

CASE REPORT**RECONSTRUCTION OF THUMB POLYDACTILY: A CASE REPORT**Budiman¹, Gladya Utami, T. Fadli Nazwan Sani

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ABSTRACT

Introduction: Polydactyly is the most common hand congenital abnormality found, next after syndactyly. The presence of these extra fingers is not merely only duplication but it is accompanied by abnormal hypoplastic structures, abnormal joints, tendon anomalies, and anomaly of the insertion of ligaments. There are many techniques proposed to reconstruct polydactyly, from simple excision of bones, ligaments, and tendons to complex reconstructions.

Patient and Method: A 21-year-old woman complaining of difficulty in grasping because the space between first and second fingers is too far away, and the extra metacarpal bone is protruding that causes snagging and stumbling on other objects. She had a history of surgical removal of polydactyl when she was 4-month-old. The patient has a limitation in hand activities.

Result: Six months after surgery, the patient was satisfied with the results. The fingers has better mobility, good appearance. No more prominent bone, and neuropathic problems, or other disorders were found.

Summary: Polydactyly is a common congenital hand occurrence. Understanding the anatomy to guide the reconstructive actions is important. It is not only about which digit would be preserved, but we have to concern about what our patient need, improving the quality, patient's hand function, and the aesthetic aspect.

Keywords: polydactyly; congenital hand; pediatric hand surgery

Latar Belakang: Polidaktili merupakan kelainan bawaan yang umum ditemui setelah sindaktili. Kehadiran jari tambahan ini bukan merupakan sekedar duplikasi, namun disertai dengan kelainan struktur anatomi yang hipoplastik, sendi abnormal, anomali tendon, dan insersi ligament. Banyak cara untuk rekonstruksi polidaktili, mulai dari yang sederhana eksisi tulang, ligamen, dan tendon hingga rekonstruksi yang rumit.

Pasien dan Metode: Seorang wanita 21 tahun mengeluh kesulitan dalam menggenggam karena jarak antara jari pertama dan kedua terlalu jauh, dan tulang metacarpal tambahan menonjol yang menyebabkan tersangkut dan tersandung pada benda lain. Pasien memiliki riwayat operasi pengangkatan polydactili ketika dia berusia 4 bulan. Pasien memiliki keterbatasan dalam melakukan aktifitas menggunakan tangan.

Hasil: Enam bulan pasca operasi, pasien merasa puas dengan hasil yang didapatkan. Mobilitas jarinya menjadi lebih baik, puas dengan penampilannya, tidak ada lagi tulang lebih yang menonjol, dan tidak ditemukan adanya kendala neuropati atau gangguan lainnya.

Ringkasan: Polidaktili merupakan kelainan kongenital tangan yang sering ditemui. Memahami anatomi penting untuk mengarahkan dalam rekonstruksi. Bukan hanya tentang digit mana yang akan dipertahankan, tetapi kita harus memperhatikan apa yang dibutuhkan pasien, meningkatkan kualitas fungsi tangan pasien, dan aspek estetika.

Kata Kunci: polidaktili, kelaianan bawaan pada tangan, bedah tangan anak

Conflicts of Interest Statement:

The author(s) listed in this manuscript declare the absence to any conflict of interest on the subject matter or materials discussed.

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INTRODUCTION

Polydactyly is characterized by the presence of an extra digit, digits, or part of a digit in hand or foot or both. Polydactyly is the common congenital anomaly. It can be found in isolation or as part of a syndrome. Polydactyly is classified into preaxial (radial or tibial), central (presence of duplicated second, third, or fourth digits), and postaxial (ulnar or fibular) types.^{1,2}

The thumb, responsible for 40% - 50% of hand function, must be able to oppose the other digits with a stable pinch. Besides trying to restore cosmesis, polydactyly surgical management also seeks to allow normal hand function. Thorough evaluation and diagnosis are among the most important aspects of overall patient management.³

