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The relationship between trapezium union, CMC joint instability and function following pollicisation.		
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Introduction

Pollicisation is the procedure of choice for severe thumb hypoplasia and selected other upper limb abnormalities. Originally developed as a treatment for traumatic amputation of the thumb, the technique of pollicisation was adapted for use in children. The procedure has evolved to a defined, structured transfer of the index finger on it's neuro-vascular pedicle. Early development of this technique is attributed to Murray[1], Gosset[2], Bennell[3], and Littler[4], who began this work on adults. Dr Andrew 'Ben' Murray, an Australian surgeon, is believed to have performed the first pollicisation, described in 1946. Murray had an interesting history, who despite having lost his leg in a shooting accident and also suffering from ulnar nerve injury, continued to operate in the post war years. He was tragically shot by a disgruntled patient in Brisbane in 1955, after refusing to give a medical certificate for back pain. Hilgenfeldt[5] preferred transfer of the long finger due to the relative similarity of length between it's proximal phalanx and the first metacarpal, however this did not gain popularity over the index transfer. Littler, Matthews, Zancolli, Riordan and Malik all published papers between 1955 and 1971 extending the early principles to children with congenital thumb hypoplasia [6-10]. Buck-Gramcko further extended this work publishing a landmark paper[11] with a case series of 114 pollicisations of children, some affected by the thalidomide tragedy. The method described by Buck-Gramcko is widely accepted and used by modern hand surgeons with minor alterations.

Muller in 1937[12] described the increasing severity of thumb hypoplasia. In 1967 Blauth[13] developed a classification which defined the five grades of increasing severity used today. Manske and McCarroll redefined Blauth's grade III in 1992 as grade IIIA and IIIB[14]. The details of Blauth's grading are shown in Table 1.

Table 1. Grading of the hypoplastic thumb

Grade	Description	
	Minimal shortening and narrowing of all structures All musculoskeletal and neurovascular structures present	

Grade	Description	
II	Reduced bony width and length Superficial muscles of opposition hypoplastic or absent Mild extrinsic abnormalities Narrowing of the first web space +/- instability at the metacarpophalageal joint	
III	Absent proximal metacarpal Unstable metacarpophalangeal joint Extensive extrinsic and intrinsic musculotendinous deficiencies	
IV	Pouce flottant or floating thumb	
V	Absent thumb	

Some question the change to Blauth's original grading and division into IIIA and IIIB. Blauth's Grade III thumb lacked a metacarpal base and therefore a stable carpometacarpal (CMC) joint. Manske's IIIA thumb is defined with a stable CMC joint but differs from Grade II through the presence of extrinsic anomalies. This approach also suggests that extrinsic anomalies develop in an orderly manner

after the intrinsic anomalies. The concept of increasing hypoplasia of all structures with increasing grade is more realistic.

Grade I hypoplasia does not require surgical treatment. Grade II is generally treated with reconstruction and an opposition transfer for improved intrinsic function. Metacarpophalangeal (MCP) joint stabilisation and first web plasty are performed as indicated. Manske Grade IIIA thumbs are generally treated identically to Grade II, with appropriate attention to extrinsic anomalies [15, 16].

The indication for pollicisation is generally accepted to be an inadequate CMC joint (Blauth Grade III, Manske Grade IIIB). It is widely accepted that a thumb without a stable, functional CMC joint will not be effective, and a four digit hand (pollicised index finger and three other digits) capable of opposition is preferred. This is often difficult for parents to accept if the hypoplastic thumb is of reasonable size. Pollicisation for grades IV and V is generally well accepted. It is recognised by parents that any vestigial thumb is non-functional.

A more recent classification retains Blauth's distinction between Grades II and III - presence or absence of the proximal metacarpal - and subdivides Grade II according to increasing severity of hypoplasia[17, 18]. Grade IIA is defined as a hypoplastic thumb with MCP joint ulna-collateral ligament laxity. Extrinsic anomalies are present but do not demand reconstruction. Grade IIB is defined as having the features of Grade IIA plus global MCP joint instability and/or extrinsic anomalies requiring reconstruction, and grade IIC is defined as having features of Grade IIB plus an inadequate CMC joint. Grade III would retain the original skeletal description of Blauth, with absence of the metacarpal base. Buck-Gramcko's sub-classification which sub-divides Grade III according to whether one third or two thirds of the metacarpal is missing may also be helpful. Under this system, grade IIC thumbs may require pollicisation if reconstruction is unlikely to achieve a functional thumb.

Alternatives to index finger pollicisation may be indicated when maintenance of five digits is paramount. Some have transferred non-vascularised bone to reconstruct the hypoplastic metacarpal[19]. A recent paper from Chow et al[20] reported a series of six cases of hemi-longitudinal metatarsal transfer in China, where the concept of a four digit hand is culturally unfavourable. Problems arise from the failure to create an adequate CMC joint, however results of grip and pinch strength in this small cohort were comparable with pollicisation as were scores on the Jebsen test of hand function. Vascularised second toe metatarsophalangeal joint transfers have gained some popularity [21]. A series by Tu[22] reported parent satisfaction and adequate grip and pinch in eleven patients who had a similar procedure.

Other congenital abnormalities of the hand may warrant pollicisation. These include; Ulnar Dimelia (mirror hand), Multi-fingered hand, Macrodactyly and Ulnar deficiency[23]. A modified approach is required, as underlying anatomy is varied and not always predictable. Outcome in these patients is directly proportional to the quality of the transposed digit, and often results will not be as successful as pollicisation for classical thumb hypoplasia.

The results of pollicisation are generally good, both for function and appearance, with superior results obtained when the index finger is of good size and normal mobility and when the forearm and wrist are stable. Those with longitudinal radial deficiencies in the forearm and consequent wrist instability obtain poorer results from pollicisation[24-27]. Some studies have included associated conditions such as radial longitudinal deficiency in their results[28, 29], which has led to the reporting of abnormally poor outcomes. Most studies analyse patients with isolated thumb hypoplasia separate to those with associated radial dysplasia.

However, even with a normal or near normal forearm and wrist, the grip and pinch strengths are significantly compromised. A number of factors may be incriminated, including instability of the reconstructed joints and weakness of the musculotendinous unit reconstructions.

Various efforts have been made to quantify the outcome of pollicisation by examining case series. Most authors have examined a combination of strength, range of motion and function, whilst others have addressed appearance, and position. Some have attempted to combine these individual outcome parameters and develop a single global score for the pollicised digit[30, 31].

It is appreciated that comparison of pollicised digits is difficult given the anatomical variation of the index finger, difference in cognitive ability between children and variation of surgical technique between surgeons and between each individual case. Most have identified those with radial deficiency to have vastly inferior results and thus examined them as their own cohort. Historical results are presented below in the categories by which our cohort has been assessed.

i) Appearance

Subjective appraisal of appearance has been assessed by some authors. Most have used questionnaires for subjective appraisal from the parent and if possible the patient. Percival [30] included cosmesis as part of his assessment method and scored this as a combination of position and appearance. Appearance was based on whether the parent deemed the new thumb to look satisfactory. This method has been applied in other studies [25-27, 32, 33]. Goldfarb et al [34] evaluated the objective features of thirty-one pollicised digits including joint angulation, length, girth and nail width. They concluded that none of the thirty-one thumbs were considered to have the appearance of a normal thumb and that a pollicised digit is narrower, with decreased nail width. The study also addressed the subjective appearance of a pollicised thumb and assessed opinions of the caregiver, therapist and surgeon. The average VAS score for appearance was 6.6 (4.4-9.7) and was significantly higher from the caregiver than both the therapist and surgeon.

ii) Position

The length of the thumb has been assessed in a normal paediatric population[35]. Measurements were expressed as a ratio of distance between

a. the thumb interphalangeal (IP) joint crease to thumb tip, and

b. thumb IP joint crease to proximal interphalangeal (PIP) joint crease of the index finger.

If the thumb tip were equal with the index PIP crease, this ratio would return a value of 100%. Examination across an age range of 1 to 18 years with n=273 (546 hands) returned a mean ratio of 70%, indicating the tip of the normal thumb is proximal to the PIP joint of the index finger.

Goldfarb et al [34] measured a mean ratio of 90% in their cohort of thirty-one pollicised thumbs, suggesting that pollicised digits are statistically significantly longer than normal thumbs.

iii) Tenderness

No published data specifically reported a measure of tenderness at the operation site or with movement of the pollicised digit.

iv) Stability

Little has been written about CMC joint stability. Lochner et al [36] examined stability of the CMC joint post-pollicisation by examination of post-operative medical records and patient and parent complaints. The study accepted the lack of documentation of instability or notable complaints to reflect clinical stability of the CMC joint. Each pollicisation in the series was not clinically examined for stability by the authors.

v) Range of motion

Range of motion (ROM) has been assessed in multiple ways. Manske[24] measured ROM using a standard goniometer at the MCP, PIP and DIP joints of the pollicised digit, with his nomenclature referring to joints of a finger, that is, the pollicised index, rather than the joints of a thumb. These figures were added together to calculate total active motion (TAM). The normal flexion/extension ROM for a thumb has been published as 185 degrees including hyperextension, and normal TAM for a finger 260 degrees[37]. The study stated that because of restriction of motion at the MCP and IP joints caused by tendon shortening in the pollicisation procedure, that results would be compared with a normal thumb, rather than a normal finger. Results were divided into patients with associated conditions (RLD, five-finger hand, mirror hand) and patients with no associated conditions. These two groups were also considered together. The group with no associated conditions were reported to have a TAM of 146 degrees and the group with associated conditions 69 degrees with an overall range of 40-75% of a normal thumb.

Vekris et al[27] also measured TAM, measuring three joints. They reported a mean of 145(120-150) degrees for patients with an isolated thumb deficiency. Patients with an associated condition had a mean TAM of 96(65-120) degrees.

Staines et al[38] reported TAM as 89(50-135) degrees in the operated hand and 110 (100-145) degrees in the unaffected hand. This study only examined subjects with isolated thumb aplasia and no associated limb abnormalities. The

authors only included the MCP and IP joints of the new thumb, rather than the three joints measured by Vekris and Manske.

Kozin et al[28] measured flexion and extension at the IP and MCP joints but did not calculate a two joint TAM. The data from this paper can be used to calculate a two joint TAM of 107 degrees but it is unclear whether this includes patients with associated abnormalities including radial longitudinal deficiency. Other movements were assessed and mean abduction (presumed to be radial abduction) was reported as 62 (30-90) degrees and mean adduction as 19 (0-80) degrees. Roper et al[29] also measured radial and palmar abduction in 9 patients and reported the means as 44 and 53 degrees respectively. This series included many with associated radial dysplasia.

Historical data for total active motion is summarised in Table 2.

Table 2:Total active motion. Measurements of Manske and Vekris measure three joints, with the figures in brackets being for two joints. The measurements for Kozin and Staines are for two joints.

		ТАМ			
Article	n	No associated abnormalities	Associated abnormalities (eg RLD)	Total	
Manske 1992	28	146 (106)	69 (46*)	98 (67*)	
Vekris 2011	25	145 (113)	96 (57*)	124* (89*)	
Kozin 1992	14	unclear	69*	107*	
Staines 2004	12	89	n/a	n/a	

^{*} Calculated from published data.

Palmar abduction was measured using the *pollexograph* in a group of 21 patients with a hypoplastic thumb (Blauth II-IV) by De Kraker et al[39]. It was unclear whether or not any of these patients had undergone pollicisation, or if there was any associated radial dysplasia. It was concluded in the same study that the pollexograph gives a lower reading than traditional measurement of palmar abduction with a goniometer, but has significantly higher inter-observer and intra-observer reliability. Results of these studies are shown in Table 3.

Table 3: Palmar abduction

		Palmar abduction		
Cohort	n	No associated abnormalities	Associated abnormalities (eg RLD)	Total
De Kraker*^	21	47.5		47.5
Roper	9			53

^{*} Measured with pollexograph.

[^]Patients had not undergone pollicisation, but did have Blauth II-IV hypoplasia.

vi) Strength

Strength measures have been reported in multiple case series. Most include grip and some form of pinch strength. All studies used standard dynamometry (JAMAR) for grip and a pinch meter for varied pinch tests including tripod, tip and key pinch. Strength is dependent on age as well as the pre-operative quality of the pollicised digit and musculo-tendinous units and the presence or absence of wrist and forearm anomalies. For these reasons raw values reported in kilograms are of little use unless divided into age categories, which is often not practical with a small number of subjects. Results are usually reported as a percentage of either the unaffected hand or of age-matched normal values.

