

A Modified Bilhaut–Cloquet Procedure for Zigzag Thumb Polydactyly Types III and IV

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Background: A review of the English literature over the least 43 years revealed only a total of 53 cases of Wassel types III, IV, and VII treated by the Bilhaut–Cloquet (B–C) procedure. Furthermore, the detailed results and range of motion were only given in 2 series (a total of 9 cases).

Methods: Four cases of Wassel types III and IV thumb duplications with zigzag deformity were treated with a modified B–C procedure. The main modification was minimal central resections at the joints aiming for joint preservation to improve the postoperative range of motion of the new thumb.

Results: The results showed an excellent overall functional score (Tada score of 5 of 5 in all cases). The mean range of motion at the metacarpophalangeal joint was excellent (60°), but the mean at the interphalangeal joint was only 20°. Cosmetically, all thumbs were “too wide” and a panel of 3 hand surgeons scored the cosmetic result (0–10 scoring system) between 5.7 and 6.7. No secondary procedures were done, and all parents were “very satisfied” despite the wide thumbs and split nails. This was attributed to the presence of an ugly preoperative zigzag appearance, and hence the comparative postoperative appearance was satisfactory.

Conclusion: The modified B–C procedure gives a satisfactory function, but the cosmetic outcome is suboptimal. (*Plast Reconstr Surg Glob Open* 2017;6:e1589; doi: 10.1097/GOX.0000000000001589; Published online 28 December 2017.)

INTRODUCTION

There are 4 types of surgical procedures for the correction of thumb polydactyly. “Simple excision” is usually done if one of the duplicates is floating or severely hypoplastic. Most cases are treated with “reconstruction”: the larger, more-functional duplicate is retained and reconstructed using techniques such as collateral ligament reconstruction, intrinsic muscle reattachment, intrinsic tendon balancing/reinsertion and soft-tissue augmentation from the excised smaller, less-functional duplicate. The third option is the “on-top plasty,” which is best suited for duplications with one thumb being adequate proximally and the other thumb containing a superior nail and pulp distally. Finally, the Bilhaut–Cloquet (B–C)

procedure is occasionally done when both thumbs are equal in length and size but neither is thought to be sufficient for reconstruction on its own. In the B–C procedure, the central parts of the duplicates are discarded, and the outer parts are combined to form the new thumb.¹

The use of the B–C procedure in thumb duplication has well-known drawbacks: the new thumb tends to be wider than normal, the new nail will be ridged in the center, and there is a variable degree of stiffness at the involved joints. Some hand surgeons clearly stated that the B–C procedure should be abandoned.^{2,3} Others recommended the B–C procedure for Wassel types I and II duplications and not for more proximal duplication types.⁴ Restricting the B–C procedure for Wassel types I and II results in stiffness at the interphalangeal joint (IPJ) only, which is well tolerated. In contrast, if the procedure is done for Wassel types III and IV, the resulting stiffness at both the IPJ and metacarpophalangeal joint (MPJ) is considered unacceptable. However, several authors still advocate the B–C procedure for types III, IV, and VII duplications.^{5–14} Except for the stiffness, these authors noted several advantages of the B–C procedure including better joint stability, alignment, and overall function.

The senior author (MMA) has used a modified B–C procedure in 4 cases of Wassel types III and IV zigzag thumb

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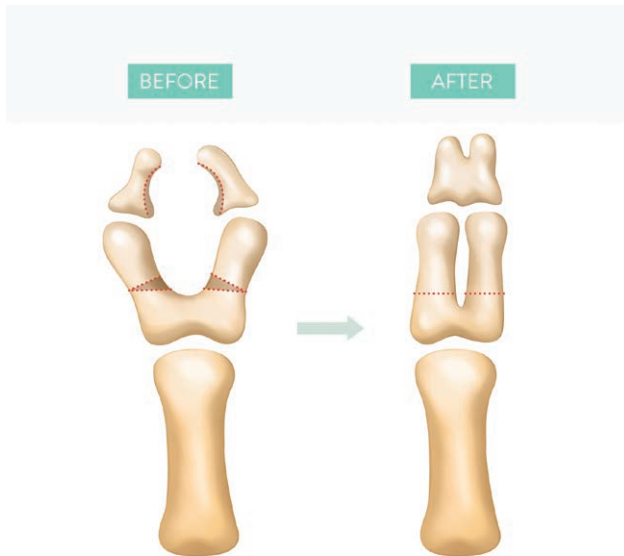


Fig. 1. The modified B–C procedure for Wassel type III thumb polydactyly. The shaded areas are excised. Each closing wedge osteotomy is fixed with a single interosseous wire.

