibromatosis. On physical examination, the right great toe and forefoot showed extreme enlargement and incredible abnormality (Figure 1a,1b). Also, along with these 2nd and 5th phalanges, deformity also existed. The second toe has no phalanges, and macrodactyly is not combined with syndactyly. The fifth toe phalanges are malrotated. The deformed toe was 7 cm in sagittal diameter and 5 cm in horizontal diameter. The plantar skin of the sole became remarkably thick. A plain radiograph of the right foot showed remarkable overgrowth of the first phalanx bone with significant deformity (Figure 2). Computed tomography and magnetic resonance imaging scans of the right foot showed that the adipose layer was apparently thickened (Figure 3a,3b).

The major aim in the treatment of foot macrodactyly is the reconstruction of a pain-free, functional foot with a good cosmetic appearance. A debulking procedure and toe reconstruction with distal phalanx amputation were planned after consideration of the treatment options. At surgery, a zigzag digital incision was made on the palmar and dorsal aspect of the toe. The incisions were aligned to match zigs and zags. An en bloc dissection included skin, fatty tissue, neurovascular bundles and proximal phalanx. Flexor and extensor tendons were slightly detached from the bone and were retracted to allow bone resection with an oscillating saw. Disarticulation was achieved through complete removal of the distal phalanx beginning from the interphalangeal joint. The tip of the finger and part of the nail were removed. The soft tissues of the dorsal and plantar aspects of the toe were also debulked. Primary closure was performed. A short leg cast was applied with medial and lateral compression. At the latest follow-up 8 months postoperatively (Figure 4a,4b,5), the patient was very satisfied with his foot and shoe wear became easier than preoperation. The patient’s only complaint remains a mild pain triggered with long-distance walks.

Discussion

Macrodactyly of the foot is a rare congenital anomaly characterized by an enlargement of the soft tissue and osseous elements. The pathogenesis of macrodactyly is not known and defined. Heredity does not ap-
pear to play a role, and our patient did not have a family history [5]. Macrodactyly is not necessarily associated with any other type of deformity, although it can be found in people with some types of syndromes, including neurofibromatosis, hemangiomatosis, arteriovenous malformations, congenital lymphedema, and Klippel-Trenaunay-Weber and Proteus syndromes [6-8].

However, not every enlargement of the foot, or any element of the foot, is macrodactyly. Enlargement of a toe can result from the presence of a hemangioma, or some other neoplasm, in which only the overlying skin and adjacent soft tissues are involved, with no radiographic evidence of an increased size of the osseous elements [9,10]. Other diseases in which “false macrodactyly” can occur are Ollier’s disease, Maffucci’s syndrome, vascular malformation, neurofibromatosis, and Milroy’s disease [1,10].

Syed et al. reported that in macrodactyly of the foot, excessive proliferation and accumulation of adipose tissue was the basic lesion. But in the hand, macrodactyly hypertrophy and tortuosity of the digital nerve were the striking features which were absent in cases affecting the foot [4,11]. Barsky AJ reported that macrodactyly is more commonly observed in males and in the foot and he defined two types of macrodactyly. Static macrodactyly, the most common, is a congenital growth which progresses in proportion with normal growth. Patients with static macrodactyly have larger toes (or toe) when they are born. It then continues to grow proportionally to the other fingers. Progressive macrodactyly, in which the digit involved has a higher rate of growth, is less common [9]. In macrodactyly, the blood vessels are frequently either of normal caliber or only slightly enlarged; the digit as a whole is thus relatively poorly vascularized. Healing potential is thereby reduced and the incidence of wound breakdown and infection is high. These complications are more likely to occur when extensive dissection and defatting have been performed [10,12].

The major goal in surgical reduction of foot macrodactyly is to obtain a pain-free and satisfactory aesthetic foot [4,13,14]. When determining the treatment, it is important to consider the severity of the deformity. Therefore, radiologically, X-ray, computed tomography (CT) and magnetic resonance imaging (MRI) are important for surgical treatment planning. Treatment varies according to which toe is affected. If surgery needs to be done, the surgeon could destroy the growth part of the bone, along with removing as much of the excess tissue as possible.