There are disparities in outcome between the studies that compared strength of the pollicised hand to the unaffected side, and studies that compared strength to established normal values. Staines et al[38] measured the unaffected side in unilateral pollicised patients without associated radial dysplasia. The grip, key pinch and tripod pinch strength were found to be 87 percent, 60.3 percent and 70 percent of age matched normal values. The study group published again five years later[40] and proposed that some studies that have evaluated hands compared to with opposite 'normal' hands might have inappropriately high outcomes, in other words that the 'normal' hand did not possess the same strength and function as an average age-matched hand from the general population. These investigators analysed the affected side against published age matched normal data and found grip, lateral (key) pinch and tripod pinch strength to be 36 percent, 25 percent and 32 percent of normal. The same group have recently published another follow up case series[31] of 22 hands in 18 patients. They have affirmed the non-operated hand may be weaker than normal dominant hands by measure of 50 percent, 58 percent and 62 percent for grip, lateral key pinch and tripod pinch strength respectively. Operated hands compared with normal non-dominant hands were reported as 29 percent, 27 percent and 31 percent for the same measures. They have also followed their cohort over time and observed age-related trends in strength, suggesting children aged three to five years often have grip and pinch strength in the normal range, for both operated and non operated hands. Patients over 5 years of age were almost unanimously below normal strength.

Vekris et al[27] reported strength comparative to the contralateral hand only, and although the paper did not discuss the specific results for associated and non-associated radial dysplasia groups, this can be gleaned from data published. Strength in those without associated radial dysplasia (n=14) was 88 percent, 64 percent and 60 percent for grip, key pinch and tip pinch respectively. For those with any associated radial dysplasia (n=7, Type 1-4 Bayne classification) strength was 52 percent, 52 percent and 47 percent for the same measures.

Manske et al[24] measured grip as well as tip, key and tripod pinch strengths. Results were expressed as a percentage of normative data and also divided into age groups. Patients with associated conditions (radial longitudinal deficiency, five fingered hand and ulnar dimelia) were analysed separately to those with isolated thumb hypoplasia. The study reported grip strength in patients with no associated conditions as 31 percent of normal, with key, tripod and tip pinch strengths 38 percent, 35 percent and 44 percent respectively. Those with

associated conditions were found to have a mean grip strength of 15 percent of an age matched normal, and key, tripod and tip pinch strengths of 14 percent, 16 percent and 16 percent respectively. Results did not suggest strength measures for patients below five years of age approached normal values.

Some studies have reported strength measures but have not divided results into those with and those without associated radial dysplasia[28, 29]. It is well established that those with associated radial dysplasia perform poorer[24, 26, 27] on strength tests, so unfortunately these results are not useful for comparison. Other studies[26] reported unilateral cases as a percentage of the non-operated hand and bilateral cases as a percentage of published normal values. Fortunately results were reported individually and it could be discerned as to which value each patient was compared. Results are included in Table 4, however low patient numbers in each group make mean scores recorded of questionable value.

Two articles[25, 32] used the scoring system developed by Percival[30] which included scores for tip pinch, pulp pinch and grasp (including strength), but these results could not be compared to normal values or with quantitative strength measures.

Studies reporting strength as an outcome post pollicisation are summated in Tables 4-7. It is noted whether strength was expressed as a percentage of agematched normal values (% normal) or a percentage of the opposite hand (% opposite hand/%opp).

Table 4: Grip strength: No associated radial dysplasia

Cohort	n	Grip Strength		
Conort		% normal	% opposite hand	
Staines 2004	10	36	35	
Vekris 2011	14		88	
Netscher 2013	22	29	60	
Manske 1992	10	31	24^	
Clark 1998	8*	64	62	

^{*5} bilateral cases compared with normals and 3 unilateral cases compared with opposite hand.

[^] Manske cited 24% strength of the opposite hand in his results for 10 subjects without associated conditions, however reported average grip strength (all subjects) in his discussion as 75% of the opposite hand.

Table 5: Grip strength: Associated radial dysplasia

Cohort	n	Grip \$	Strength
Conort		% normal	% opposite hand
Vekris 2011	7		52
Manske 1992	18	15	
Clark 1998	7*	8	29

^{* 3} bilateral cases compared with normals and 4 unilateral cases compared with opposite hand.

Table 6: Pinch strength: No associated radial dysplasia.

Oak and	Tip Pinch		Tripod Pinch		Key Pinch		
Cohort	n	%normal	%орр	%normal	%орр	%normal	%орр
Staines 2004	10			32	46	25	41
Vekris 2011	14		60				64
Netscher 2013	22			31	47	27	44
Manske 1992	10	44	27	35	26	38	24
Clark 1998	8*	64	63			61	53

^{* 5} bilateral cases compared with normals and 3 unilateral cases compared with opposite hand.

Table 7: Pinch strength: Associated radial dysplasia

Cahant		Tip F	Pinch	Tripod	Pinch	Key I	Pinch
Cohort	n	%normal	%орр	%normal	%орр	%normal	%орр
Vekris 2011	7		47				52
Manske 1992	18	16		16		14	
Clark 1998	7*	3	28			7	29

 $^{^{}st}$ 3 bilateral cases compared with normals and 4 unilateral cases compared with opposite hand.

vii) Function

Functional measures have also been assessed in multiple ways, making comparison between studies difficult. The Jebsen Hand Function Test (JHFT) has been used in modified form by multiple examiners. Manske[24] used 6 activities and reported the times for each as a percentage of normal. On average for the six

tasks, those with no associated conditions performed at 101 percent and those with associated conditions performed at 133 percent of the time of aged matched normal subjects.

Netscher[31] used the pegboard functional dexterity test and a modified Jebsen test and concluded that the JHFT 'correlates better with a subjective sense of ability than the pegboard and is more appropriate in these patients'. The heavy object item was not used so five activities were tested. On average, patients performed in significantly slower time in three activities. The other two activities were also slower but these were not statistically significant. Results were not expressed as a percentage. These results were similar to results from the same cohort of patients published earlier[38, 40].

Some examiners have measured function using a set battery of tasks[24, 27, 29] or other methods like a Perdue board[28]. These tasks are helpful to get an indication for usage of the new thumb, but are not standardised and comparable between trials.

Buffart et al [41] found a correlation between activity performance on functional tasks and quantitative measures including strength and range of motion in a cohort of twenty-two children with radial longitudinal deficiency. They suggested the that a larger range of motion was a more linear correlation with function than strength. They also concluded the greater the degree of radial deficiency, the greater the deficit in all three measures.

viii) Satisfaction

Golfarb et al[34] asked the surgeon, therapist and caregiver to complete a visual analogue scale (VAS), focussed on cosmesis rather than overall function. The caregiver recorded a significantly higher score (mean = 7.3) compared with the surgeon (mean = 6.4) and the therapist (mean = 6.0). Ceulemans et al [32] also used a VAS, however encompassed aesthetic and functional outcome. They reported the mean result for the examiner to be 7.8 and the parent 7.6.

ix) Radiology

The created CMC joint has recently been examined by Lochner et al[36] in a group of 85 pollicisations in 74 patients, by retrospective review of medical records. 20 patients did not have bony union between the index metacarpal head and base. Of these only three were clinically unstable requiring surgical stabilisation. 10 patients had evidence of CMC subluxation on plain radiographs of which one was symptomatic requiring reconstruction. Physeal arrest in the index proximal phalanx was noted in 21 of these patients.

Stress view radiographs have not been examined in any case-series to date. Integrity of the index metacarpal base-head complex and stability of the created CMC joint under stress post-pollicisation have not been studied. Systems used to quantify radiological displacement of the CMC joint in other populations[42] were deemed non-applicable to our cohort given the altered anatomy of the thumb base.

Surgical technique

The technique of pollicisation of the index finger has been described in depth by many[4, 43-46] and is perhaps best appreciated in the description by Buck-Gramcko[11]. In brief, the index finger MCP joint becomes the new thumb CMC joint. The finger PIP joint becomes the thumb MCP joint and the DIP joint becomes the thumb IP joint. The extensor indicis proprius and extensor digitorum communis are shortened by the length of the skeletal shortening and become respectively the extensor pollicis longus and abductor pollicis longus musculotendinous units of the new thumb. Some have demonstrated improved IP joint motion and flexion strength with shortening of the the flexor digitorum profundus[47] but most allow this to shorten with time unless pollicisation is performed at an older age. The first dorsal and palmar interossei are shortened and connected to the mobilised lateral bands of the index finger extensor mechanism to become the abductor pollicis brevis and adductor pollicis of the thumb respectively. Appropriate skin incisions allow redistribution of skin flaps to create a first web of satisfactory depth and width.

An integral part of the success of a pollicisation is the creation of a new CMC joint and there are a number of principles in reconstruction which would appear to be necessary:

- 1. Optimal positioning of the new thumb ray in palmar abduction, radial abduction and appropriate rotation;
- 2. Placement of the thumb ray in an anterior plane to that of the finger CMC joints;
- 3. Hyperextension of the index finger MCP joint to prevent a hyperextension deformity of the new CMC joint.

It is difficult to satisfy all of the above parameters and place the cancellous surfaces of the index finger metacarpal head and base in direct apposition.

Most use the method described in Buck-Gramcko's landmark 1971 article [11] in which he advises rotation of the index metacarpal head into full flexion to prevent an hyperextension deformity of the new CMC joint, although some have found case to case variability in this position[48]. Buck-Gramcko suggested retention of the metacarpal base to be necessary in cases with relatively short phalanges. In these cases, the metacarpal head was fixed to the base using one or two Kirschner wires. If the phalanges were of normal length, his initial description did not retain the metacarpal base, and the metacarpal head was sutured to the joint capsule and carpal bones. Subsequently most, including Buck-Gramcko, have preferred to retain the base [23, 44-46]. The plane of osteotomy of the metacarpal is varied, with a transverse osteotomy at the metacarpal base and an oblique osteotomy in either coronal or sagittal planes all described[48]. Some prefer K-wire fixation to promote head to base union as described by Buck-Gramcko [11, 23, 29, 46]. Some eschew this [44]. Manske wrote of the importance of a fibrous union rather than a bony union between the retained base and head [24, 45], creating a pseudarthrosis at this articulation. He proposed that using sutures rather than K-wires for fixation permitted increased mobility of the new thumb.

The surgical technique used for the patients in this cohort was recently published [18] and is included as an appendix (Appendix 3). Of note is the preference for bony union at the metacarpal base-head junction which stabilises the new trapezium. No patients in the cohort underwent flexor tendon shortening.

Aims of this study

The impact of bone union between the new trapezium and the metacarpal base on functional outcomes has not been determined. Most authors agree that an inadequate CMC joint, or a hypoplastic first metacarpal is the indication for pollicisation in congenital thumb hypoplasia. The pollicisation procedure aims to create an adequate CMC joint, but the stability of the new joint has not been well addressed in any study of pollicised patients. The primary outcomes of range of motion, strength and function have been well documented in isolation, but have not been related to either bony union or to CMC joint stability. This study addresses these questions, with the primary aims outlined below.

- 1. What is the union rate between metacarpal head (new trapezium) and metacarpal base when this is the surgeons intended outcome?
- 2. If non-union occurs, is the metacarpal head/base complex stable?
- 3. Does this bony union alter the stability of the new CMC joint?
- 4. Does bony union of the new trapezium to the metacarpal base or CMC joint stability have any effect on functional outcome, including strength and range of motion?

Secondary aims were to assess results according to the presence or absence of a forearm/wrist anomaly and to assess the function of the opposite thumb when it was previously considered to be clinically normal.

Materials and Methods

Ethics and governance approval was sought from the Human Research Ethics Committees of Northern Sydney Local Health District, the Sydney Children's Hospital Network and North Shore Private Hospital. All committees provided their approval (see Appendix 1). The University of Sydney Human Research Ethics Committee acknowledged the right to proceed with this study (Appendix 2).

Between 1990 and 2012 pollicisation of a digit was performed on 90 hands in 79 patients by a senior surgeon in Sydney, Australia. The procedure was performed at one of three centres. 35 patients with 44 affected hands were able to attend a follow up appointment for data collection for this study. Of the 44 patients who were unable to attend, 14 were unable to be contacted, 3 had subsequently died from associated pathology or other causes following the procedure and 27 were unable to attend at convenient times.

Of the 35 patients who were able to attend, 19 were male and 16 female. There were 16 right hands, 10 left hands and 9 bilateral cases. Median age at operation was 1 year 10 months (Range 10 months-14 years 6 months). Of the 40 patients with thumb hypoplasia, 14 were classified as a Blauth grade III, 14 as a grade IV and 12 as a grade V. Four patients did not present with typical thumb hypoplasia. Two were cases of ulnar dimelia, one a five fingered hand with rudimentary thumb and associated Townes-Brocks Syndrome and another with a hypoplastic triphalangeal thumb.

