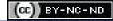
# Middle Phalangectomy for the Correction of Macrodactyly of the 2<sup>nd</sup> Digit in an Adult Patient: A Case Report

Surgery Section

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### **ABSTRACT**

Macrodactyly, a rare congenital anomaly characterised by localised gigantism of one or more digits, presents significant challenges in therapeutic management due to the absence of established guidelines. The present case report explored the surgical management of an 18-year-old female patient with macrodactyly of the second digit, addressing discomfort, footwear issues and psychological distress. The study adapts techniques from prior paediatric cases to suit adult care needs, particularly focusing on middle phalangectomy as a feasible treatment option for isolated macrodactyly. Clinical examination and radiographic assessments confirmed primary macrodactyly affecting the patient's left second toe. The surgical intervention involved middle phalangectomy, debulking of fibrofatty tissue, joint capsule and tendon repair, along with a Winograd wedge resection procedure. Postoperative care included pain management and wound dressing. Despite encountering delayed wound healing and mild necrotic changes, the patient reported manageable pain levels and expressed satisfaction with the cosmetic outcome and improved footwear options at the six-month follow-up. The present case report contributes valuable insights into the management of macrodactyly, highlighting the efficacy of middle phalangectomy in adult patients. Preoperative assessment for peripheral arterial disease is emphasised to mitigate neurovascular complications. Proactive patient counseling regarding potential postoperative complications is crucial for informed decision-making. Overall, the findings underscore the importance of tailored surgical approaches in addressing macrodactyly and improving patients' quality of life.

# Keywords: Congenital, Giantism, Phalanges, Surgery

#### **CASE REPORT**

A clinically healthy 18-year-old female presented for the evaluation and management of her left 2<sup>nd</sup> digit congenital deformity. The onset of this deformity was noticed by the patient's parents immediately after birth, as they observed hypertrophy of her left second toe compared to the adjacent digits. The patient described experiencing discomfort during physically demanding activities and facing challenges in finding appropriate footwear. Additionally, the patient expressed a desire to hide her feet in public due to emotional distress caused by the noticeable appearance of her feet.

The patient's medical history revealed that she takes medication for anxiety management and an oral contraceptive. She is a non smoker and consumes minimal alcohol. The maternal history indicated an uncomplicated pregnancy, marked by normal growth patterns and the achievement of expected physical and cognitive developmental milestones, with no known associations with syndromic conditions. Furthermore, there is no reported family history of similar congenital deformities among known family members.

Clinical examination revealed the absence of any other abnormalities in the feet. The left second digit exhibited a normal range of motion across both the interphalangeal and metatarsophalangeal joints. Notably, a semiflexible lateral deviation of the 2<sup>nd</sup> toe was observed, as illustrated in [Table/Fig-1]. Both clinical and radiological assessments most closely support a diagnosis of primary macrodactyly affecting the 2<sup>nd</sup> digit of the left foot. Importantly, there are no features indicative of alternative diagnosis, such as tuberous sclerosis complex, neurolipomatous macrodactyly, proteus syndrome, or giantism.

Radiographic examinations suggest enlargement of both the middle and distal phalanx of the left second digit. Clinical observations and radiographic assessment indicate the presence of fibrofatty tissue surrounding the bones of the 2<sup>nd</sup> digit. Further examination reveals a wider 2<sup>nd</sup> toenail plate compared to the contralateral 2<sup>nd</sup> toe. A medial deviation of the distal phalanx is noted, attributed to the deformity in the middle phalanx, which measures 20 degrees [Table/ Fig-2a,b]. Importantly, no cutaneous lesions indicative of Klippel-Trenaunay-Weber syndrome are observed.



[Table/Fig-1]: Preoperative weight-bearing photograph of left 2nd digit macrodac-



ateral X-ray of left 2nd toe macrodactyly

Additionally, comprehensive neurovascular assessments involving laser Doppler, Ankle-brachial Index (ABI), monofilament testing and vibration perception assessments all yield results within normal parameters. Due to the absence of other discernible signs and symptoms, further diagnostic tests were not deemed necessary.

After multiple discussions emphasising the potential risks, benefits, complications and side-effects of surgical intervention, the patient chose to proceed with surgical treatment under general anaesthesia.

# **Operative Technique**

The procedure was performed under intravenous general anaesthesia. A preoperative injection of 6 mL of 0.75% naropin plain was administered via a 2<sup>nd</sup> ray block technique for local anaesthesia. The site was prepared using betadine solution and the extremities were appropriately draped to maintain sterility. The left limb was exsanguinated, and a calf tourniquet was inflated to a pressure of 270 mmHg. A linear incision was made just lateral to the centerline of the left 2<sup>nd</sup> digit, starting proximal to the Proximal Interphalangeal Joint (PIPJ) and extending distal to the Distal Interphalangeal Joint (DIPJ) area. The superficial fascia was meticulously separated, with a primary focus on preserving the integrity of the neurovascular structures.