Polydactyly is the fourth most common congenital anomaly after clubfoot, cleft lip/palate, and spina bifida, according to a 5-year audit of Pennsylvania Department of Health records. While Thumb duplication occurs in 0.08 to 1.4 per 1000 live births and is more common in American Indians and Asians than in other races.^{4,5} It occurs in a male-to-female ratio of 2.5 to 1 and is most often unilateral.⁴ Postaxial polydactyly is predominant in black infants; it is most often inherited in an autosomal dominant fashion, if isolated, or in an autosomal recessive pattern, if syndromic.⁶ A prospective San Diego study of 11,161 newborns found postaxial type B polydactyly in 1 per 531 live births (1 per 143 black infants, 1 per 1339 white infants); 76% of cases were bilateral, and 86% had a positive family history.⁶ While in Indonesia, scholarly literature on the number of polydactyls that is yet to be available.

Thumb duplications result from a failure of the radial-ulnar axis of the hand plate to form or differentiate. The zone of polarizing activity (ZPA) in the posterior part of the developing limb bud is the principal signaling center.

The ZPA expresses sonic hedgehog protein, which controls the formation of radial-ulnar features. Abnormal expression of Hox genes, bone morphogenic protein, and the Gli-3 gene all play a role in the evolution of thumb duplications. Genetic consultation is not indicated, as most cases are sporadic and unilateral. However, the presence of a triphalangeal thumb is known to exhibit an autosomal dominant inheritance pattern.⁷

Thumb duplications are classified under the more commonly used Oberg, Manske, and Tonkin (OMT) classification as malformations, because of their failure of formation/differentiation of the hand plate in the radial-ulnar axis. The deformity itself can be further subdivided based on the specific form of duplication. The Wassel system, developed in 1969, is most familiar among pediatric hand surgeons for its simplicity and is the most widely used system in the recent literature.⁸

This table below describes seven types of radial polydactyly according to the level of the skeleton at which duplication occurs. Type IV is the most common type of thumb polydactyly, followed by type II and type VII. Despite its widespread use, however, this system has several limitations when it comes to surgical planning.⁸ For example, the Wassel classification often fails to identify the origin of the extra digit, a key morphologic characteristic necessary for the process of surgical planning. Additionally, the system does not align classification types with unique operative techniques; hence, the management of Wassel types II, IV, and VI may be entirely similar.⁹ In clinical practice, this terminology is best used as a basic framework for the management of a thumb duplication.¹⁰

Table 1. Wassel Classification: Types I to VII based on the level of duplications¹¹

Type	Description	% of Polydactyly
I	Bifid distal phalanx	2%-6%
II	Duplicate distal phalanx	15%-17%
III	Bifid proximal phalanx	6%-9%
IV	Duplicate proximal phalanx	43%-46%
V	Bifid metacarpal	10%-12%
VI	Duplicate metacarpal	3%-4%
VII	Triphalangism	6%-20%

PATIENT AND METHOD

A 21-year-old woman complained of difficulty in grasping because the space between the 1st and 2nd digit is too far, and there is a bone in between which was protruding, and causes snagging and tripping on objects. According to the patient's, she was told by his mother that she had previously undergoes thumb removal operation at the age of 4-month. Preoperative x-ray demonstrated a Wassel type VI according to the Wassel's classification and triphalangeal thumb of the left hand with evidence of previous operation. The patient planned for surgical correction. We would opt to perform osteotomy of metacarpals than perform ray amputation to extra metacarpal in order to prevent gaping. Many triphalangeal thumbs can be quite functional which will not require of surgery.⁸

The patient asked to revise the space between the 1st and 2nd finger to be closer than before. So we performed the osteotomy of the 1st metacarpal, then we transported it to the top of the duplicated metacarpal, which osteotomy has also been done.