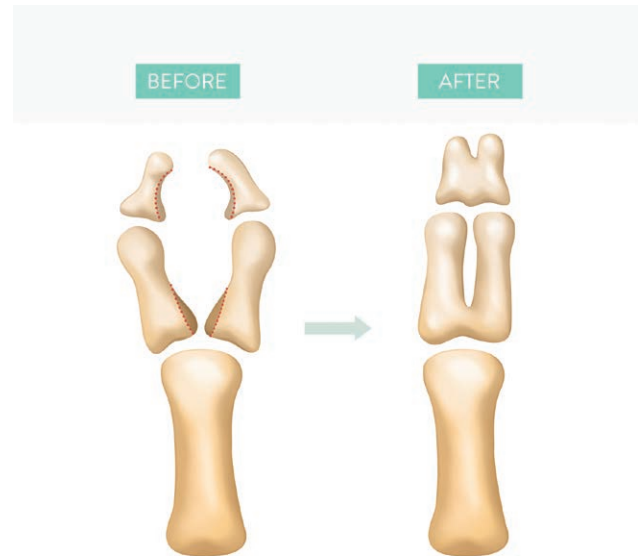


Fig. 2. The modified B–C procedure for Wassel type IV thumb polydactyly. The shaded areas are excised.

duplications aiming to minimize the stiffness at both IPJ and MPJ. The current communication describes the modification and reports on the results of these 4 cases.

PATIENTS AND METHODS

This is a retrospective study of a total of 4 cases of zigzag thumb polydactyly types III ($n = 1$) and IV ($n = 3$) treated with a modified B–C procedure. The aim of the modification was to minimize the stiffness at the IPJ and MPJ by minimizing resections at the joints. However, the modification was expected to result in wide thumbs. This disadvantage was explained to the parents before surgery.

Surgical Technique

In type III zigzag polydactyly (Fig. 1), the central cortices of the distal phalanges are removed. A closed wedge osteotomy is done at the level of the diaphysis of proximal phalanx, and the osteotomy is fixed with a single interosseous wire. Towel clips are applied to approximate the phalanges and correct the zigzag deformity, and transverse K-wires are used to maintain this approximation. The towel clips are removed, and a thumb spica cast is applied for 3 weeks. The transverse K-wires are removed in the clinic at time of cast removal.

In type IV zigzag polydactyly (Fig. 2), the central cortices of the distal phalanges are removed. Central resections from the proximal phalanges are done at the level of the metaphysis, and lower diaphysis was also excised. Towel clips are applied to approximate the phalanges and correct the zigzag deformity, and transverse K-wires are used to maintain this approximation. The towel clips are removed, and a thumb spica cast is applied for 3 weeks. The K-wires are removed in the clinic at the time of cast removal.

In all cases, repair of the nail bed is done using absorbable sutures under magnification. No extrinsic tendon repositioning is done.

Table 1. Assessment of the Cosmetic Outcome

Parameter	Description
A, Size	
Nail width	Compared to the normal contralateral side, the size is reported as either:
Pulp circumference	Similar: if the size discrepancy is within 3 mm
Width at the level of the proximal phalanx	Too wide: if the discrepancy is more than 3 mm
B, Lateral prominence	Reported as either present or absent
At the level of the IPJ	
At the level of MPJ	
C, Ridging of the nail	Reported as either present or absent

Table 2. Functional Assessment

Parameter	Description
A, Active range of motion	Documented as degrees of motion at the joints
At the IPJ	
At the MPJ	
Combined motion at both IPJ and MPJ	
B, Extension lag	Documented as degrees of extension lag at the joints
At the IPJ	
At the MPJ	
C, Joint stability	The joints are stressed, and any instability is measured in degrees
At the IPJ	
At the MPJ	
D, Malalignment	Any malalignment of the thumb ray is measured in degrees