Twelve hands in ten patients required additional surgery for associated radial longitudinal deficiency, which was performed prior to pollicisation. The 28 remaining hands with thumb hypoplasia did not require any wrist/forearm corrective surgery.

Revision surgery was performed on five hands. Two had the first web space revised and scar debulked. One child had a fall and on subsequent examination was found to have an aneurysm which was removed. The child with a five fingered hand had a metacarpal base-head non union which was fixed with circlage wires. The final revision involved a flexor digitorum superficialis IV to extensor pollicus longus tendon transfer for lack of radial abduction and opposition.

The limb abnormality was associated with a generalised syndrome in 21 cases, with the distribution shown in Table 8. In three patients a pattern of congenital abnormalities was observed but no syndrome or association was identified. One patient had limb abnormality associated with Valproate embryopathy.

Table 8: Distribution of genetic anomalies

Genetic Abnormality	Number
Nil	10
VACTERL association	10
Holt-Oram Syndrome	5
Unidentified	3
Goldenhar Syndrome	2
TAR (Thrombocytopaenia Absent Radius)	1
Baller-Gerold Syndrome	1
Townes-Brock Syndrome	1
Rothmund-Thomson Syndrome	1
Valproate Embryopathy	1

Assessment

Examination was performed by two examiners; a fellow of hand surgery, and a masters student. Radiographs were reviewed by the supervising surgeon.

i) Appearance

Size of the pollicised digit was rated as good, fair or poor as was the contour of the thenar eminence according to Table 9.

Table 9: Appearance

Rating	Size	Contour
Good	The width and length of the pollicised digit were very similar to the contralateral thumb	The thenar eminence was of sufficient bulk to give the hand a very similar appearance to the contralateral side. The first web space commenced at the neo-MCP joint.
Fair	The width and length of the pollicised digit were marginally different to the contralateral thumb	The thenar eminence was of comparable bulk to give the hand a satisfactory appearance compared with the contralateral side. The first web space commenced close to the neo-MCP joint.
Poor	The width and length of the pollicised digit were considerably different to the contralateral thumb	The thenar eminence was of considerably different bulk to give the hand an unsatisfactory appearance compared with the contralateral side. The first web space did not commence close to the neo-MCP joint.

Any difference in colour of the pollicised digit or transposed skin flaps was noted.

ii) Position

The distance from the tip of the new thumb to the PIP joint of the next ulnar digit was measured, with the thumb adducted to the side of that adjacent digit. These measures were repeated on the contralateral hand.

Measurements were recorded as a positive value if the thumb extended distal to the index PIP joint and a negative value if the thumb remained proximal to the joint line. Results were divided into those thumbs shorter than, equal to and longer than this reference point.

The MCP and IP joints of the new thumb were observed for radial or ulnar deviation, which was quantified in degrees using a standard goniometer. Deviation was also assessed for the MCP and IP joints of the opposite thumb.

iii) Tenderness

Pressure tenderness was assessed in both the anatomical snuff box and at the CMC joint of both thumbs.

iv) Stability

Ligamentous stability was tested using a standard clinical radial and ulnar collateral stress test at the CMC joint. For the test, the new trapezium was held in a fixed position, and radial or ulnar stress applied to the new metacarpal. An end point was determined by the examiner and the result quantified with a standard goniometer. An unstable joint was determined to deviate greater than a total of fifty degrees in both directions, or not have a firm end point, indicating lack of ligamentous and soft tissue integrity.

v) Range of motion

Joint range of motion at the wrist joint was measured with a standard goniometer. The wrist was assessed for subluxation in both a radio-ulnar plane and dorsal-volar plane, and subsequently categorised as stable or unstable. MCP and IP flexion of the thumb was measured with a hand goniometer. Radial abduction was measured as the angle between the first and second metacarpals. Palmar abduction was measured both at the thumb tip and at the metacarpal using the *pollexograph*™ which gives a measurement in degrees. Range of motion readings from the pollexograph were compared with published paediatric normal values[49] which were not age-matched. Retroposition was assessed in millimetres using a ruler, and compared with normal values, again not age-matched.

All range of motion measures were for active movement. If a patient was unable to complete a movement they were excluded from analysis for that movement.

It was observed whether the patient could make a fist on each hand, which entailed flexion of the IP and MCP joints of all fingers with a flexed and palmar abducted thumb on the outside of these fingers.

vi) Strength

Both intrinsic and extrinsic strength was measured on both hands. The ability of the patient to perform each strength task was rated on a scale of 0-4 as shown in Table 10.

Table 10: Ability of patient to perform strength tasks

Not able to perform task	0
Grips but can't perform task	1
Abnormal but performs task	2
Minimally abnormal	3
Normal	4

If a patient was able to achieve levels 3 or 4, each measure was repeated three times, recorded and a mean calculated.

Power grip was assessed using a JAMAR dynamometer. The dynamometer aperture was adjusted according to hand size. If there was doubt as to which setting would be more successful, both were trialled and the greater measure taken.

Tripod pinch, key pinch and tip pinch were calculated using a pinch dynamometer. Some children were unable to produce sufficient force to obtain a reading on the scale.

Palmar abduction and opposition force were measured on both thumbs using the *Rotterdam Intrinsic Hand Myometer*^m, for which reliability has been affirmed in children[50].

Strength measures were compared with aged matched norms as well as the contralateral hand. Due to the large variability of age in our cohort, it was sometimes necessary to use different normative data sets when analysing the same parameter. For grip strength: one data set [51] was used for 4-12 year olds, another for 12-19 year olds[52] and another for any patient greater than 19 years old[53]. For pinch strength: one data set was used for 5-12 year olds[54], one for 12-19 years olds[52] and another for those greater than 19 years old[53]. The literature contains data for the Rotterdam Intrinsic Hand Myometer for children aged 4-12 years old[55] and data is accessible for those greater than 12 years old [56]. No normative data was available for children less than four years old.

vii) Function

Function was assessed using a modified Jebsen Test of Hand function (JHFT) [57] which included the tasks listed in Table 11. Each patient was prompted to use their pollicised digit to complete each task. If patients were not able complete the task using this digit, a result was noted in the 'fingers' column in shown in Table 11. The patient was given one subsequent attempt at the task using their thumb, and if unsuccessful were excluded from analysis.

Table 11: Modified Jebsen Test of Hand Function

Task	Time (sec) for R		Time (sec) for L	
	Thumb	Fingers	Thumb	Fingers
Turn over 5 cards				
Pick up 5 small objects and place in can				
Stack 4 checkers				
Place 5 large light objects on board				
Place 5 large heavy objects on board				

Cards slightly larger than standard paying cards were used. Children were asked to manipulate and turn the card on a table, and not permitted to slide the card to the edge of the table and then turn.

Small beads were scattered in a small area and the child instructed to pick up one at a time, and place in an adjacent empty can.

Empty standard sized tin cans were used as light objects, which were placed on 5 cards placed next to them. A full standard sized tin can (375g) was used as the heavy object which was placed on to the same cards at the same distance.

Results were compared with age-matched normals reported by Taylor et al [58]. The data for non-dominant hands was used in all cases for comparison. Data was not available for children under six years of age. Patients in our cohort aged twenty and above were compared with the values published for those aged nineteen, as this was the upper limit of Taylor's study population.

viii) Satisfaction

After tests were completed, the examiners subjectively rated both the function and cosmesis as excellent, good, fair or poor. Parents were asked to complete a questionnaire with the same parameters. Patients were asked to complete an additional questionnaire if at an appropriate age of understanding.

ix) Radiology

Five x-ray views were taken of each patient. These included postero-anterior (PA) views of the hands and thumb, as well as of the forearms if the patient had any type of radial dysplasia. A lateral view of the thumb was also taken. Two stress-view radiographs of both CMC joints were taken, one with ulnar stress applied to the joint, and one with radial stress applied. In each case the stress was applied by an examiner by manually fixing the proximal bone (the new trapezium and retained metacarpal base), and providing the stress to the bone distal to the joint (the new thumb metacarpal). Stress-view radiographs were taken as a PA view. Each image was evaluated in the radiology department to ensure an adequate view of the CMC joint space. Attention was paid to prevent any pain when the stress was applied. Parents were used when necessary for restraint or positioning of a child.

Radiographs were evaluated for three parameters, demonstrated in Figures 1-4.

1. Bony Union: Was there fusion between the metacarpal head and base?

Figure 1: PA radiograph demonstrating bony union at the new trapezium and retained metacarpal base



2. Trapezium stability: Was the metacarpal head-base complex stable under stress?

Figure 2: Stress view radiograph. Red arrow indicating metacarpal head/base and yellow arrow indicating CMC joint. These X-rays demonstrate non-union and instability of the trapezium.



3. Carpo-metacarpal joint stability: Was there CMC joint angulation or displacement when placed in a position of radial or ulnar stress?

The methods shown in Figure 3 were used. Two lines were drawn on the longitudinal axes of the metacarpal and trapezium (lines A and B). An additional two transverse lines were drawn in the planes of the articular surfaces of the CMC joint (lines C and D). An average of the two angles subtended by these lines (angles X and Y) was calculated when both radial and ulnar stress was applied. If

the angle total (radial + ulnar stress) was greater than or equal to fifty degrees, the CMC joint was deemed unstable.

Figure 3: Measurement of angulation

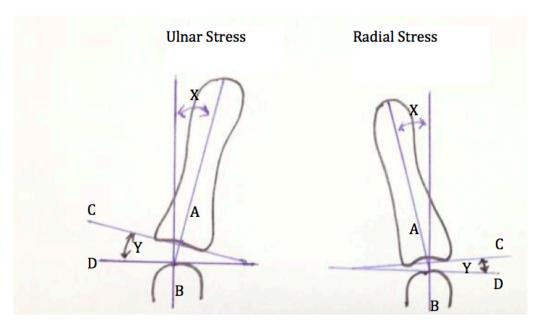


Figure 4: Measurement of angulation on a radiograph.



Displacement was measured as demonstrated in Figure 5. A point was drawn at the midpoint of each bony articular surface at the CMC joint, point A and point B. After radial or ulnar stress was applied, the percentage of displacement of point A on point B was measured, in relation to the width of the articular surface of the

metacarpal base. If the two points were measured as being separated by greater than 25% of the articular width in either direction, the CMC joint was deemed unstable.

Figure 5: Measurement of displacement

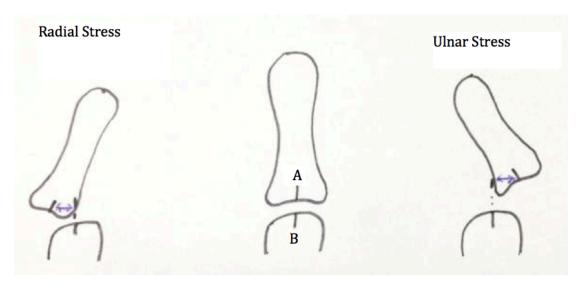


Figure 6: Measurement of displacement on a radiograph. This joint is considered to be stable as the displacement is less than 25%.



Each radiograph was reviewed by both examiners as well as the supervising surgeon.

Statistical Analysis

For statistical analysis SPSS 20.0 software was used. Descriptive statistics were used to measure the distribution of grip strength, range of motion, JHFT and the pattern of thumb use between the group with isolated thumb hypoplasia and those with associated RLD. The results were compared with age related normal values of range of motion, grip strength and JHFT as percentages. For comparisons between groups, statistical analyses were performed using the t-test for grip strength, JHFT and range of motion variables. The chi-square test was used for thumb use pattern analysis between groups. Statistical significance was set at $\alpha \le 0.05$.

Results

Assessment

i) Appearance

Colour abnormalities were observed between the border of the transposed flap and hand in three patients with darker skin. One 17 year old child had begun to grow hair on the skin transposed to his thenar eminence.

The subjective evaluation of digit size and contour is shown in Table 12.

Table 12: Evaluation of digit size and contour

		Good	Fair	Poor	Not recorded*	Total
	No assoc cond.	22	4	0	2	28
Digit Size	Assoc cond.*	7	8	0	1	16
	All	29	12	0	3	44
	No assoc cond.	14	12	1	1	28
Contour	Assoc cond.*	4	11	0	1	16
	All	18	23	1	2	44

^{*} Associated conditions include radial longitudinal deficiency, ulnar dimelia, five-fingered hand and hypoplastic triphalangeal thumb.

ii)Position

The mean distance from the tip of the thumb to the PIP joint crease is shown in Table 13. Six pollicised digits were shorter than the crease, thirteen equal to it and twenty-two longer. Three measurements were not recorded.