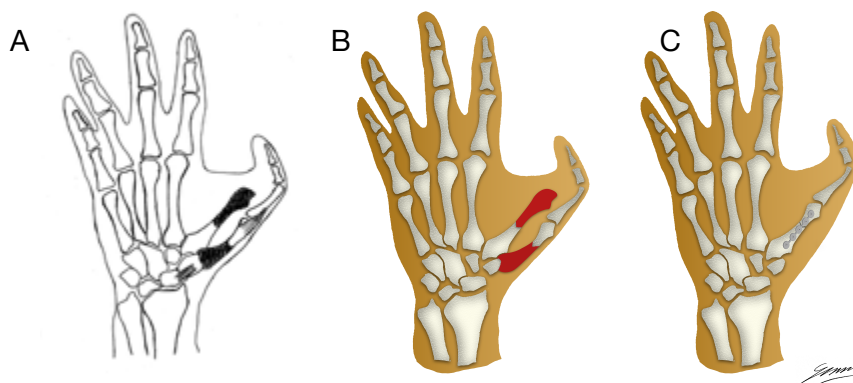
The surgical technique were simplified without tendon or flap transfer. The thumb must be active function before surgery. Amalgamated metacarpals were fixed with fixation plates. Healing was smooth, and the patient was discharged with no occurrence of surgery-related complications.

RESULT

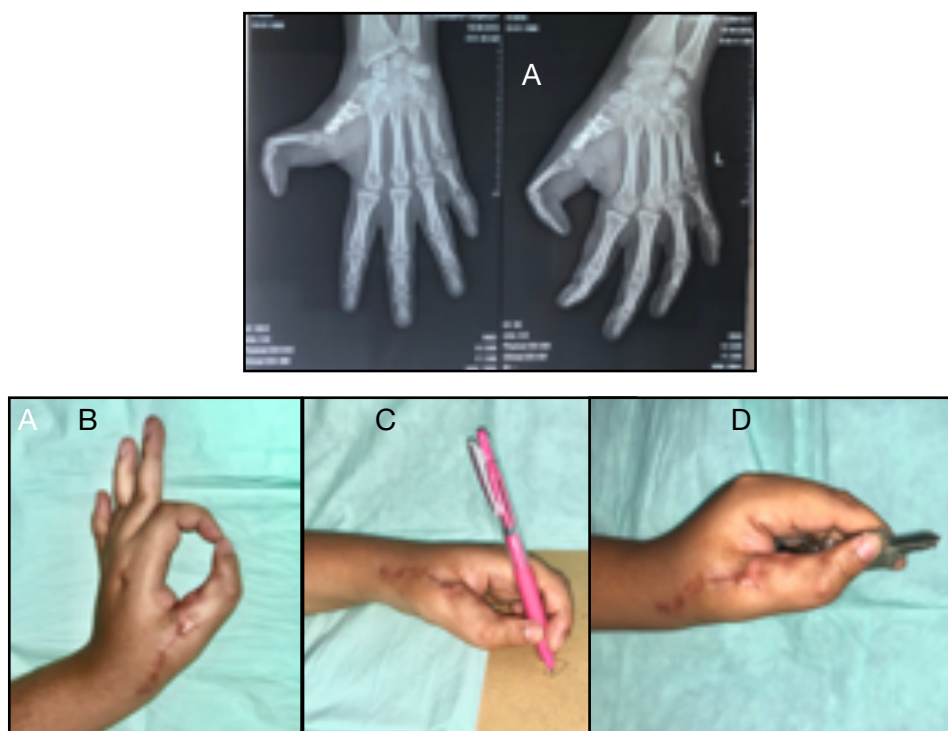
On the follow up, patient had satisfied abduction, adduction, flexion, extension, and pinching movement. Objective evaluation based on the assessment of a range of motion and stability of the MCP and IP joints. The metacarpals union was observed on the x-ray films and compared with the other side. The patient said she feels satisfied with the result; she pleased with the appearance that has no gap between 1st & 2nd finger, and the ability of return to work with no more protruding bone that causes snagging other objects. Unfortunately, the patient lost control so we cannot review gripping and pinching strength with goniometer.