Assessment

Cosmetic and functional outcome measures were documented at final follow-up as shown in Tables 1 and 2. Furthermore, the overall functional outcome was measured using the scoring system of Tada et al. (1983) as shown in Table 3. Finally, the pre- and postoperative clinical and radiological illustrations of the 4 cases

Table 3. Tada Overall Functional Scoring for Treated Duplicate Thumbs

Parameter	Description	Score*
Overall active range of motion at both the IPJ and MPJ	More than 70°	2
	50°–70°	1
	Less than 50°	0
Joint stability	Absent or less than 5°	1
	More than 5°	0
Malalignment	Absent or less than 10°	2
	10°–20°	1
	More than 20°	0

*A total score of 4–5 is considered as good, 2–3 as fair, and 0–1 as poor.

(Figs. 3–6) were shown to a panel of 3 hand surgeons. A list of 6 questions was given to the panel as shown in Table 4. The aim of the first 2 questions was to evaluate

and score the overall cosmetic results. As mentioned in the introduction, the use of the B–C procedure for thumb polydactyly is a controversial issue. Hence, questions #3–6 were given to the panel to explore this controversial issue (Table 4). Finally, the parents were asked 2 questions regarding the use of the thumb in daily activities, and to assess the overall satisfaction as shown in Table 5.

RESULTS

Demographic data are shown in Table 6. The pre- and postoperative clinical and radiological illustrations are shown in Figures 3–6. Assessment of the cosmetic outcome (as per Table 1) was similar in all cases: all size parameters showed a discrepancy of more than 3 mm (ie, too



Fig. 3. Case #1 with type IV duplication. A, Preoperative appearance, (B) marking, (C) immediate postoperative appearance, (D) final postoperative appearance, (E) preoperative X-ray, and (F) final postoperative X-ray. Note that bony union was obtained at the distal phalanges and not at the proximal phalanges.

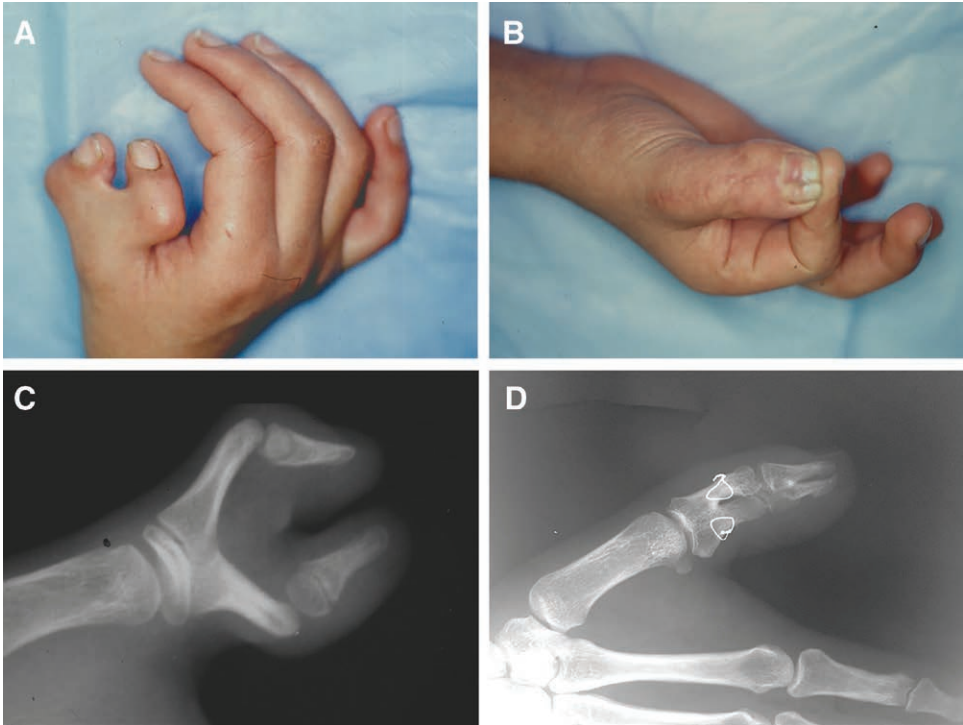


Fig. 4. Case 2 with type III duplication. A, Preoperative appearance, (B) final postoperative appearance, (C) preoperative X-ray, and (D) final postoperative X-ray. There was bony union at the distal and proximal phalanges.

