Supplementary Material

Methods of Outcome Assessment and Scoring

Dijkman et al (2016)

Rotterdam Outcome Assessment System for Radial Polydactyly (see Table 1 in Dijkman et al, for fully scoring):

- Active range of motion and deviation of the interphalangeal (IP) and metacarpophalangeal (MCP) joints were measured using handheld goniometer.
- Instability was assessed by applying lateral stress to the joints.
- Palmar abduction was measured using the Pollexograph.
- Visual analog scale (0–100):
  - Appearance of the scar
  - Residual prominence at amputation site
  - Thumb size
  - Pulp appearance
  - Nail appearance

- Patient reported pain: never, when cold, with use, and constant.
- Patient reported satisfaction: maximal, reasonable, moderate, and dissatisfied.

Strength:
- Tip, tripod, and key pinch strengths assessed using a pinch strength dynamometer.
- Each measurement was performed three times, and the mean of these three maximum voluntary contractions was calculated to ensure measurement accuracy.

Appearance:
- Visual analog scale (extremely ugly–perfectly normal looking)
- Appearance of the scar
- Residual prominence at the amputation site
- Thumb size
- Pulp
- Nail
- Overall aesthetic outcome

Thumb size:
- Length was measured from palpable base of the proximal phalanx to the tip.
- Circumference was measured at the IP joint with the joint in neutral position.
- Pulp circumference was measured at the level of the lunula.
- Nail width was measured at the lateral edges, where the lateral nail folds meet the hyponychium.


Cosmetic assessment: clinically assessed regarding nail width and cosmetic acceptability.

- Measuring the width of nail plate quantified nail growth.
- The terms “hypertrophic scar” and “atrophic pulp” were based on the perceptions of the patients or their guardians.

Additional measures:
- Grip strength was measured using a Jamar dynamometer.
- Tip and chick pinch were measures with a pinch gauge.
- Growth potential:
  - Retarded radial growth was defined as the thumb circumference of the distal and/or proximal phalanx being <75% of that on the normal side.
  - Longitudinal growth was represented by the longitudinal length of the thumb between the carpometacarpal joint and the tip of thumb in the resting position.

- Instability: X-rays of thumbs were taken in posteroanterior or view in full extension, stressed radially and ulnarily. The contralateral thumb was used as the reference.

- First web space contracture: defined as a loss of >25% of intermetacarpal angle between the first and second metacarpal bones, when compared with the normal side.

- Stiffness: defined as a loss of >25% of the sum of active IP joint (IPJ) and MCP joint range of movement with reference to the normal side. Ulnar deformity was defined as >5 degrees of ulnar angulation at the IPJ or >20 degrees of angulation at the MCP joint in the resting position. Radial instability was defined as >5 degrees of angulation at the IPJ or >20 degrees at the MCP joint when the examiner subjected the thumb to maximal stress from the resting position. In cases of bilateral polydactyly, the digit with the less affected parameter was taken as the normal side for comparison.

- Joint deformity: unclear how measured
- Bone growth: unclear how measured

Overall scoring:

The authors reference the scoring system by Tada et al; however, they give a maximum score or 20, which is not in line with the original Tada et al’s scoring. It is therefore unclear how the score is achieved.

Maillet et al (2007)

- Areas assessed (further details not given):
- Nail appearance
- Length and width of the thumb compared with the opposite thumb
- Alignment
- MCP and IP joints stability and arc of mobility
- Active abduction and opposition
• Strength of the grip between the thumb and second finger and the strength of the palmar grip.
• Function was assessed by assessing the patient’s manipulation of various objects.
• Determination of the alignment and the quality of bony fusion in the case of the Bilhaut technique (based on radiographs).
• Satisfaction of children’s parents was assessed using a simple scale: very satisfied, satisfied, not very satisfied, and not at all satisfied

Scoring (based on the Tada scoring system):
• Range of active motion of the IP and MCP joints:
  ° 70 degrees = 2; 50 to 70 degrees = 1; and <50 degrees* = 0
Stability:
  ° Absent = 1 and present = 0
Overall score:
  ° Good = 4 to 5; fair = 2 to 3; and poor = 1 or less

Larsen et al (2005)