Figures 1. Preoperative X-ray showed a Wassel type VI with triphalangeal thumb of the left hand



Figures 2. Design of Operate Procedure : (A)The insertion and the origin of abductor pollicis brevis. (B) Osteotomies of metacarpals, black areas were resected, no tendon or flap transfer. (C) Transported the radial digit to the top of the ulnar metacarpal then fixed it with fixtation plate and screw



Figures 3. Evaluation after surgery: (A) Postoperative X-ray showed (B-D) Excellent pinching and gripping function

DISCUSSION

Surgical intervention is required for all duplicated thumbs, as the potential for improved function and appearance, exceeds the risk of surgery. We must overcome more than just structural problems of the bone to achieve surgical correction. In all cases, the goals are a stable mobile thumb of appropriate size, shape, stability and mobility as well.⁸

Thumb movements, functional use, and status of the thenar musculature must be considered, because it is important. In Wassel types IV-VII, the thenar musculature may vary from normal to hypoplastic.^{7,12} The most important muscle of the thenar group is the abductor pollicis brevis (APB), which able to produce excellent opposition of the thumb.¹³ It arises from the scaphoid tubercle and trapezium¹⁴, and from its insertion into the base of the proximal phalanx, this muscle can stabilize the metacarpophalangeal joints (MCPJ) in abduction, flexion, and pronation and can assist extension of the terminal phalanx.¹³

Therefore, detailed history and physical examination are important,⁷ to perform surgical correction. In this case, we only did bony reconstruction, the resections of the distal of ulnar's metacarpal and the proximal of radial's

Our treatment is determined by the frequency of different methods used for treating duplicated thumbs and found that the overwhelming majority of cases were treated with resection and reconstruction (85%). Simple ablation alone (5%), the Bilhaut-Cloquet procedure (8%), pollicization (1%), and the on-top plasty (1%) are used much less frequently.^{7,12} The expected outcome of surgery in the case of osteotomy is correction of the deformity¹⁵.

In this case we performed metacarpals resection because we concerned with what our patient need, that is to bring the patient's thumb and index finger close, and address the prominent bone between them. The method used is to move the phalanges and an osteotomy metacarpal of the radial thumb to an ulnar thumb's metacarpal which was previously also reconstructed by performing a metacarpal osteotomy.

With reconstructive osteotomies for this case, we can make the finger closer and no more prominent bone due to the patient's wishes. However, a literature said if we do ray amputation,

we would opt to remove a digit including its metacarpal, and this surgery can cause a decrease in the pinch and grip strength as well¹². Amalgamated metacarpals were fixed with fixation plates. We chose plate and screw fixation because we concern about the stability of the thumb.

At the age of 1 and 2 year, where the pinch function is developing, reconstruction is typically performed, and also before the development of fine motor skills. The main goals of reconstruction are an the restoration and maintenance of function and pinch activity, also improved appearance.¹⁶

This surgical correction is improving the thumb function and what our patient need from the previous surgery at the age of 4-month. The metacarpal osteotomy is the best solution for this case. Repair of function is the main thing to consider in this operation while aesthetics is the added value.

SUMMARY

Polydactyl is a common occurrence. An understanding of anatomy and the reconstructive actions undertaken is not only about lifting a finger, but also improving the quality, function of the patient's hand, and the aesthetic aspect. An understanding of the type of polydactyl, the operation to be carried out, and an overview of the design of the operation are necessary, so we believed an osteotomy procedure to achieve a narrow distance between 1st and 2nd digit is the best way to do for this case. The patient and parents were pleased with the appearance and better function for pinching and gripping because the space between 1st and 2nd digit is no longer far in the distance, no more a prominent bone as well. In the follow-up evaluations; the union metacarpals were amalgamated, the moves were stable, no neuropathic problems, or other disorders were found.

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