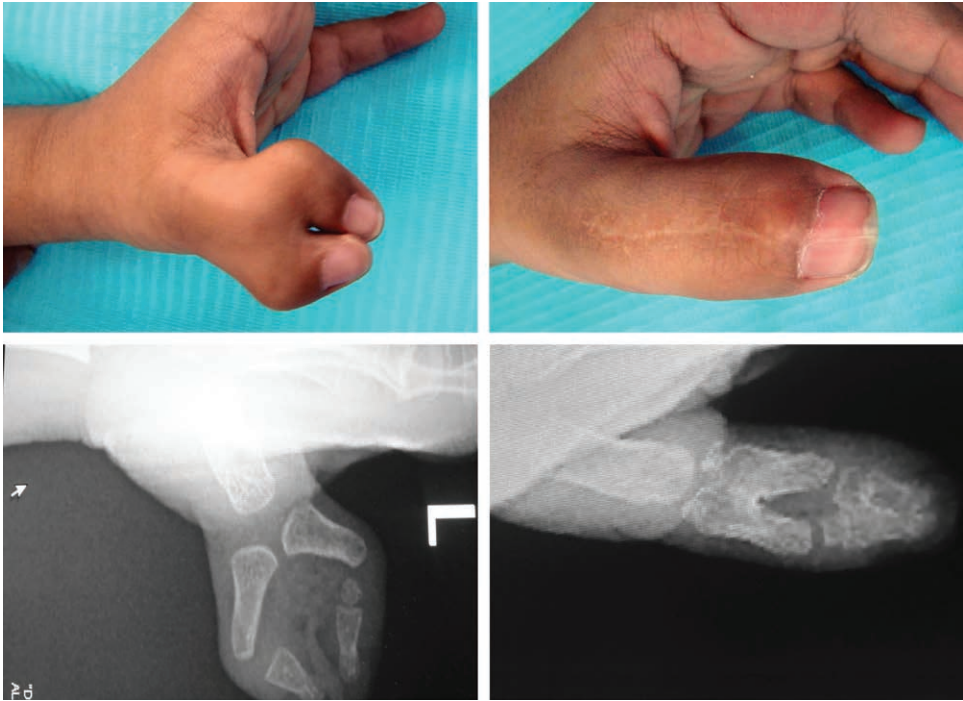


Fig. 5. Case 3 with type IV duplication. A, Preoperative appearance, (B) final postoperative appearance, (C) preoperative X-ray, and (D) final postoperative X-ray. There was bony union at the distal and proximal phalanges.



Fig. 6. Case 4 with type IV duplication. A, Preoperative appearance, (B) final postoperative appearance, (C) preoperative X-ray, and (D) final postoperative X-ray. There was bony union at the distal phalanges only.

Table 4. Questions Given to the Panel of Hand Surgeons

1	Do you think that this cosmetic result is acceptable?
2	How would you score the cosmetic result out of a scale of 0–10?
3	For this case of zigzag duplication, will you go for the B–C procedure or you will go for reconstruction (meaning resection of one component along with collateral ligament reconstruction, osteotomies, K-wires, with soft tissue/bony augmentation where appropriate)?
4	If one chooses to go for reconstruction, do you think that this can be accomplished for this zigzag duplication in one stage or do you think that a second stage will likely be required?
5	Do you think that the B–C procedure should be abandoned?
6	If you think that the B–C procedure still has a place for types III/IV, what would be the best indication for choosing it if both components are approximately equal in length? (choose one answer):
	A, Each component is very small in width, and hence the new thumb will be too narrow.
	B, If the preoperative examination shows significant instabilities of the joints and thus postoperative instability is likely regardless of the technique of reconstruction.
	C, Zigzag duplication.