Table 13: Thumb tip to PIP joint flexion crease distance

	Number of thumbs proximal to PIP joint	Mean distance proximal to PIP joint (mm)	Number of thumbs at PIP joint	Number of thumbs distal to PIP joint	Mean distance distal to PIP joint (mm)
No associated conditions	2	5.5	7	16	4.8
Associated conditions*	3	6.7	5	4	7.3
Other	1	20	1	2	6.5
Total	6	8.5	13	22	5.4

^{*} Associated conditions include radial longitudinal deficiency, ulnar dimelia, five-fingered hand and hypoplastic triphalangeal thumb.

Radial or ulnar deviation at the IP joints in a resting position was apparent in five affected thumbs and four unaffected thumbs. MCP joint deviation was observed in three affected and three unaffected thumbs. There was no tendency to a radial or ulnar direction.

iii)Tenderness

No children exhibited tenderness with pressure applied over their anatomical snuff box or CMC joint.

iv)Stability

Clinically, six operated thumbs exhibited instability at the CMC joint. Of non-operated thumbs, twelve CMC joints were clinically unstable by our own parameters.

v)Range of Motion

Range of motion at the wrist, ability to form a fist and stability of the wrist are summated in Tables 14 and 15.

Table 14: Range of motion: Wrist. All measures expressed as a mean in degrees.

	Flexion	Extension	Radial Deviation	Ulnar Deviation
No associated conditions	64	47	34	31
Associated conditions*	31	12	21	7
All	52	35	29	22

^{*} Associated conditions include radial longitudinal deficiency, ulnar dimelia, five-fingered hand and hypoplastic triphalangeal thumb.

Table 15: Wrist stability and ability to form a fist

	Fist^	Stable wrist^
No associated conditions	24/24	24/28
Associated conditions*	5/16	11/15
All	29/40	35/43

[^]Data not collected for some children due to compliance

Range of motion of the new thumb is shown in Table 16. All measurements are expressed in degrees with the exception on retroposition which is expressed in millimetres.

Table 16: Range of motion: Thumb

	IP joint + MP joint flexion	Radial Abduction	Palmar Abduction	Retroposition (mm)
No associated conditions	91	47	49	20
Associated conditions*	43	46	53	4
All	74	47	50	14

^{*} Associated conditions include radial longitudinal deficiency, ulnar dimelia, five-fingered hand and hypoplastic triphalangeal thumb.

Range of motion at the CMC joint is multi-planar, and was measured with palmar abduction, radial abduction and retroposition. These measurements were then expressed as a percentage of normative values, which are summarised in Table 17.

Table 17: CMC joint range of motion, mean percentage of normal values.

	Palmar Abduction (SD)	Radial Abduction (SD)	Retroposition (SD)
Isolated Thumb hypoplasia	80.1 (20.5) n=28	78.0 (21.8) n=28	70.7 (46.0) n=24
RLD	88.6 (23.9) n=12	69.4 (20.5) n=12	35.4 (18.7) n=4
Other*	86.5 (36.5) n=4	89.6 (55.0) n=4	25.7 (18.2) n=2
Total	83.0 (22.7) n=44	76.7 (25.5) n=44	63.0 (44.4) n=30

^{*}Other = Ulnar dimelia, five fingered hand, hypoplastic tri-phalangeal thumb

^{*} Associated conditions include radial longitudinal deficiency, ulnar dimelia, five-fingered hand and hypoplastic triphalangeal thumb.

vi)Strength

Strength measurements are shown in Tables 18-20. Values recorded are expressed as a percentage of normal values, and also as a percentage of the non-operated hand. Patients with surgery performed on the contralateral hand (pollicisation or reconstruction) were excluded from the latter. Patients who scored 0, 1 or 2 for ability to complete the grip strength task were excluded.

Grip strength measurements were compared to available age-matched normal values and shown in Table 18. Due to the large age variability in our cohort, three different sets of published normals were used for ages four to twelve, thirteen to eighteen and nineteen and older[51-53]. No data was available for children under four years of age.

Table 18: Strength: Grip, measured with JAMAR dynamometer

Condition	Mean percentage of age- matched normal values (SD)	Mean percentage of contralateral hand
Isolated Thumb hypoplasia	51.2 (20.1) n=28	89.6 n=13
RLD	8.3 (4.6) n=10	7.9 n=4
Other*	54.0 (45.6) n=2	61.6 n=2
Total	40.6 (26.4) n=40	69.4 n=19

^{*}Other = Ulnar dimelia, five-fingered hand

Two sets of normative data were also used for comparison of measures of tip, tripod or key pinch, which were divided into those aged five to twelve years old, and those thirteen and above [52, 54]. No data was available for children under five years of age. Results are shown in Table 19.

Table 19: Strength: Pinch, measured with a pinch dynamometer

	Tip pinch		Tripod	l Pinch	Key Pinch	
	% norms (n)	% contra hand (n)	% norms (n)	% contra hand (n)	% norms (n)	% contra hand (n)
Isolated Thumb hypoplasia	22.1 (17)	51.5 (8)	27.6 (15)	42.2 (6)	27.9 (14)	38.9 (8)
RLD	1.7 (1)	3.75 (1)	2.0 (1)	2.9 (1)	0.8 (1)	1.3 (1)
Other*	20.9 (1)	56.3 (1)	6.0 (1)	20.0 (1)	14.2 (1)	28.5 (1)
Total	21.0 (19)	47.2 (10)	24.9 (17)	34.5 (8)	25.3 (16)	34.1 (10)

^{*}Other = Five-fingered hand

Intrinsic muscle strength was measured with the Rotterdam Intrinsic Hand Myometer, for which normative values exist for children aged four to twelve [55] and also for adults [56]. Unfortunately no data exists for those aged between thirteen and adulthood. Children in this age bracket were compared with adult values. Results are shown in Table 20.

Table 20: Strength: Palmar Abduction and Opposition, measured with Rotterdam Intrinsic Hand Myometer

	Palmar Abduction		Opposition		
	% norms (n)	% contralateral hand (n)	% norms (n)	% contralateral hand (n)	
Isolated Thumb hypoplasia	21.2 (26)	82.0 (13)	14.9 (26)	93.0 (13)	
RLD	5.6 (11)	43.3 (4)	4.6 (12)	28.1 (5)	
Other*	26.2 (3)	59.6 (3)	12.0 (3)	45.0 (3)	
Total	17.3 (40)	70.9 (20)	11.7 (41)	70.7 (21)	

^{*}Other = Two Ulnar dimelia, one five-fingered hand

Both grip and pinch strengths were assessed on the normal, non-operated side and compared to normative values. Patients with surgery performed on this hand (pollicisation or reconstruction) were excluded from this analysis. Results are shown in Table 21.

Table 21: Strength: Normal, non-operated hand expressed as percentage of age-matched normal values.

	Grip (n)	Tip pinch (n)	Tripod pinch (n)	Key pinch (n)
Isolated Thumb hypoplasia	78.6 (14)	52.1 (10)	56.9 (12)	63.7 (12)
RLD	75.8 (5)	31.2 (4)	73.6 (4)	54.6 (4)
Other*	94.1 (3)	22.9 (3)	47.3 (3)	58.3 (3)
Total	80.2 (22)	42.0 (17)	58.9 (19)	61.1 (19)

^{*} Other = Two Ulnar dimelia, one five-fingered hand

vii)Function

Results from the Jebsen test of hand function were compared with available agematched norms [58]. Results are displayed in Table 22 as a percentage of these norms, with a higher percentage reflecting a slower time to complete the task.

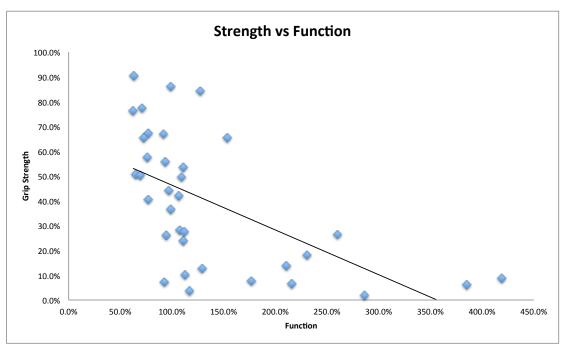
Table 22: Jebsen Test of Hand Function, percentage of age-matched normal values

	n	Isolated Thumb hypoplasia (SD)	RLD (SD)	Other* (SD)
Turning Cards	32	127.6 (115.9) n=22	267.8 (150.4) n=9	120.4 n=1
Stacking Checkers	34	74.7 (28.1) n=23	206.9 (220.5) n=10	80.0 n=1
Pellets in Cans	34	75.2 (33.2) n=23	106.8 (64.2) n=10	73.3 n=1
Lifting Light objects	36	117.3 (37.9) n=23	250.7 (106.3) n=12	119.4 n=1
Lifting heavy objects	33	119.3 (32.2) n=23	238.1 (95.9) n=9	109.4 n=1
Total Time	36	100.3 (41.7) n=23	229.1 (113.7) n=12	98.5 n=1

^{*} Five-fingered hand

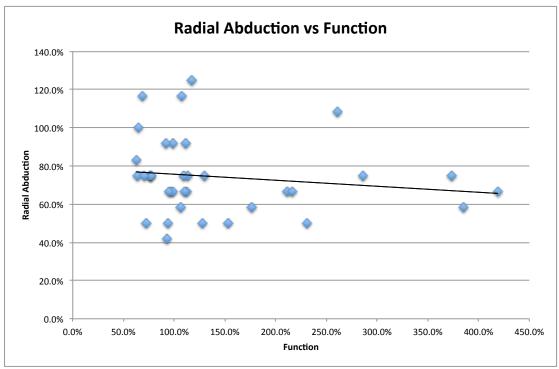
Higher grip strength and better radial abduction range of motion were indicative of a lower total time on the JHFT. Figures 7-8 show the relationship between these variables. Note that a lower percentage for the JHFT is equivalent to a better result than a higher percentage.

Figure 7: Grip Strength vs Function



^{*} Grip strength and the total time for Jebsen Hand Function Test (JHFT) expressed as percentage of agematched normal values.

Figure 8: Radial Abduction vs Function



^{*} Radial abduction and the total time for Jebsen Hand Function Test (JHFT) expressed as percentage of agematched normal values

viii)Satisfaction

Table 23 details the results of parent and doctor satisfaction questionnaires.

Table 23: Satisfaction

	Doctor Satisfaction		Parent Satisfaction	
	Function	Cosmesis	Function	Cosmesis
Excellent	19	10	15	14
Good	9	19	13	16
Fair	11	12	4	3
Poor	2	0	3	2

ix)Radiology

Radiographs were evaluated for metacarpal head/base bony union, metacarpal head/base stability and CMC joint stability; the raw numbers are presented in Table 24.

Table 24: Evaluation of radiographs

	Metacarpal head/ base bony union		Metacarpal head/ base stability		CMC joint stability	
	Yes	No	Stable	Unstable	Stable	Unstable
Isolated Thumb Hypoplasia	25	3	28	0	21	6
RLD	8	4	9	3	9	3
Other*	4	0	4	0	4	0
Total	37	7	40	3	34^	9

 $^{{}^{\}star} O ther \ conditions \ include \ ulnar \ dimelia, \ five-fingered \ hand, \ triphalangeal \ thumb$

Table 25 demonstrates the distribution of CMC joint stability and metacarpal head/base bony union.

Table 25: Bony Union vs CMC joint Stability

		CMC joint stability		
		Stable	Unstable	Total
	Union	28	8	36
Bony Union	Non-Union	6	1	7
	Total	34	9	

[^]One child did not have a stress-view performed

The relationship of palmar abduction range of motion with CMC joint stability and metacarpal head/base bony union is shown in Tables 26-27.

Table 26: Palmar Abduction range of motion, measured to thumb tip on the Pollexograph, compared with CMC joint stability: Affected Thumbs

	Palmar Abduction ROM as mean % of age-matched normal (SD)				
	Stable CMC joint	Unstable CMC joint	p value		
Isolated thumb hypoplasia n=27	81.7 (20.5) n=21^	76.9 (22.5) n=6^	0.62		
Thumb hypoplasia with RLD n=12	89.7 (25.2) n=9	85.1 (23.8) n=3	0.79		
Thumb hypoplasia with other associated conditions* n=4	86.5 (36.5) n=4				
Total n=43	84.4 (23.3) n=34	79.6 (21.8) n=9	0.58		

[^] One child did not have a stress-view performed

Table 27: Palmar Abduction range of motion, measured to thumb tip on the Pollexograph, compared with bony union: Affected Thumbs

	Palmar Abduction ROM as mean % of age-matched normal (SD)				
	Bony Union	Non-Union	p value		
Isolated thumb hypoplasia n=27	81.5 (21.2) n=25	68.6 (4.8) n=3	0.31		
Thumb hypoplasia with RLD n=12	88.6 (23.6) n=8	88.6 (28.0) n=4	1.00		
Thumb hypoplasia with other associated conditions* n=4	86.5 (36.5) n=4				
Total n=43	83.6 (23.0) n=37	80.0 (22.7) n=7	0.71		

 $^{{}^*\}mathrm{Other}$ conditions include ulnar dimelia, five-fingered hand, triphalangeal thumb

^{*}Other conditions include ulnar dimelia, five-fingered hand, triphalangeal thumb

The relationship of radial abduction range of motion with CMC joint stability and metacarpal head/base bony union is shown in Tables 28-29.