Objective assessment:
Thumb pinch strength:
• Digital pinch meter
• Scoring: > 66% of control group = 2; 33 to 66% of control group = 1; and <33% of control group = 0

Range of motion:
• Ranges of motion at the IP and MCP joints were determined separately using a digital goniometer
• Scoring: > 66% of control group = 2; 33 to 66% of control group = 1; and <33% of control group = 0

Joint stability:
• Defined as more than 5 degrees lateral deviation at IP joint, or, at least, 20 degrees at the MCP level (tested manually).
• The score for the more unstable of these two joints was used (as per Ogino et al, 1996)
• Scoring: instability present = 0 and instability not present = 1

Alignment:
• Alignment of the IP and MCP joints was measured digitally by measuring any ulnar or radial deviation from the central axis with a goniometer
• Scoring: < 10 degrees = 2; 10 to 20 degrees = 1; and >20 degrees = 0
• Overall scoring:
  ° >7 = good
  ° 4 to 7 = fair
  ° < 4 = poor

Subjective assessment:
• Patients graded functional and cosmetic results on a visual analog scale (not further details)

Ogino et al (1996)

Subjective outcomes
Patient/parent subjective feelings on function and cosmetic result.

Objective outcomes
• Range of motion: recorded as the sum of the active motions of the IP and MCP joints.
  ° Scoring: 90 degrees or less = 0; 91 to 130 degrees = 1; and 131 degrees or more = 2.
• Joint stability: whichever joint was more unstable between IP and the MCP joints was evaluated and its score was used. Joints were tested manually for lateral instability. It was rated from −1 to 1.
  ° Scoring: IP joint: 5 degrees or more = −1 and 4 degrees less = 1. MP joint: 20 degrees or more = −1 and 19 degrees or less of instability = 1.
• Thumb alignment: rated from 0 to 2 points
  ° Scoring: 9 or less = 2; 10 to 19 = 1; and 20+ = 0.

Overall Scoring
• 4 or 5 = good overall result
• 2 or 3 = fair
• 1 or less = poor

Ganley and Lubahn (1995)

Functional results: satisfied or not satisfied
Cosmetic results: satisfied or not satisfied
Patient satisfaction surveys: conducted by telephone and personal interview.

Townsend et al (1994)

Functional
• IP joint deviation/ laxity: unacceptable if >15 degrees
• MCP joint deviation/laxity: unacceptable if >30 degrees
• Patient/examiner overall assessment: unacceptable if they felt results unacceptable for other reason

Cosmetic
• Girth: unacceptable if one-third more or less than normal thumb
• Length: unacceptable if <80% of normal thumb
• Patient/examiner overall assessment: unacceptable if they felt results unacceptable for other reason

Goffin et al (1990)

Areas assessed (further details not given):
• Hand function (writing)
• Lateral stability of IP and MCP joints
• Opening and depth of the first web
• Frontal and sagitta deviation
• Articular mobility of the MCP and IP
• Aesthetic
• Radiological appearances
• Parents' opinion of result

No scoring system was used.

**Andrew and Sykes (1987)**

Assessment via interviews (patients and parents) using a standard proforma of outcomes, pre- and postoperative. Questioning was focused around functional and cosmetic disability caused by the thumb pre- and postoperative. A particular note was made of whether the patient had been teased.

Cosmetic assessment in comparison to contralateral side (no further details given):

• Length
• Circumference
• Overall shape
• Angulation at rest were noted

Cosmetic assessment in comparison to contralateral side (no further details given):

• Range of movement of the joints of the thumb was measured, after noting the flexor crease pattern of the thumb.

**Tada et al (1983)**

Scoring, as originally described:

Range of active motion of the IP and MCP joints:

- 70 degrees = 2; 50 to 70 degrees = 1; and <50 degrees = 0
- Stability:
  - Absent = 1 and present = 0
- Malalignment:
  - <10 degrees = 2; 10 to 20 degrees = 1; and >20 degrees = 0
- Overall score:
  - Good = 4 to 5; fair = 2 to 3; and poor = 1 or less

• Collateral ligament stability of MCP and IP joints
• Pinch, chuck pinch, power grip, and span grasp
  No scoring system was used.