Table 5. Questions Answered by the Parents

Questions	Choices Given for Answering the Question
The use of the thumb in daily activity	Excellent Occasional Rarely used
Overall satisfaction with the outcome	Very satisfied Somewhat satisfied Not satisfied

wide). Note should be given that size comparison was not possible in case #1 because the contralateral thumb was hypoplastic, but the appearance was also judged as “too wide.” All 4 cases had lateral prominences at the IPJ. Case

Table 6. Demographic Data of the 4 Patients

Patient	Figure Number	Wassel Type	Age at Surgery (y)	Follow-up (y)
1	Figure 3	IV	1	5
2	Figure 4	III	10	10
3	Figure 5	IV	1	2
4	Figure 6	IV	1	0.5

#2 also had a lateral prominence at the MPJ. Finally, the nail was ridged in all 4 cases.

The functional results are shown in Table 7. Note should be given that none of the cases had an extension lag, joint instability, or malalignment. Because all cases showed a combined IPJ–MPJ active range of motion greater than 70° and none of the cases showed joint instability or malalignment, all cases qualified for a Tada score of 5 of 5 (as per Tada criteria³ in Table 3).

Tables 8 and 9 show the answers to the questions by the panel of hand surgeons. Although all surgeons considered the result cosmetically acceptable, the mean cosmetic scores (of 10) varied from 5.7 to 6.7 only, indicating that the overall cosmetic outcome was still suboptimal (Table 8). Table 9 shows that all 3 surgeons supported the use of the B–C procedure for the cases presented. Furthermore, all surgeons did not advocate the opinion of abandoning the B–C procedure and thought that the best indication of the procedure in Wassel types III and IV would be the zigzag deformity.

Finally, the parents of all cases reported the excellent use of the thumb in daily activities by all children, and all parents were “very satisfied.”

DISCUSSION

This article demonstrates an excellent functional outcome (Table 7) and a Tada functional score of 5 of 5 in all cases treated by the modified B–C procedure. Cosmetically, however, all thumbs were “too wide,” and the mean cosmetic score by the panel did not reach a score of 7 in any of the cases (Table 8). Yet, the panel thought that the overall cosmetic result is “acceptable” (Table 8), and all parents were “very satisfied” with the result. This may be due to the fact that all of our cases had an ugly zigzag deformity, and, in comparison, the postoperative appearance gives the impression of a good cosmetic outcome despite the excessive width and the split-nail appearance. We emphasize the importance of preoperative discussion with the family about what can be expected, with special attention regarding the aesthetic outcome. Furthermore, the importance of an informed consent of the possible complications, aesthetics, and long-term results/function is also emphasized in these cases.

A review of the English literature over the least 43 years revealed only a total of 53 cases of Wassel types III, IV, and VII treated by the B–C procedure^{2,3,5,6,8,9,11,12,14–18}, and these cases are summarized in Table 10. However, the detailed results and range of motion were only given in 3 series.^{2,5,14} Dijkman et al.² reported the overall results in 8 cases (3 type II and 5 type IV cases) without specifying the

Table 7. Functional Results in 4 Cases of Thumb Polydactyly Treated with the Modified B–C Procedure

Parameter	Case 1	Case 2	Case 3	Case 4
A, Active range of motion				
At the IPJ	20°	25°	20°	15°
At the MPJ	65°	60°	55°	60°
Combined motion (IPJ and MPJ)	85°	85°	75°	75°
B, Extension lag				
At the IPJ	No extension lag	No extension lag	No extension lag	No extension lag
At the MPJ	at either joint	at either joint	at either joint	at either joint
C, Joint stability				
At the IPJ	No instability	No instability	No instability	No instability
At the MPJ	at either joint	at either joint	at either joint	at either joint
D, Degrees of malalignment	0	0	0	0

Table 8. Assessment of the Cosmetic Results of the 4 Cases by 3 Hand Surgeons (Surgeons Are Labeled as I, II, III)

Questions*	Case 1			Case 2			Case 3			Case 4		
	I	II	III	I	II	III	I	II	III	I	II	III
Is the cosmetic result acceptable (yes or no)?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Score the cosmetic result out of a scale of 1–10	5	7	8	4	6	7	6	7	7	5	6	7
Mean cosmetic score for every case	6.7			5.7			6.7			6		

*The questions are related to questions #1 and 2 from Table 4.