Table 28: Radial Abduction range of motion compared with CMC stability: Affected Thumbs

	Radial Abduction ROM as mean % of age-matched normal (SD)				
	Stable CMC joint	Unstable CMC joint	p value		
Isolated thumb hypoplasia n=27	77.4 (20.9) n=21	81.9 (27.6) n=6^	0.66		
Thumb hypoplasia with RLD n= 12	74.1 (21.0) n=9	55.6 (12.7) n=3	0.19		
Thumb hypoplasia with other associated conditions* n=4	89.6 (55.0) n=4				
Total n=43	77.9 (25.9) n=34	73.1 (26.3) n=9	0.63		

[^] One child did not have a stress-view performed

Table 29: Radial Abduction range of motion compared with Bony Union: Affected Thumbs

	Radial Abduction ROM as mean % of age-matched normal (SD)				
	Bony Union	Non-Union	p value		
Isolated thumb hypoplasia n=28	80.0 (22.0) n=25	61.1 (9.6) n=3	0.16		
Thumb hypoplasia with RLD n=12	72.9 (23.0) n=8	62.5 (14.4) n=4	0.43		
Thumb hypoplasia with other associated conditions* n=4	89.6 (55.0) n=4				
Total n=44	79.5 (26.5) n=37	61.9 (11.6) n=7	0.09		

^{*}Other conditions include ulnar dimelia, five-fingered hand, triphalangeal thumb

^{*}Other conditions include ulnar dimelia, five-fingered hand, triphalangeal thumb

The relationship of retroposition with CMC joint stability and metacarpal head/base bony union is shown in Tables 30-31.

Table 30: Retroposition range of motion compared with CMC Stability: Affected Thumbs

	Retroposition ROM as mean % of age-matched normal (SD)				
	Stable CMC joint	Unstable CMC joint	p value		
Isolated thumb hypoplasia n=23	69.0 (41.5) n=17	86.3 (55.4) n=6	0.43		
Thumb hypoplasia with RLD n=4	35.4 (18.7) n=4				
Thumb hypoplasia with other associated conditions* n=2	25.7 (16.2) 11=2				
Total n=29	59.4 (40.0) n=23	86.3 (55.4) n=6	0.18		

 $^{^{\}mbox{\sc h}}$ One child did not have a stress-view performed

Table 31: Retroposition range of motion compared with Bony Union: Affected Thumbs

	Retroposition ROM as mean % of age-matched normal (SD)				
	Bony Union	Non-Union	p value		
Isolated thumb hypoplasia n=24	73.3 (47.2) n=21	52.5 (38.8) n=3	0.47		
Thumb hypoplasia with RLD n=4	28.9 (16.7) n=3	54.7 n=1	0.31		
Thumb hypoplasia with other associated conditions* n=2	25.7 (16.2) 11=2				
Total n=30	64.6 (46.4) n=26	53.1 (31.7) n=4	0.63		

^{*}Other conditions include ulnar dimelia, five-fingered hand

^{*}Other conditions include ulnar dimelia, five-fingered hand

The relationship of grip strength with CMC stability and metacarpal head/base bony union is shown in Tables 32-33.

Table 32: Grip Strength compared with CMC joint stability

	Grip Strength as mean % of age-matched normal (SD)				
	Stable CMC joint	Unstable CMC joint	p value		
Isolated thumb hypoplasia (n=27)	56.0 (19.8) n=21	38.3 (14.2) n=6	0.05		
Thumb hypoplasia with RLD (n=10)	9.0 (5.4) n=7	6.7 (0.6) n=3	0.48		
Thumb hypoplasia with other associated conditions* (n=2)	54.0 (45.6) n=2				
Total (n=39^)	44.9 (27.5) n=30	27.7 (19.4) n=9	0.09		

^{*}Other conditions include ulnar dimelia, five-fingered hand

Table 33: Grip Strength compared with bony union

	Grip Strength as mean % of age-matched normal (SD)				
	Bony Union	Non-Union	p value		
Isolated thumb hypoplasia (n=28)	53.7 (19.3) n=25	30.6 (17.2) n=3	0.06		
Thumb hypoplasia with RLD (n=10)	9.3 (4.9) n=7	6.0 (3.5) n=3	0.32		
Thumb hypoplasia with other associated conditions* (n=2)	54.0 (45.6) n=2				
Total (n=40)	44.6 (25.9) n=34	18.3 (17.5) n=6	0.02		

^{*}Other conditions include ulnar dimelia, five-fingered hand

 $^{^{\}wedge}$ One child did not have a stress-view performed

The relationship of palmar abduction strength with CMC joint stability and metacarpal head/base bony union is shown in Tables 34-35.

Table 34: Palmar abduction strength measured with the Rotterdam Intrinsic Hand Myometer, compared with CMC joint stability.

	Palmar Abduction strength (RIHM) as mean % of age-matched normal (SD)				
	Stable CMC joint	Unstable CMC joint	p value		
Isolated thumb hypoplasia (n=25)	21.8 (16.7) n=19	15.0 (12.6) n=6	0.37		
Thumb hypoplasia with RLD (n=11)	5.9 (3.4) n=9	4.2 (0.2) n=2	0.52		
Thumb hypoplasia with other associated conditions* (n=3)	26.2 (13.4) n=3				
Total (n=39)	17.6 (15.5) n=31	12.3 (11.7) n=8	0.37		

 $^{{}^*\}mathrm{Other}$ conditions include ulnar dimelia, five-fingered hand, triphalangeal thumb

 $Table\ 35: Palmar\ abduction\ strength\ measured\ with\ the\ Rotterdam\ Intrinsic\ Hand\ Myometer,\ compared\ with\ bony\ union$

	Palmar Abduction strength (RIHM) as mean % of age-matched normal (SD)			
	Bony Union	Non-Union	p value	
Isolated thumb hypoplasia (n=26)	20.6 (14.3) n=23	26.4 (32.5) n=3	0.58	
Thumb hypoplasia with RLD (n=11)	5.4 (3.4) n=7	5.9 (3.0) n=4	0.83	
Thumb hypoplasia with other associated conditions* (n=3)	26.2 (13.4) n=3			
Total (n=40)	17.9 (14.2) n=33	14.7 (21.8) n=7	0.63	

 $[\]hbox{*Other conditions include ulnar dimelia, five-fingered hand, triphalangeal thumb}$

The relationship of opposition strength with CMC joint stability and metacarpal head/base bony union is shown in Tables 36-37.

Table 36: Opposition strength measured with the Rotterdam Intrinsic Hand Myometer, compared with CMC joint stability.

	Opposition strength (RIHM) as mean % of age-matched normal (SD)			
	Stable CMC joint	Unstable CMC joint	p value	
Isolated thumb hypoplasia (n=25)	15.7 (7.4) n=19	10.7 (8.6) n=6	0.18	
Thumb hypoplasia with RLD (n=12)	4.8 (1.7) n=9	3.8 (0.4) n=3	0.31	
Thumb hypoplasia with other associated conditions* (n=3)	12.0 (2.7) n=3			
Total (n=40)	12.2 (7.6) n=31	8.4 (7.6) n=9	0.20	

 $^{{}^*\}mathrm{Other}$ conditions include ulnar dimelia, five-fingered hand, triphalangeal thumb

Table 37: Opposition strength measured with the Rotterdam Intrinsic Hand Myometer, compared with bony union

	Opposition strength (RIHM) as mean % of age-matched normal (SD)			
	Bony Union	Non-Union	p value	
Isolated thumb hypoplasia (n=26)	14.7 (7.8) n=23	17.1 (11.9) n=3	0.64	
Thumb hypoplasia with RLD (n=12)	4.9 (1.6) n=8	3.9 (1.1) n=4	0.28	
Thumb hypoplasia with other associated conditions* (n=3)	12.0 (2.7) n=3			
Total (n=41)	12.1 (7.6) n=34	9.5 (9.9) n=7	0.44	

 $^{{}^*}$ Other conditions include ulnar dimelia, five-fingered hand, triphalangeal thumb

The relationship of tripod pinch strength with CMC joint stability and metacarpal head/base bony union is shown in Tables 38-39.

Table 38: Tripod pinch strength measured with the pinch dynamometer, compared with CMC joint stability.

	Tripod Pinch Strength as mean % of age-matched normal (SD)				
	Stable CMC joint	Unstable CMC joint	p value		
Isolated thumb hypoplasia (n=14)	32.7 (16.2) n=10	19.8 (8.9) n=4^	0.16		
Thumb hypoplasia with RLD (n=1)	2.0 n=1				
Thumb hypoplasia with other associated conditions* (n=1)	6.0 n=1				
Total (n=16)	27.9 (18.4) n=12	19.8 (8.9) n=4	0.42		

^{*}Other conditions = five-fingered hand

Table 39: Tripod pinch strength measured with the pinch dynamometer, compared with bony union

	Tripod Pinch Strength as mean % of age-matched normal (SD)				
	Bony Union	Non-Union	p value		
Isolated thumb hypoplasia (n=15)	28.9 (16.5) n=13	19.3 (4.6) n=2	0.44		
Thumb hypoplasia with RLD (n=1)	2.0 n=1				
Thumb hypoplasia with other associated conditions* (n=1)	6.0 n=1				
Total (n=17)	25.6 (17.6) n=15	19.3 (4.6) n=2	0.63		

^{*}Other conditions = five-fingered hand

[^] One child did not have a stress-view performed

The relationship of key pinch strength with CMC joint stability and metacarpal head/base bony union is shown in Tables 40-41.

Table 40: Key pinch strength measured with the pinch dynamometer, compared with CMC joint stability.

	Key pinch Strength as mean % of age-matched normal (SD)				
	Stable CMC joint	Unstable CMC joint	p value		
Isolated thumb hypoplasia (n=14)	28.2 (19.6) n=11	26.8 (12.7) n=3	0.91		
Thumb hypoplasia with RLD (n=1)	0.8 n=1				
Thumb hypoplasia with other associated conditions* (n=1)	14.2 n=1				
Total (n=16)	25.0 (19.7) n=13	26.8 (12.7) n=3	0.88		

^{*}Other conditions = five-fingered hand

Table 41: Key pinch strength measured with the pinch dynamometer, compared with bony union.

	Key pinch Strength as mean % of age-matched normal (SD)				
	Bony Union	Non-Union	p value		
Isolated thumb hypoplasia (n=14)	29.1 (18.1) n=13	12.8 n=1	0.36		
Thumb hypoplasia with RLD (n=1)	0.8 n=1				
Thumb hypoplasia with other associated conditions* (n=1)	I				
Total (n=16)	28.0 (17.8) n=15	12.8 n=1	0.20		

^{*}Other conditions = five-fingered hand

The relationship of tip pinch strength with CMC joint stability and metacarpal head/base bony union is shown in Tables 42-43.

Table 42: Tip pinch strength measured with the pinch dynamometer, compared with CMC joint stability.

	Tip Pinch Strength as mean % of age-matched normal (SD)				
	Stable CMC joint	Unstable CMC joint	p value		
Isolated thumb hypoplasia (n=16)	24.9 (14.0) n=13	11.3 (7.6) n=3	0.13		
Thumb hypoplasia with RLD (n=1)	1.7 n=1				
Thumb hypoplasia with other associated conditions* (n=1)	20.9 n=1				
Total (n=18)	23.1 (14.2) n=15	11.3 (7.6) n=3	0.19		

^{*}Other conditions = five-fingered hand

 $Table\ 43: Tip\ pinch\ strength\ measured\ with\ the\ pinch\ dynamometer,\ compared\ with\ bony\ union.$

	Tip Pinch Strength as mean % of age-matched normal (SD)				
	Bony Union	Non-Union	p value		
Isolated thumb hypoplasia (n=17)	21.2 (12.8) n=15	29.2 (22.9) n=2	0.48		
Thumb hypoplasia with RLD (n=1)	1.7 n=1				
Thumb hypoplasia with other associated conditions* (n=1)	20.9 n=1				
Total (n=19)	20.0 (12.8) n=17	29.2 (22.9) n=2	0.88		

^{*}Other conditions = five-fingered hand

The relationship of hand function with CMC joint stability and metacarpal head/base bony union is shown in Tables 44-45.