The intermediate phalanx was excised to allow for decompression and alignment of the digit. After the phalanx excision, a portion of the fibrofatty tissue was removed from the medial and lateral sides of the digit. An ostectomy was then performed on the prominent medial and lateral condyles of the distal and proximal phalanx using a rongeur. An elliptical excision was carried out on the plantar aspect of the 2<sup>nd</sup> toe, allowing for further debulking of the fibrofatty tissue. Alignment, position and circumference were assessed and deemed adequate. Image intensification was utilised to ensure appropriate correction of the medial and lateral condyles. The area was flushed with saline.

The plantar and dorsal aspects of the joint capsule and flexor tendon were repaired in a corrected position using a bone tunnel in the distal phalanx with 3-0 Ethibond and 2-0 Vicryl. Joint capsules were repaired using 2-0 Vicryl with the toe held in a corrected position. The extensor tendon was retensioned and repaired in the corrected position using 3-0 Ethibond and 2-0 Vicryl. The skin was reapproximated with 3-0 Nylon suture.

A Winograd wedge resection procedure was then performed to reduce the width of the  $2^{\rm nd}$  toenail. Excess distal fibrofatty tissue was excised through the Winograd incisions. The area was flushed with saline and the skin was reapproximated and closed with 2-0 nylon suture.

A regional block was performed with ropivacaine 0.75% and 1 mg of dexamethasone for lasting postoperative analgesia. After the procedure, the patient was partially weight-bearing in a modified Robert-Jones compression dressing until the sutures were removed at three weeks. Pain was managed with ice packs, paracetamol and ibuprofen. Breakthrough pain was addressed with oxycodone; however, this was not necessary.

The patient received the standard postoperative care protocol as prescribed by the treating surgeon. This protocol included a structured postoperative follow-up plan, which involved 24-hour postoperative phone support and scheduled, in-person postoperative visits at weeks 1, 2, 3, 6, 12 and 24. Additional visits were arranged as deemed clinically necessary.

During the initial weeks following the surgery, the patient reported manageable levels of pain. It is important to note that experiencing pain in the early postoperative period is an expected aspect of the recovery process and is not considered a complication. At the 3<sup>rd</sup>-

week postoperative appointment, clinical observation revealed the presence of mild necrotic changes affecting the 2<sup>nd</sup> digit [Table/Fig-3a,b]. Neurovascular compromise emerged as a primary concern during the course of this procedure. Vigilant and regular assessments were performed, including measurements of capillary refill and monitoring digital and foot temperatures.



**[Table/Fig-3a,b]:** Three weeks postoperative photograph of the dorsal aspect and apex of the  $2^{\rm nd}$  toe. Some mild necrosis can be noted at the distal lateral aspect of the toe.

Delayed wound healing was a notable complication in this case. The patient sought clinical attention eight weeks following the procedure due to the persistence of delayed wound healing and mild ongoing exudation from the lateral wedge resection site. At this point, no additional signs of infection were apparent, and the condition was managed through ongoing wound dressings and the application of topical antiseptic creams administered by the patient at home.

During the six-month postoperative review, the patient showed remarkable progress with no reports of pain. She expressed satisfaction with the cosmetic results of the procedure and was pleased with her expanded choice of footwear. It is worth noting that a mild lateral deviation of the second digit persisted at this point, a characteristic that was deemed acceptable by both the patient and the surgeon.

A noteworthy observation was the continued delayed wound healing on the dorsolateral aspect of the second digit [Table/Fig-4]. It was hypothesised that this localised issue may be attributed to suture extrusion. Weight-bearing X-rays were also reviewed six months postoperatively, displaying the acceptable position and alignment of the 2<sup>nd</sup> digit [Table/Fig-5].





[Table/Fig-4]: Partial weight-bearing image of left foot six months postoperatively. [Table/Fig-5]: Weight-bearing AP X-ray of left foot six months postoperatively. (Images from left to right)

## **DISCUSSION**

Macrodactyly, a rare congenital condition, has been documented in medical literature since as far back as 1840 [1]. Macrodactyly is a congenital anomaly characterised by localised gigantism of one or more digits, most frequently observed in the lower limbs [2]. Primary macrodactyly is an enlargement of the fingers or toes without limb enlargement, neurological or vascular abnormalities, or any features suggestive of a syndrome [3]. Secondary macrodactyly is associated with an underlying disease, such as neurofibromatosis and Klippel-Trenaunay-Weber syndrome. The incidence of macrodactyly in toes is reported to occur in 1/18,000 births [3]. True macrodactyly is a hamartomatous enlargement of all the elements of the affected digit [4]. Although the exact cause of true macrodactyly is unknown, it appears that it may have a genetic origin [5].