Table 9. Panel Opinion for Questions Related to the Use of B–C Procedure in Thumb Polydactyly (the 3 Hand Surgeons in the Panel Are Labeled as I, II, III)

Questions*	Case #1			Case #2			Case #3			Case #4		
	I	II	III	I	II	III	I	II	III	I	II	III
Will you go for B–C procedure or reconstruction for this case?	B–C	B–C	B–C	B–C	B–C	B–C	B–C	B–C	B–C	B–C	B–C	B–C
If one chooses reconstruction, do you think a second stage will likely be required?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Do you think that the B–C procedure should be abandoned?	All 3 surgeons answered “no”											
The best indication for the B–C procedure in types III/IV polydactyly	All 3 surgeons choose “the zigzag deformity”											

*These questions are related to questions 3–6 from Table 4.

results for the type IV cases, and hence we could not compare their results to ours. Table 11 compares our results to the results of the other 2 series.^{5,14} The 4 cases of Abid et al.⁵ had zigzag Wassel type IV thumb duplication. In all cases, only one nail was retained, and central bony excision was unequal at the distal phalanges and equal at the proximal phalanges. In the series of Tonkin and Bulstrode¹⁴ of 5 cases, 1 nail was retained (with unequal bony excision at the distal phalanges) in 1 case. The remaining 4 cases had classic B–C procedure with shared nails and equal bony excisions. However, Tonkin and Bulstrode¹⁴ made special attention to physal matching and joint congruity to improve the range of motion. Table 11 clearly demonstrates that the cosmetic results in our series are inferior to the cosmetic results of the other 2 series. Functionally, however, our patients had better range of motion and better functional scores. The range of motion was severely restricted at both IPJ and MPJ in the series of Abid et al.⁵ However, the range motion was better in the series of Tonkin and Bulstrode,¹⁴ indicating that keeping special attention to joint congruity while doing the classic B–C procedure will improve the range of motion especially at the MPJ. In fact, the differences in range of motion

between our series and their series are small and probably do not justify the “too wide” appearance of the thumbs in our series. The senior author has decided to stop using the modified technique and to start utilizing the Tonkin technique in future cases.

The senior author (MMA) believes that the main indication for using the B–C procedure in Wassel types III and IV is the presence of a zigzag deformity, and this was also the opinion of the panel. Although Wassel²¹ did not use the B–C procedure in his series, he stated that the B–C procedure is the best option for the zigzag deformity. Evans⁷ and Abid et al.⁵ also stressed that the B–C procedure was the technique of choice in zigzag types III and IV thumb duplications. Horii et al.²² treated 11 cases of type III thumb duplication with “reconstruction” and reported excellent results in all 11 cases, but none of the cases had a zigzag deformity. In contrast, Kawabata et al.²³ treated the zigzag deformity with “reconstruction” and reported poor results (with regard to instability, malalignment, and extension lag), despite doing secondary procedures in almost all patients. The need for secondary procedures if one elects to treat zigzag thumb dupli-

Table 10. Wassel Types III, IV, and VII Treated by the B–C Procedure and Reported in the English Literature Since 1974