Table 44: Jebsen hand function test (JHFT), total time to complete tasks, compared with CMC stability. Values are percentages of age-matched normal values.

	Jebsen Test of Hand function (Total time as a percentage of age matched normal)			
	Stable CMC joint	Unstable CMC joint	p value	
Isolated thumb hypoplasia (n=22)	87.8 (23.5) n=16	131.6 (63.5) n=6^	0.03	
Thumb hypoplasia with RLD (n=12)	228.5 (114.4) n=9	231.2 (146.8) n=3	0.97	
Thumb hypoplasia with other associated conditions* (n=1)	98.5 n=1			
Total (n=35)	136.9 (94.7) n=26	164.8 (101.9) n=9	0.46	

[^] One child did not have a stress-view performed

 $Table\ 45: Jebsen\ hand\ function\ test\ (JHFT),\ total\ time\ to\ complete\ tasks,\ compared\ with\ Bony\ Union.\ Values\ are\ percentages\ of\ age-matched\ normal\ values.$

	Jebsen Test of Hand function (Total time as a percentage of age matched normal)				
	Bony Union	Non-Union	p value		
Isolated thumb hypoplasia (n=23)	100.5 (43.7) n=21	97.8 (1.8) n=2	0.93		
Thumb hypoplasia with RLD (n=12)	217.7 (109.0) n=8	252.1(136.6) n=4	0.64		
Thumb hypoplasia with other associated conditions* (n=1)	98.5 n=1				
Total (n=36)	131.7 (83.5) n=30	200.7 (132.5) n=6	0.10		

^{*} Other conditions = five-fingered hand

^{*} Other conditions = five-fingered hand

The analysis of relative risk of instability of the CMC joint with union of the metacarpal head/base following pollicisation is shown in Tables 46-47.

Table 46: Cross tabulation between instability at the CMC joint and non-union of trapezium following pollicisation.

		Distribution with instability at CMC joint according to union or non-union of metacarpal head/base		Total	
			Stable	Unstable	
	Non Union	Number	6	1	7
Non-Unior Distribution	Non-Union	% Union	85.7%	14.3%	100.0%
with union	Union	Number	28	8	36
	Union	% Union	79.5%	20.5%	100.0%
Total		Number	34	9	43
		% Union	80.4%	19.6%	100.0%

Table 47: Risk estimate analysis of instability at the CMC joint and non-union of trapezium following pollicisation.

	Value	95% confidence interval	
	value	Lower	Upper
Relative risk of CMC joint instability after union	1.4	0.21	9.7

The analysis of relative risk of instability of the trapezium (metacarpal head/base complex) with non-union of the metacarpal head/base following pollicisation is shown in Tables 48-49.

 $Table\ 48: Cross\ tabulation\ between\ instability\ of\ trapezium\ and\ non-union\ of\ trapezium\ following\ pollicisation.$

		Distribution of trapezium instability according to union or non-union		Total	
			Stable	Unstable	
	Non Union	Number	4	3	7
Distribution	1	% Union	57.1%	42.9%	100.0%
with union		Number	37	0	39
	Union	% Union	100.0%	0.0%	100.0%
Total		Number	41	3	44
Total		% Union	93.5%	6.5%	100.0%

Table 47: Risk estimate analysis of instability of the trapezium following non-union of the metacarpal head/base.

			nfidence interval	
	Value	Lower	Upper	
Relative risk of instability of the trapezium after non-union	35.0	1.9	613.8	

Discussion

Assessment

i) Appearance

As expected an aesthetic difference was elucidated between the thumbs of children with isolated thumb hypoplasia and those with associated radial longitudinal deficiency. Digit size received higher scores than contour in both groups. Previous investigation of appearance of the pollicised thumb have not included thenar contour[32, 34], and although this subjective examination was not a focal point of this study, any future study should include an objective measure of thenar bulk.

ii) Position

Our results reflected those of Goldfarb [34] in demonstrating that pollicised digits remain longer than normal thumbs. Twenty-two pollicisations reached distal to the PIP joint line, with another thirteen equal to it. Only six of forty-one measured thumbs were proximal to the PIP joint.

The success of a pollicisation relies on, among other factors, the ability of the new thumb to oppose all digits. There are several factors which make this a challenge for the surgeon. Firstly, the joint structure of the basal thumb joint does not approximate that of a normal thumb CMC joint. Secondly, reconstruction of intrinsic musculature does not restore the strength of normal opposition. Thirdly, the IP joints of the transferred index finger are limited in active motion, as demonstrated in our cohort and in the literature [24, 27, 38]. Surgeons may compensate for these factors by increasing length of the pollicised digit. This was reflected in our results with thirty-five of forty-one thumbs equal to or longer than the PIP joint crease of the next ulnar digit. Surgeons can also shorten the tendon of the flexor digitorum profundus which may provide improved IP joint motion and flexion strength[47], however this was not performed in these cases.

iii) Tenderness

Tenderness was not elucidated over the CMC joint or the anatomical snuff box in any patient. Pain was not an inhibiting factor in measurement of strength or range of motion.

iv) Stability

Six thumbs were determined to be unstable with clinical stress testing, with all six of these also proving to be unstable on X-ray. Three thumbs assessed as stable with manual testing were considered unstable when examined with X-ray. Radiologic stability will be discussed in section ix) radiology.

Staines et al [38] have published the hypothesis that the non-operated hand in a patient with a unilateral pollicisation is inferior in strength and function to published normative data, despite not having any clinical diagnosis. Our results concur with this, including clinical assessment of stability of the non-operated thumb, which showed twelve unstable CMC joints on that side.

This test for clinical stability has not been applied to a normal population and cannot be validated. Furthermore, it is accepted that it is difficult to apply this test for stability in a standard manner, however the same method and criteria were utilised in all patients.

v) Range of motion

As expected, range of motion at the wrist was markedly reduced in those with radial longitudinal deficiency. Our mean values for wrist range of motion for those with isolated thumb hypoplasia were within one standard deviation of a normal population [53] except for wrist extension, which at 47 degrees was reduced (normal SD 67.3 + /-11.2 degrees), and radial deviation, which at 34 was increased (normal SD 20.5 + /-6.0 degrees).

The ability to make a fist was achieved in all children with no associated radial longitudinal deficiency. Those with RLD were limited by lack of wrist extension and limited digit range of motion.

The results recorded for IP joint and MCP joint flexion were not directly comparable with previous measures of total active motion (TAM), as our measures did not include flexion at the CMC joint. However, our results showed a difference between those with isolated thumb hypoplasia (91 degrees) and associated RLD (43 degrees), consistent with the difference shown between these groups in TAM.

Essentially TAM of the new thumb is a reflection of the pre-operative range of movement at the PIP and DIP joints of the index finger. Many of our cohort, in particular those with radial longitudinal deficiency, had reduced pre-operative finger ROM, for reasons including tendon deficiencies of the extensor indicis proprius and extensor digitorum muscles, partial syndactyly of the pollicised digit and joint stiffness of the IP joints of the index finger. A reported deficiency in TAM in a pollicised digit is not necessarily a reflection of the operation itself, but can be attributed to these or other pre-operative limitations. This infers that comparison of TAM in a series of pollicisation cases does not necessarily provide any useful information about differences in the result of the surgery, only in difference of pre-operative function.

Measurement of palmar abduction with the pollexograph provided a similar mean value to that previously reported in children with thumb hypoplasia (table 50), however it must be recognised that only a small number of children have been assessed using the same technique. Our measurement of 50.2 degrees was

marginally below the normal range of 57.7 (55.0-60.4) degrees[49], which may reflect a decreased range of motion at the basal thumb joint and/or alteration in the mechanics of the reconstructed abductor pollicus brevis.

Table 50: Palmar abduction: Comparison to the literature

	n	Palmar abduction		
Cohort		No associated abnormalities	Associated abnormalities (eg RLD)	Total
De Kraker*^	21	47.5		47.5
Roper	9			53
Our cohort (thumb tip)*	44	55.1	53.0	50.2
Our cohort (metacarpal)*	44	39.4	43.7	38.5

^{*} Measured with pollexograph

Components of CMC joint range of motion; palmar abduction, radial abduction and retroposition, were correlated to CMC joint stability measured by stress view radiographs. This has not been reported in any published study. In children with isolated thumb hypoplasia, palmar abduction measured on the pollexograph was slightly higher for those with a stable CMC (81.7% vs 76.9% of normal values, p =.62). The same was observed for those with associated RLD (89.7% vs 85.1% of normal values, p=.79). We were unable to demonstrate statistical significance. Radial abduction and retroposition were both observed to be lower in those with stable CMC joints and isolated thumb hypoplasia (77.4% vs 81.9%, p=.664 and 69.0% vs 86.3%, p=.43). The same trend was observed in those with associated RLD and this difference was also not statistically significant.

All measures of CMC range of motion were higher in those with bony union at the created first metacarpal head-base junction. In those with isolated thumb hypoplasia and bony union; palmar abduction was 81.7% vs 76.9% of normal values, p=.62, radial abduction 80.0% vs 61.1%, p=.16, and retroposition 73.3% vs 52.5%, p=.48. This data suggests that those with a union at the created trapezium maximise range of motion at the created CMC joint, and those without may rely on movement at other thumb joints for function. The suggestion of a pseudarthrosis at this articulation [24, 45] in cases of non-union does not appear to facilitate increased movement based on our data, although the numbers in our cohort did not allow significant differences to be established.

vi) Strength

This study compared the pollicised digit with both the opposite hand and with established age matched normative data. As expected, and consistent with studies to date, our grip strength results showed a markedly higher average

[^]Patients had not undergone pollicisation, but did have Blauth II-IV hypoplasia. Measured to the thumb tip.

score for those without associated radial longitudinal deficiency (RLD), 51% compared with 8% for those with associated RLD, when compared with agematched normals. The mean grip strength of 51(31-71)% was higher in our patient group compared with that reported by Manske[24]; 31(8-52%) and Netscher[31]; 29%.

Mankse reported tip, tripod and key pinch strengths in those with isolated thumb hypoplasia as 44, 35 and 38% of normal values, which were higher than our values of 22, 28 and 28%. Younger children had difficulty handling the pinch dynamometer, and many patients struggled to hold the device independently and still produce enough force to register a score on the scale. Close to half of the cohort were rated one out of four for ability, or 'grips but can't perform task', and excluded from analysis. This reduced the number of results when compared with grip strength analysis, for which forty out of a possible forty-four hands were able to complete as 'normal' or 'minimally abnormal'. For each pinch strength measure, some children were able to produce the lowest value on the scale with their first attempt, but then not able to move the needle past zero with their subsequent two attempts. These two results of zero were included in the average score, and this had noteworthy impact on our results.

Intrinsic strength, measured with the Rotterdam Intrinsic Hand Myometer (RIHM), was of similar magnitude to pinch values. In those with isolated thumb hypoplasia, palmar abduction was 21.2%, and opposition 14.9% of age-matched normal values. There were not any published studies that have used this device to measure strength in pollicised thumbs available for comparison. It is reasonable that intrinsic strength will be significantly more compromised than extrinsic strength, with both the effect of transferred musculo-tendinous units and the reduced excursion of the IP joints playing a role. It is also suggested that instability of the basal joint of the new thumb will negatively effect the strength of these intrinsics, with a subluxing CMC joint providing a far less reliable fulcrum for movement.

It has been established [31, 38, 40] that the contralateral, non-pollicised hand is weaker than normal non-dominant hands by between 50-87% for grip strength, 58-60% for key pinch and 62-70% for tripod pinch. Tip pinch was not reported. Our results support this, showing grip strength of the non-operated hand to be 80.2% of normal values, with key and tripod pinch strengths 61.1% and 58.9% respectively. Tip pinch was 52.1% of normal. For this analysis, children with any previous surgery of the contralateral hand were excluded. Children with unilateral RLD or other disorders such as ulnar dimelia did not return significantly lower results for their non-pollicised hand when compared with children with isolated thumb hypoplasia.

The relationship between instability and muscle weakness was observed in our study for both intrinsic and extrinsic muscle testing, best demonstrated in those with isolated thumb hypoplasia. Grip strength was significantly higher for those with stable CMC joints, 56.0% of normal (n=21), compared with unstable CMC joints, 38.3% (n=6), p=.05. Tripod, key and tip pinch strength were all higher, without statistical significance, in those with stable CMC joints; 32.6% vs 19.8% (p=.16), 28.2% vs 26.8% (p=.91) and 24.1% vs 11.3% (p=.13) respectively. Intrinsic muscle groups were also higher, although this difference was again non-

significant, with palmar abduction strength 21.8% vs 15.0% (p=.36) and opposition 15.7% vs 10.7%, stable to unstable. The smaller patient numbers as well as low measurements make it difficult to draw conclusion for those with radial longitudinal deficiency. Children with pollicised digits for other pathology such as ulnar dimelia, five-fingered hand and tri-phalangeal thumb were all observed to have stable CMC joints.