The objectives of surgical intervention for pedal macrodactyly encompass the reduction of deformity, enhancement of aesthetic outcomes, improvement in overall quality of life, alleviation of psychological distress and facilitation of comfortable footwear usage. The spectrum of surgical treatments available for managing foot macrodactyly is diverse and encompasses joint arthroplasty, arthrodesis, debulking fibrofatty tissue, various skin plasty techniques, phalangectomies and digital amputation [2-6].

Given the infrequency of this condition, there exists a paucity of case reports and case series delineating surgical intervention techniques and resultant outcomes. Josh F et al., described the correction of a similar presentation of Macrodactyly on a three-year-old child [6]. We have incorporated elements from this approach and implemented minor procedural adjustments to align with the present case adult patient presenting isolated macrodactyly of the second digit.

Despite multiple case studies and case series that have been published on this subject matter, there is no consensus or accepted protocol for surgical correction of this rare congenital deformity [5,7,8]. For the present case patient, the primary objective in treating 2<sup>nd</sup> toe macrodactyly is to provide stability, alleviate pain or discomfort, and enhance the aesthetic appearance of the toe [3,7]. In 1998, Kotwal PP and Farooque M, introduced a digit-shortening technique for macrodactyly, which involved phalangectomy. Their case series revealed that despite the combination of procedures employed, patients experienced some degree of stiffening in the operated digit. However, the authors underscored that achieving improved cosmesis outweighed concerns regarding increased stiffness due to joint removal [1]. The Kotwal PP and Farooque M middle planectomy technique entails excising the middle phalanx through a lateral inverted T incision. Subsequently, the joint capsule from the proximal interphalangeal joint is sutured to the joint capsule of the distal interphalangeal joint and stabilised using a percutaneous K-wire [1].

There have only been several case reports or series that have detailed the use of phalangectomy in various combinations of procedures [1,2,9,10]. For example, Josh F et al., described their experience using a middle phalangectomy and a double pedicled digital artery flap to treat a three-year-old girl with macrodactyly of the second digit. Their approach involved both dorsal and plantar incisions, with the use of a percutaneous K-wire. In the case of this young child, the outcome yielded an acceptable cosmetic appearance. However, the toe remained in a dorsiflexed position after the removal of the K-wire. Given the child's age, it was hypothesised that this would self-correct as she matured [6].

In the current case, an innovative approach was adopted by introducing a bone tunnel for joint capsule and tendon repair, which aimed to enhance stability, allowing for tape fixation

without the need for an intramedullary K-wire. Additionally, a bilateral Winograd wedge resection procedure was incorporated to reduce the size of the enlarged 2<sup>nd</sup> toenail and further debulk the distal toe, ultimately aiming to improve the overall cosmesis of the digit.

#### **Patient Perspective**

"At my six-month follow-up appointment after the surgery, I was genuinely pleased with the progress I had made. I couldn't believe the relief-no more pain! The surgery had truly been a game-changer for me. But what really brought a smile to my face was the cosmetic outcome. My second toe, which had caused me so much distress and discomfort, now looked remarkably better.

One of the most exciting changes was the newfound freedom in choosing my footwear. Before the surgery, finding comfortable shoes that accommodated my condition was a daily struggle. But now, I could wear a wider range of shoes that not only fit better but also looked stylish. It was a tremendous boost to my confidence.

Of course, it's not all smooth sailing. I did notice that a slight deviation in the position of my second digit was still there. However, both my surgeon and I agreed that it was a reasonable trade-off for the improved overall appearance and comfort. The fact that I no longer experienced pain or had to deal with limited shoe options made the residual deviation a minor concern in the grand scheme of things.

All in all, I'm incredibly satisfied with the results of the surgery and grateful for the positive changes it has brought to my life."

# **CONCLUSION(S)**

The utilisation of a middle phalangectomy in combination with the Winograd wedge resection technique appears to present a viable approach when addressing an isolated case of primary pedal macrodactyly involving the second toe. It is imperative to emphasise the importance of assessing for peripheral arterial disease as part of the preoperative evaluation, given the elevated potential for neurovascular complications. To mitigate this risk, it may be appropriate to consider a staged approach to the procedure. Furthermore, it is crucial to counsel patients before surgery, ensuring they are well-informed about potential postoperative complications, such as excess necrosis or infection, which could necessitate amputation. This proactive communication with patients helps manage expectations and fosters a more informed decision-making process.

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