Authors	Number of Cases and Wassel Type	Comment
Abid et al. ⁵	4 cases (all type IV)	Excellent cosmetic results but significant stiffness, with mean range of motion of 10° at the IPJ and 25° at the MPJ
Baek et al. ¹⁶	5 cases (all type III)	Paper aimed to describe the technique—no results
Cooney et al. ¹⁵	2 cases (type not specified)	No results for the B–C cases
Dijkman et al. ²	5 cases (all type IV)	The author compared the results of these 5 B–C cases to type IV cases treated with reconstruction and reported better MPJ stability with the B–C procedure. However, all cosmetic parameters were worse with the B–C procedure and the authors recommended abandoning the B–C procedure
Dobyns et al. ¹⁹	2 cases (Type IV)	The result of one patient only was given at 19-y follow-up, “good function but stiff”
Ganley and Lubahn ⁸	3 cases (2 type III and 1 type IV)	No detailed range of motion given but the authors reported, “satisfactory function and appearance despite the stiffness”
Hartrampf et al. ⁹	3 cases (all type IV)	No detailed range of motion, “good function, stability and alignment”
Iwasawa et al. ¹¹	4 cases (1 type III and 3 type IV)	No range of motion given. Excellent cosmetic results at the nail, detailing the technique of nail repair
Maillet et al. ¹²	3 cases (all type IV)	No range of motion given. A good outcome was reported in 2 cases and a fair outcome in 2 cases
Naasan and Page ²⁰	4 cases (2 type III and 2 type IV)	No range of motion results. Both type III cases required reoperation because of excessive width/angulation). One type IV patient required 2 operations to correct nail problems
Ogino et al. ¹⁶	3 cases (1 type III and 2 type IV)	No range of motion or detailed results, but the overall outcome was considered good in all cases
Samson et al. ¹⁷	4 cases (all type IV)	No detailed range of motion given. Stiffness was reported in all cases, and it was severe in 1 case. The overall result was excellent in 1 case, good in 2 cases, and poor in 1 case
Tada et al. ³	4 cases (all type IV)	No detailed range of motion given for the B–C cases, but all had “significant stiffness” at both the IPJ and MPJ. The authors recommend abandoning the B–C procedure
Tonkin and Bulstrode ¹⁴	5 cases (1 type III, 1 type IV, 3 type VII)	Detailed results were given including the range of motion. The mean motion at the IPJ was 13° and at the MPJ was 55°. Tada scores were 5 in 3 cases, 4 in 1 case, and 3 in 1 case
Townsend et al. ¹⁸	2 cases (both type IV)	No detailed range of motion or results given. Severe stiffness was noted in both cases, and the authors recommended abandoning the B–C procedure except for cases with severe hypoplasia of both duplicates

Table 11. Comparison Between Our Results in the Current Series and the Results of Abid et al.⁵ and Tonkin and Bulstrode¹⁴

Parameter	Current Series (n = 4 cases)	Abid et al (2010) (n = 4 Cases)	Tonkin and Bulstrode (2007) (n = 5 Cases)
Mean active motion at the IPJ	20°	10°	13°
Mean active motion at the MPJ	60°	25°	55°
Stability at the IPJ/MPJ	Stable in all cases	Stable in all cases	IPJ instability in 1 case. All other cases had stable IPJ. The MPJ was stable in all 5 cases
Malalignment	No malalignment	No malalignment	No malalignment
Mean Tada functional score (of 5)	5	“Good” score by a modified Tada score	4.4
Nail width/thumb width	All cases were “too wide”	Not specifically measured but illustrations showed “similar” width	All cases were “similar” in width (within 3 mm of the contralateral side)
Overall cosmetic assessment	Acceptable, but the mean panel score for the 4 cases was low and varied from 5.7 to 6.7	Excellent (no panel scoring was done)	Excellent (no panel scoring was done)

cations with “reconstruction” was also reported by our panel (Table 9).

Both Tada et al.³ and Dijkman et al.² clearly stated that the B–C procedure should be abandoned. Other authors did not report a single case of B–C procedure in their series²⁴ or only used it for types I and II duplications.⁴ We share the opinion of the panel (Table 9) that the B–C procedure has a place in the treatment of thumb polydactyly, and the best indication would be in zigzag deformities.

Tonkin has also shown excellent results utilizing the classic B–C procedure for all types of thumb duplications.^{14,25}

In conclusion, the aim of the modified B–C procedure in this series was to improve the range of motion, but at the expense of accepting a wide thumb. The modification obtained an excellent range of motion at the MPJ but a poor result at the IPJ. The reason for that is unclear, but it may be related to the presence of a severe preoperative joint incongruity at the IPJ. If one looks at the preoperative

x-rays (Figs. 3–6), the base of the distal phalanx is articulating with the side of the head of the proximal phalanx because of the zigzag deformity. A simple joint reduction is not expected to produce a congruent IPJ. Failure to obtain a near-full range of motion and the relatively poor cosmetic outcome made the senior author decide to try the Tonkin technique in future patients with zigzag deformity and compare the results to the current series.

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