Bony union at the head-base junction of the metacarpal may also influence postoperative strength of the pollicised digit. In those with isolated thumb hypoplasia and bony union, grip strength averaged 53.7% of aged matched normal values, compared with 30.6% in those with non-union, p=.06. A difference was also observed in those with associated RLD, 9.3% vs 6.0% (p=. 32), and a significant difference in the total cohort (44.6% vs 18.3%, p=.02). Those with bony union were also found to have higher pinch and intrinsic strength measures, however we were unable to demonstrate clinical significance.

vii) Function

All children were asked to complete the functional tasks, regardless of age. Most children were able to complete some of the five prescribed activities, with only one child aged two with a hypoplastic tri-phalangeal thumb, as well as a six year old with ulnar dimelia unable to complete any of the tasks. Of the Jebsen Hand Function Test (JHFT) tasks, turning the cards and lifting heavy objects proved the most difficult, with five children unable to complete, one of whom could not complete with either pollicised digit. Some children unable to use their pollicised digit to produce an adequate tip pinch to grasp a card on a flat surface were able to complete the card turning task using a 'scissor' grasp between two digits, usually the fourth and fifth fingers. Age matched normal values were not available for children under six years of age.

If a child was able to lift the heavy object, there was not a large amount of difference in the time taken to move five full cans compared with five empty cans. Full cans were assessed subsequent to empty cans so may have been subject to some repetition bias in testing. However this similarity is also observed in the normative data, with all age-groups having a less than 0.5 second difference between the two weights. The 'heavy' objects could perhaps be heavier to give a true reflection of both strength and ability to form a grasp.

Children with isolated thumb hypoplasia performed better on the JHFT than those with associated radial longitudinal deficiency (100.3% vs 229% of normal values). This is consistent with findings by Buffart et al [41] and is reflective of the combination of superior strength and range of motion seen in this group (Figures 7-8). It is also of note that function of the pollicised digit in those with isolated thumb hypoplasia approached the normal population when measured with the JHFT (100.3%) when their grip strength (51.2%) and range of motion (70.7-80.1%) did not. This is reflective of the development of 'trick' movements and adaptive strategies employed by those with pollicised digits, and is suggestive that the digit may be of near normal function, despite inferior strength and range of motion.

In children with isolated thumb hypoplasia, total time to complete the JHFT was significantly lower for those determined to have stable CMC joints than those with unstable CMC joints (87.8% vs 131.6% of normal values, p<0.05). There was a smaller difference in those with radial longitudinal deficiency which was not significant. This preliminary data highlights the importance of stability of the created CMC joint in ultimate result of the pollicisation procedure.

Bony union did not have significant impact on the function of the pollicised digit. This analysis was limited by the small number of thumbs identified to have non-union between the head and base of the first metacarpal.

viii) Satisfaction

Satisfaction was recorded as a subjective measure of cosmesis and function by both the examiners and parents. Some older patients were able to complete this themselves. Patients and parents were more likely than the examiners to rate their function as excellent or good (80% vs 68%). A similar picture was seen for cosmesis with 86% of patients and parents rating the look of the operated hand as excellent or good compared with the examiners 71%.

Our method of assessment was much more basic than that of Goldfarb et al [34] but showed a similar trend of the patient and caregiver rating the cosmesis of the hand higher than the surgeon.

ix) Radiology

Thirty-seven of forty-four (84%) thumbs demonstrated bony union between the base and head of the first metacarpal. One child had received corrective surgery for a non-union following her original pollicisation, so for the purpose of discussion this child must be included as a failure to unite, making the actual non-union rate eight of forty-four (18%). None of the thumbs with a united trapezium were unstable at the head/base junction. Of the remaining seven without bony union, three demonstrated some instability (41%) at the head/base junction, suggesting a firm fibrous union is not always achieved. The relative risk of instability of the head/base complex after non-union was 35.0.

Thirty-four of forty-three (79%) hands were deemed stable at the CMC joint. Of those considered unstable, only one had trapezial non-union. This is suggestive that there may be increased stress placed across the CMC joint when union occurs at the base of the metacarpal. The relative risk of CMC joint instability after trapezial union was 1.4.

As for clinical instability, there were limitations in the assessment of radiological instability. The application of stress across a joint was difficult to standardise, with clinical judgement of angular and translational endpoint used as the determinant of position for x-ray. Furthermore, the assessment of stability was only assessed in the radial-ulnar plane. The CMC joint could be unstable in a dorsal-volar plane and this has not been assessed.

There is currently no definition or accepted measurement technique of CMC stability in patients post-pollicisation. The anatomy of the basal thumb joint is considerably different given the transfer of a MCP joint to assume the role of a

CMC joint. This includes differences in the articular surfaces, ligamentous structures and muscle excursions. For these reasons application of existing measures of CMC stability [42] were not valid for our population. However, we consider the parameters we developed (radial and ulnar angulation and displacement) and the definitions of instability (greater than fifty degrees angulation and twenty five percent displacement) to be logical and reasonable and to provide a superior indication of stability/instability than the few methods previously documented [36, 42].

Conclusion

In this case series of 44 hands that have undergone pollicisation, the union rate between MC head and base was 82%. For those with non-union, there is a relative risk of 35.0 for trapezial instability. Those patients with bony union demonstrated a significant increase in grip strength. Although there was a trend towards increased ROM, greater strength measurements for pinch, and improved functional results in those patients with bony union, these differences did not reach statistical significance in this number of patients.

There was a relatively increased risk of instability of 1.4 at the CMC joint following union of the MC head and base. Strength and motion measurements tended to be greater in those patients with stable CMC joints although only the measurement of grip strength reached statistical significance in those with isolated thumb hypoplasia. There were statistically significant better functional results in those patients with isolated thumb hypoplasia and stable CMC joints.

This study suggests that the aim of obtaining union of the new trapezium to the MC base is of benefit. The increased risk of CMC joint instability does not appear substantial enough to negate this conclusion. Regrettably, the number of patients in the study group does not allow stronger conclusions to be claimed. However, this rigorous study did confirm, with statistical significance, those claims previously documented in the literature of superior results in patients with thumb hypoplasia against those with RLD and the tendency for weaker strength in the supposedly normal hands of patients undergoing unilateral pollicisation of an affected hand.

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was truly appreciated and hopefully this project can go some way toward improving the outcomes of pollicisation in the future.

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Appendix 1: Ethics Approval - Site Specific





02 May 2012

Prof Michael Tonkin Dept of Hand Surgery and Peripheral Nerve Surgery Building 36, Level 4 RNSH. St Leonards. NSW 2065

Dear Prof Tonkin.

Re: SITE SPECIFIC ASSESSMENT (SSA) APPROVAL

PROTOCOL- 1205-121M, (SSA-LNR) AURED NEAF REF: LNR/12/HAWKE/66 AURED SSA REF: LNRSSA/12/HAWKE/123 STUDY INVESTIGATORS: Prof Michael Tonkin,

STUDY TITLE: Assessment of stability of the reconstructed thumb CMC joint following

pollicisation and its relation to thumb function

I am pleased to inform you that on the 2 May 2012, the delegate of the Chief Executive authorised the Site Specific Assessment for the above study on behalf of Northern Sydney Local Health District (NSLHD).

It is noted that the approval covers the following NSW Health site:

Royal North Shore Hospital (Department of Hand Surgery & Peripheral Nerve Surgery

The documentation included in the approval is as follows:

- Site Specific Assessment (SSA) Form for Low and Negligible Risk Research NSW, NSW LNR SSA Version 2.0 (2011)
- Application Form for Ethical and Scientific Review of Low and Negligible Risk Research, NSW LNR Version 2.0 (2011)
- Patient Information Sheet & Consent Form, Master, Version 1.1 dated 28 March 2012
- Site Specific, RNSH, Patient Information Sheet & Consent Form, Version 1.0 dated 29 March 2012
- Invitation Letter, Version 1.1 dated 28 March 2012.
- Site Specific Invitation Letter, Version 1.0 dated 29 March 2012.

It is noted that the Ethical & Scientific Approval for this project was reviewed and approved by Northern Sydney Local Health District (NSLHD) Human Research Ethics Committee who is accredited under the NSW Health model for single ethical review of multi-centre research.

At this time, we also remind you that, in order to comply with the *Guidelines for Good Clinical Research Practice (GCRP) in Australia*, and in line with NSLHD HREC policy, the Chief Investigator is responsible to ensure that:

 The HREC is notified of anything that might warrant review of the ethical approval of the project, including unforeseen events that might affect the ethical acceptability of the project.

> Research Office Level 13, Kolling Building Royal North Shore Hospital St Leonards NSW 2065 Tel (02) 9926 4590 Fax (02) 9926 6179





- The HREC is notified of all Serious Adverse Events (SAEs) or Serious Unexpected Suspected Adverse Reactions (SUSARs) in accordance with the Serious Adverse Event Reporting Guidelines. Please refer to the Research Office website.
- 3. Proposed amendments to the research protocol or conduct of the research that may affect the ethical acceptability of the project are submitted to the HREC on an amendment form (including any relevant attachments). For multi-centre studies, the Chief Investigator should submit to the Lead HREC and then send the amendment approval letter to the investigators at each of the sites so that they can notify their Research Governance Officer.
- Proposed changes to the personnel involved in the study are submitted to the HREC on a Change in Personnel Form (accompanied by the investigator's CV where applicable).
- Change in Personnel Form (accompanied by the investigator's CV where applicable).

 5. The HREC must be provided with an annual progress report for the study by the 31st October each year. For multi-centre studies the Chief Investigator should submit to the Lead HREC on behalf of all sites. The annual report acknowledgment from the Lead HREC should be submitted to the Research Governance Officer.
- The HREC must be provided with a final report upon completion of the study. For multi-centre studies the Chief Investigator should notify the Lead HREC and the investigators at each site should notify the relevant Research Governance Officer.
- The HREC must be notified, giving reasons if the project is discontinued at a site before the expected date of completion.

Internet:

www.northernsydneyresearch.com.au

Approval lasts for five (5) years, therefore your approval will expire on 02/05/2017. Should you require an extension an amendment form should be submitted.

Yours sincerely,

Susan Siwakoti Ethics Officer Research Office

NORTHERN SYDNEY LOCAL HEALTH DISTRICT

Research Office Level 13, Kolling Building Royal North Shore Hospital St Leonards NSW 2065 Tel (02) 9926 4590 Fax (02) 9926 6179







The Sydney Children's Hospitals Network

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GADATA\Research\GOVERNANCE-SSA Applications\Approval Letters & Memos\LNR-12-SCHN-242 SSA Ltr to Michael Tonkin.docx

16 July 2012

Dr Michael Tonkin Department of Hand Surgery and Peripheral Nerve Surgery Royal North Shore Hospital Building 36, Level 4 ST LEONARDS NSW 2065

Site Authorisation Letter

Dear Dr Tonkin,

HREC reference number: SSA reference number:

HREC/12/HAWKE/66 LNRSSA/12/SCHN/242

Project title:

Assessment of Stability of the Reconstructed Thumb CMC Joint Following Pollicisation and its Relation to Thumb Function

NSW Sites listed:

The Sydney Children's Hospital's Network - Westmead

Site-specific approved documentation:	Version	Date
Parent Information Sheet	1.0	2 Apr 2012
Participant Information Sheet	1.0	2 Apr 2012
Assent Form	1.0	2 Apr 2012
Consent Form	1.0	2 Apr 2012
Letter of Invitation to Parents/Participants	1.0	29 Mar 2012

Thank you for submitting your application for this project. I am pleased to inform you that authorisation has been granted for this study to take place at the above site.

The following conditions apply to this research project. These are additional to those conditions imposed by the Human Research Ethics Committee that granted ethical approval:

- Proposed amendments to the research protocol or conduct of the research which may affect the ethical acceptability of the project, and which are submitted to the lead HREC for review, are copied to the research governance officer;
- Proposed amendments to the research protocol or conduct of the research which may affect the ongoing site acceptability of the project are to be submitted to the research governance officer.

Yours sincerely,

Mr James Cokayne Research Governance Manager



13 June 2012

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Prof Michael Tonkin Sydney Hand Surgery Associates Suite 1, Level 4 North Shore Private Hospital Westbourne St, St Leonards, NSW, 2065

Assessment of Stability of the Reconstructed Thumb CMC joint following pollicisation and its relation to thumb function.