The surgical options include debulking of soft tissues, amputation of the distal phalanx, middle phalanx, or the whole ray, and epiphysiodesis by stapling or destroying the epiphysis to prevent additional longitudinal growth without controlling the circumferential growth. Debulking procedures can be performed with epiphysiodesis to reduce the size of the toe, but it must sometimes be repeated because of progressive recurrence of the macrodactyly [10,15-17].

In macrodactyly of the toes, the Tsuge method, which involves excision of the distal phalanx with soft tissue reconstruction using a dorsal flap, along with nail preservation, is generally recommended [18]. In our case, a 23-year-old patient accepted amputation of the distal phalanx at the interphalangeal joint combined with the removal of the fibro-fatty tissue. He did not desire the preservation of the nail. With the flap proposed for coverage of the fingertip, sensation is preserved. Active motion was preserved and metatarsophalangeal joint stability was maintained by the collateral ligament [4]. Although the patient seemed satisfied with the result of treatment, the final appearance was not as good as expected.

Because of the rarity of the condition, few reports pertaining to the results of the surgical treatment of this condition have been published. Bulut et al. reported 3 cases of ray amputation with good cosmetic and functional results. They suggest that ray amputation is an effective single-stage treatment method for cases of advanced or recurrent macrodactyly of the lesser toes that provides acceptable cosmetic and functional results [19]. Primary advantages are the acceptable cosmetic results and the reduction in the size of the involved digit, which can only be achieved by this procedure. Ray amputation is not suggested in macrodactyly of the great toe because of the important role of the great toe in normal stepping and walking, as well as the unacceptable cosmetic result [6]. Hendrix et al. applied amputation of the finger phalanx and painful lump excision of the 23-year-old male patient who was...
operated on before from the 2nd and 3rd fingers, and the resulting defect closed with a rectus abdominis flap. Their surgical reconstruction of the macrodactyly included phalangectomy of the base of the second digit of the left foot, ray resection of the third digit at the metaatarsophalangeal joint level, and resection of the dorsal and plantar mass. The defect was covered with a rectus abdominis free-tissue flap and a split-thickness skin graft [20]. Hop et al. searched for all cases of macrodactyly treated with ray resection and combined them with their three cases to provide an overview of the results of this treatment. Their report was similar to the other reports: this technique can lead to successful results in macrodactyly of the forefoot [10]. Sabapathy et al., in two cases, shortened the toes by amputation and, in an attempt to improve the appearance, transplanted the nails as free grafts onto the amputation stumps. But in the first case, the new nail was deformed due to the absence of nail folds. Therefore, in the second case, the lateral nail folds and eponychial folds were included in the composite grafts, and the result was better [12]. Dautel et al., in a case report, shortened the great toe by removing the distal two-thirds of the distal phalanx after isolating the nail as an island flap in a subperiosteal plane. Being isolated as an island flap, the nail complex was mobilized to its new position on top of the foreshortened toe. They advocate the use of a vascularized pedicled transfer because of the safety of nail grafting compared with conventional non-vascularized nail grafts [8]. Katz JB reported a progressive macrodactyly. The patient was a 48-year-old man who presented with a gross deformity of his right foot and ankle. The toes of his foot, although always large and deformed, had started to grow again when he was approximately 43. The surgical decision was amputation of the fifth toe and partial ray resection of the fifth metatarsal. They had a wound-healing complication and the patient discharged 19 days after surgery [21].

No consensus has been reached regarding the treatment of foot macrodactyly. Many surgical options are available for the treatment of macrodactyly of the foot. Thus, the most appropriate technique should be chosen for the patient individually. For severe macrodactyly of the great toe, amputation of the first phalanx could be an alternative treatment.

Conflict of interest statement
The authors have no conflicts of interest to declare.

References
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