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 from the new thumb.

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. Others recommended the B–C procedure for Wassel types I and II duplications and not for more proximal duplication types. 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, IV, and VII duplications, 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. Except for the stiffness, these authors noted several advantages of the B–C procedure including better joint stability, alignment, and overall function.

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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.
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 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.

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.
(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

| Parameter                  | Description | Score* |
|----------------------------|-------------|--------|
| Overall active range of motion at both 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.

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.
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 #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 criteria 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, and these cases are summarized in Table 10. However, the detailed results and range of motion were only given in 3 series. Dijkman et al. reported the overall results in 8 cases (3 type II and 5 type IV cases) without specifying the

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**Table 4. Questions Given to the Panel of Hand Surgeons**

| Question | Options |
|----------|---------|
| 1. Do you think that this cosmetic result is acceptable? | Yes/No |
| 2. How would you score the cosmetic result out of a scale of 0–10? | 1-10 |
| 3. For this case of zigzag duplication, will you go for the B–C procedure or will you go for reconstruction (meaning resection of one component along with collateral ligament reconstruction, osteotomies, K-wires, with soft tissue/bony augmentation where appropriate)? | Yes/No |
| 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? | Yes/No |
| 5. Do you think that the B–C procedure should be abandoned? | Yes/No |
| 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): | Option A/B/C |
| 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 |

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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
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 physeal 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 duplic-

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**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 |

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**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 |
| Score the cosmetic result out of a scale of 1–10 | 5 | 7 | 8 | 4 |
| Mean cosmetic score for every case | 6.7 | 5.7 | 6.7 | 5 |

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**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 |        |        |        |        |
| Will you go for B–C procedure or reconstruction for this case? | 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 |
| 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” |       |       |       |

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*These questions are related to questions 3–6 from Table 4.
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 series 24 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.

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. 15     | 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”, “good function, stability and alignment” |
| Hartrampf et al. 9    | 3 cases (all type IV)           | No detailed range of motion given.                                                                 |
| 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 10    | 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 15° 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 Bulstrode 14

| 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) |

Tonkin has also shown excellent results utilizing the classic B–C procedure for all types of thumb duplications. 12,25

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 articu-
lating 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 ob-
tain 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 defor-
mity and compare the results to the current series.

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