Dear Prof Tonkin

Thank you for submitting this proposal to the North Shore Private Hospital Ethics Committee for consideration. This application was considered at the Ethics Committee Meeting on 7 June 2012.

The documentation reviewed for this approval included:

- 1. Letter from Prof. Tonkin to Dr Leslie dated 1 May 2012.
- Letter from the Co-Chairperson of the Northern Sydney Local Health District dated 28 March 2012, indicating approval as a low/negligible risk project to be performed at Royal North Shore Hospital and Children's Hospital Westmead.
- NSW Health Application Form for Ethical and Scientific Review of Low and Negligible Risk Research, including patient information sheets and consent forms, dated 15 February 2012.
- 4. Data collection forms.
- 5. Invitation letter Version 1.0, dated 1 May 2012.
- Sydney Hand Surgery Associates Parent Information Sheet and Parent/Participant Consent Forms, Version 1.0, dated 1 May 2012.

The Committee was supportive of this study and approved it to proceed as a Low/Negligible Risk Study, subject to minor changes, indicating North Shore Private Hospital Ethics Committee as the approving committee and providing appropriate contact details for this committee.

Ethics Committee approval is valid for 3 years from the date of the approval letter. It is a requirement of North Shore Private Hospital Ethics Committee that you submit annual reports of progress of the study as well as a final report of the outcome of the study at its completion. You first progress report will be due on 10 October 2012.

Yours sincerely

Garth I Leslie

Chair, North Shore Private Hospital Ethics Committee

Appendix 2: Ethics Approval - The University of Sydney



RESEARCH INTEGRITY

Human Research Ethics Committee

Web: http://sydney.edu.au/research_support/ethics/human/ Email: ro.humanethics@sydney.edu.au Address for all correspondence:

Level 6, Jane Foss Russell Building - G02 The University of Sydney NSW 2006 ÁUSTŔALIÁ

Ref: GD/JG

02/08/2012

Professor Michael Tonkin Department of Hand Surgery, Royal North Shore Hospital Email: mtonkin@surgery.usyd.edu.au

Dear Professor Michael Tonkin

Title: Assessment of stability of the teconsttucted thumb CMC joint following pollicisation and its relation to thumb function [protocol number 15139]

Masters Student: Nathan Trist

The Executive of the Human Research Ethics Committee (HREC), has reviewed your study to include the Masters student – Nathan Trist and acknowledges your right to proceed under the authority of Northern Sydney Local Health District (NSLHD) Human Research Ethics Committee.

The Human Research Ethics Committee advises that you consult with The University of Sydney Audit and Risk Management Office (http://sydney.edu.au/audit_risk/) to ensure that University of Sydney staff/students and premises are adequately covered for the purpose of conducting this research project.

Any modifications to the study must be approved by the Northern Sydney Local Health District (NSLHD) Human Research Ethics Committee. A copy of the approved modification, approved progress report and any new approved documents must be provided to The University of Sydney HREC for our records.

Please do not hesitate to contact Research Integrity (Human Ethics) should you require further information or clarification.

Yours sincerely

Professor Glen Davis

Chair

Human Research Ethics Committee

cc: Nathan Trist; email address; ntri7963@uni.sydney.edu.au

Appendix 3. Surgical Technique

Incisions: The first limb begins dorsally and distally at the index-middle web, and extends proximally and obliquely to the radial border of the hand proximal to the index finger MCP joint. The second limb extends from the proximal point of the first limb onto the palmar aspect of the proximal phalanx to meet the origin of the first limb in the index-middle web space. The third limb extends proximally from the palmar limb, in the line of the index-middle intermetacarpal space. These flaps are transposed when the index finger is rotated and recessed proximally. A number of subtleties of modification cater for specific demands. The palmar incision in the digit should be extended to just proximal to the proximal interphalangeal (PIP) joint when the index finger is well-developed and mobile. A longer thumb is preferable if there is significant index finger stiffness as greater length compensates for lack of mobility. In this instance the palmar incision is moved proximally towards the basal finger crease. A longitudinal incision extended distally from the dorsal limb incision to the PIP joint allows access to the extensor mechanism and its lateral bands for construction of thumb intrinsic mechanisms and the extrinsics, EPB and abductor pollicis longus (APL). The third palmar limb may be moved radially to incorporate excision of a Grade 3 or Grade 4 thumb. Alternatively, the excision of such may be incorporated into the second, more radial limb.

Palmar dissection: The neurovascular bundle of the index-middle web is identified. A radial neurovascular bundle is usually present. However, the radial digital artery to the index finger may be very small, perhaps even absent, in Grade 5 hypoplasia which is accompanied by index finger hypoplasia. The neurovascular bundles on either side of the digit are mobilised using microsurgical instruments and magnification. Inspection of the second common digital artery will determine the level of bifurcation into digital arteries to the adjacent sides of the index and middle fingers. The radial digital artery to the middle finger is tied off. The neurovascular pedicle is dissected proximally. A neural ring is relatively common but can usually be attended to by intraneural dissection of the common digital nerve. An awareness of the possibility of arterial compromise with proximal recession of the digit, either due to a neural ring or fascial structures, should prevent this complication.

Rarely, anomalies of the common digital artery demand an alteration in strategy. The vessel may arise from the deep palmar arch. In this instance the artery is short and may not allow proximal recession of the digit without compromising its arterial supply. It may be necessary to divide the deep arch, following preliminary clamping and assessment of any compromise in vascularity to the hand, to gain length. In one instance, I have found absence of a palmar common digital artery but with a large dorsal metacarpal artery connecting to the palmar system at the head-neck junction. Pollicisation was performed with the digit nourished by this vascular pedicle.

A1 and A2 pulleys are divided. The A3, A4 and A5 pulleys become the thumb A1, oblique and A2 pulleys respectively. Some routinely shorten the flexor digitorum profundus (FDP), but I have not found this necessary unless pollicisation is performed at greater than five years of age. A z-shortening can be performed

proximal to the wrist to avoid increasing the possibility of adhesions within the dissected area of the palm.

The dissection of the intrinsic muscles, the first dorsal and first palmar interossei, begins on the palmar side, mobilising the musculotendinous units to the MCP joint level, but protecting the neural supply of each.

Dorsal dissection: Thin dorsal flaps are elevated until the dorsal venous architecture is identified so that one or two veins, along with superficial dorsal nerves, can be mobilised separately from the flaps and the underlying digit. This prevents kinking of vessels, compromising venous return, when the digit is recessed proximally.

The extensor mechanism is inspected to assess the presence or absence of EIP and the quality of extensor digitorum communis (EDC). Excursion is often poor when radial deficiency accompanies thumb hypoplasia. Subsequent dissection of the extrinsic extensors and the intrinsic contributions to the extensor mechanism are performed before division of the extensors and with the skeleton intact. This allows distal mobilisation of the extensor mechanism to the level of the PIP joint, separating the lateral band contributions to this level, but maintaining continuity with the first dorsal interosseous and the first palmar interosseous muscles on radial and ulnar sides respectively. Release of the intrinsic attachments to either side of the base of the proximal phalanx must respect the integrity of the capsule and ligaments of what will become the new CMC joint. Although some recommend ablation of the blood supply to the physis of the metacarpal, others prefer not to interfere with any contribution which may maintain the integrity of the physis of the proximal phalanx. Premature physeal closure and a short first metacarpal in the reconstructed thumb is a consequence of growth plate compromise of the index finger proximal phalanx.

At this point, the EIP and EDC may be divided at the level of the MCP joint. Any remaining attachments of the intrinsic musculature are then dissected in a submuscular periosteal manner from the metacarpal diaphysis. Retractors are then placed around the head-neck junction of the metacarpal, protecting all other structures, particularly the palmar neurovascular bundles, whilst an osteotomy is performed at the head-neck junction of the metacarpal. In the young child, a Beaver blade or small osteotome is most satisfactory for the purpose. Some bone nibblers can be used to flower the metaphyseal perimeter of the head of the metacarpal by simply breaking bone fragments, which remain attached to the periosteum. This leaves bone with osteogenic potential to assist in bone union of the new trapezium to the metacarpal base (see below). The physis is removed using a fine curette and Beaver blade so that the new trapezium will not grow longitudinally. If ossification has occurred in the head of the metacarpal, it is easy to establish that the growth plate has been adequately removed. Care needs to be taken when ossification has not occurred, so that the articular surface of the metacarpal head is not breached.

CMC joint reconstruction: An integral part of the success of a pollicisation is the creation of a new CMC joint and there are a number of principles in reconstruction which would appear to be necessary:

- Optimal positioning of the new thumb ray in palmar abduction, radial abduction and appropriate rotation;
- Placement of the thumb ray in an anterior plane to that of the finger CMC joints;
- Hyperextension of the index finger MCP joint to prevent an hyperextension deformity of the new CMC joint.

It is difficult to satisfy all of the above parameters and obtain bony apposition between the index finger metacarpal head and base. Buck-Gramcko suggested retention of the metacarpal base to be necessary only in cases with relatively short phalanges. In these cases, the metacarpal head was fixed to the base using one or two K-wires. If the phalanges were of normal length, his initial description did not retain the metacarpal base and the metacarpal head was sutured to the joint capsule and carpal bones. Subsequently, most, including Buck-Gramcko, have preferred to retain the base. The plane of osteotomy incision of the metacarpal is varied, with both a transverse osteotomy at the metacarpal base and an oblique osteotomy in either coronal or sagittal planes being described. Some prefer K-wire fixation to promote head to base union as described by Buck-Gramcko. Some eschew this. Manske wrote of the importance of a fibrous union rather than a bony union between the retained base and head^[24,25], creating a pseudarthrosis at this articulation. He proposed that using sutures rather than K-wires for fixation permitted increased mobility of the new thumb.

A concern is one of possible instability of the new trapezium. However, the effect on functional outcomes according to the presence or absence of bone union between the metacarpal head (new trapezium) and the metacarpal base has not been determined. My preference is to aim for bone union whilst satisfying the above criteria of positioning.

An oblique osteotomy leaving the bone longer dorsoradially allows a satisfactory compromise in positioning the thumb optimally and maintaining some bone to bone contact. A fine K-wire is placed antegrade through the flexed metacarpal head and phalanges of the index finger and then driven retrograde into the carpus with the thumb in the desired position, removing the wire at five weeks. Before fixing the thumb to the carpus in this manner, two gauge 2-0 Ticron sutures are placed through the base of the metacarpal and into the metacarpal head, to be tightened following wire fixation of the thumb to the carpus. This method compromises the position of pronation, as 90° only is possible if one is to maintain an anterior lie of the new trapezium in relationship to the metacarpal base and some bone to bone apposition. 30° of radial abduction and 40° of palmar abduction is ideal. The less mobile digit may be fixed at lesser angles of radial and palmar abduction. Passive joint motion and the quality of the extrinsic and intrinsic motors play a role in this decision.

Tendon reconstruction: The EIP, if present, is shortened and re-sutured to the central extensor mechanism to the proximal interphalangeal joint of the index finger. Most refer to this as a construction of EPL function. However, the insertion of the central slip into the middle phalangeal base of the index finger mimics EPB anatomy of the thumb, rather than EPL anatomy. The new tendon

does simulate the adductor-retropulsion action of EPL. The tension of repair should be firm but less than full. Too tight a repair will result in retropulsion of the pollicised digit, particularly if a balance is not achieved following the reconstruction of APB. EDC helps stabilise the position of the new thumb metacarpal, moreso if its route and positioning are modified to better mimic the function of APL. It is attached to the periosteum at the dorso-radial aspect of the index proximal phalanx, avoiding the growth plate. If EIP is absent, EDC is used for EPB construction.

Although Buck-Gramcko advises dividing the lateral bands, shortening and resuturing them to create an APB and an adductor from the first dorsal interosseous and the first palmar interosseous respectively, I tend to concertina these tendons without dividing them and suture them together under as firm a tension as is possible. It is necessary to mobilise both lateral bands to beyond the PIP joint of the index finger, particularly that from the first dorsal interosseous so that its ability to abduct and rotate is optimal. This also decreases a tendency of the lateral bands to hyperextend the new MCP joint of the thumb. A gauge 5-0 Ticron suture is used to secure the tendon reconstructions.

When thumb hypoplasia is accompanied by radial hypoplasia, there is often a camptodactyly of the index finger. My preference is to deal with any significant flexion deformity of the new thumb MCP joint at a second procedure, for fear of interfering with the viability of the pollicised digit.

The tourniquet is released to check the vascularity of the thumb. Flaps are then refashioned so that they may be sutured into position with a pleasing contour. The skin tension within the flaps will assist the musculotendinous reconstruction in maintaining the position of the thumb once the wire is removed.