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Original Article

Effects of the modified Bilhaut-Cloquet surgery on the treatment of congenital duplication of thumb in children

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Abstract: Objective: To study and compare the clinical therapeutic effects of the modified Bilhaut-Cloquet surgery on the treatment of congenital duplication of thumb (CDT) in children. Methods: A retrospective study was conducted on the 110 children with CDT who received surgical treatment in our hospital from November 2014 to November 2016. Namely, all the general information, conditions, operative methods, postoperative recovery qualities, complications, etc. of the patients were evaluated. Besides, the correlations between different operative methods and postoperative recovery conditions were explored. Results: Because some patients lost contact, eventually the follow-up data of 100 patients were collected and among these patients, no one had wound infection, necrosis, internal fixation removal, bone nonunion, myasthenia or relapse, etc. Among them, Bilhaut-Cloquet surgery was applied on 59 patients (Group A), and the other 41 patients were treated with the modified Bilhaut-Cloquet surgery (Group B). The incidence of adverse events in Group A (10.17%) was higher than that in Group B (4.88%). The scores of range of motion and appearance in Group B were all higher than those in Group A. However, the scores of joint stability and angular deformity in Group B were lower than those in Group A (All $P<0.05$). The excellent and good rate of Group B (95.12%) was significantly higher than that of Group A (83.05%). The logistic regression analysis indicated that the modified Bilhaut-Cloquet surgery was related to a better postoperative recovery. Conclusion: The modified Bilhaut-Cloquet surgery can decrease the incidence of adverse events, improve the scores of range of motion and appearance and enhance the overall therapeutic effects.

Keywords: Congenital duplication of thumb, operative timing, ossification of thumb

Introduction

Congenital deformity of thumb is a kind of common congenital malformation with polydactyly as one of the commonest types [1]. It appears as a splitting duplication of the thumb, and its anatomical morphology occurs in a variety of variations. The clinical treatment of this disease was quite complex and most patients still could not be cured completely and return to absolute normal through the treatment [2]. Plenty ofresearches reported that the incidence is generally 0.08%-0.14%, the cause may be that the necrosis of the ectoderm and axis anterior mesoderm is disturbed 8 weeks before the embryonic development [3]. Consequently, the major goal of the treatment was to improve the appearances and limb functions of patients. It is also important to confirm the pathological anatomy and morphological features of the thumb. The vast majority of clinical cases were treated by surgery. However, the operative method and the appropriate time to have an operation of children were problems that we needed to consider clinically. Because the condition of congenital duplication of thumb (CDT) in children is quite complicated, the clinical manifestations are different, so it is necessary to choose the surgical method according to the actual situation [4]. On the one hand, it is necessary to remove the extra knuckles to improve the appearance of the thumb, on the other hand, the surgery is to obtain the stability of the joints, a good line of force, the maximum degree of joint mobility and strong grip function. At present, the most commonly used method is Blihaut-Cloquet surgery, which has achieved certain clinical efficacy. But there are several defects, such as splitting, malposition and unaesthetic outcomeof nail etc., and these

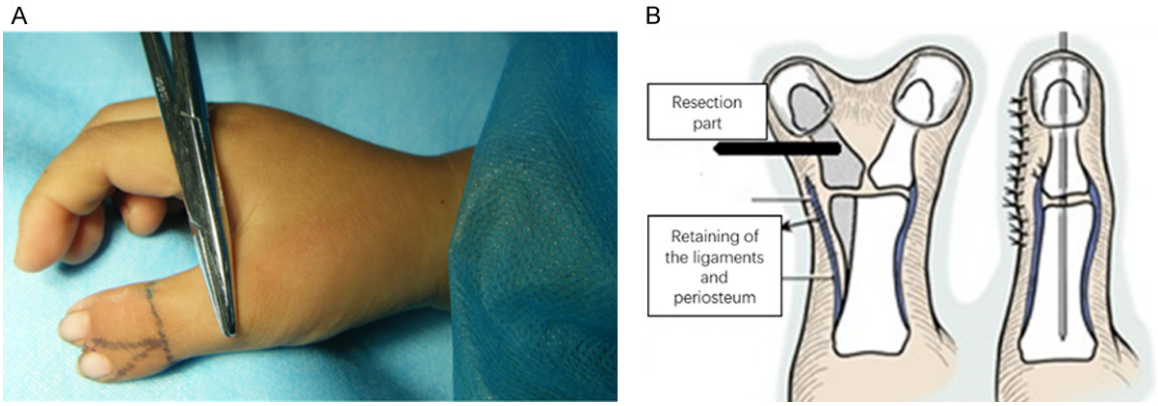

Figure 1. A. Traditional Bilhaut-Cloquet surgery; B. Surgical anatomy

many postoperative complications directly affect the overall effect of the operation [5]. Now, this Blihaut-Cloquet surgerywas modified by our hospital via preserving a better developed finger, in order to get more satisfying effects in functional and aesthetic aspects. The postoperative complications and aesthetic effects were significantly better than the traditional Blihaut-Cloquet surgery. This paper was a retrospective study on CDT of the children who received treatment in our hospital and aimed to explore the clinical therapeutic effects of the modified Bilhaut-Cloquet surgery on the treatment of CDT.

Materials and methods

Criteria for inclusion and exclusion

The study objects were 100 children with CDT who have received surgical treatment in the surgical department of our hospital from November 2014 to November 2016. The clinical data of these included patients were collected for the further retrospective study.

Inclusion criteria: Children who met the diagnostic criteria of CDT, who were diagnosed by clinical physicians and who belonged to Wassel Type-II; children who were 14 years old or below; patients who met the standard of surgical treatment.

Exclusion criteria: Children with incomplete clinical materials; patients with other illnesses.

At length, a total of 100 cases were included as study objects. This study met the requirements of Ethics Committee of our hospital and was approved by them.

Data collection

All the data of the included patients were collected, such as their ages, sexes, types of polydactyly, operative methods, detailed surgical conditions, complications and postoperative recovery condition, etc.

Grouping

There were two kinds of operative methods being applied to the patients, one was the conventional Bilhaut-Cloquet surgery, and the other was the modified Bilhaut-Cloquet surgery. There were 59 cases (35 males and 24 females; 36 cases of right sides, 14 cases of left sides, 8 cases of bilateral side and 1 case of syndactyly) in the Group who underwent the conventional Bilhaut-Cloquet surgery (Group A). At the same time, there were a total of 41 cases (24 males and 17 females; 22 cases of right sides, 10 cases of left sides, 8 cases of bilateral side and 1 case of syndactyly) in the Group who underwent the modified Bilhaut-Cloquet surgery (Group B).

As for every patient in Group A, a wedge incision was cut from the dorsal part of thumb, which was passing the nail and nail bed and reaching from the proximate end to the bifurcation site of thumb. Firstly, subcutaneous tissues were separated from the site of nail bed, so the bone and articular facet of thumb were exposed. Then longitudinal wedge incisions were performed at the articular facets, bones and tendons of two thumbs. Furthermore, after the joint capsule between the thumbs was rejected, thumbs were ready to be recombined. Afterwards, the articular facets of left phalanxes were connected meticulously and fixed with

Table 1. Comparison of the basic information of patients in two groups

Groups	Age	Sex ratio		Deformity type			
		Male	Female	Right	Left	Bilateral	Syndactyly
Group A (n=59)	1.78±0.34	35	24	36	14	8	1
Group B (n=41)	1.83±0.61	24	17	22	10	8	1
t/X^2	t=0.381	X^2=0.831		X^2=1.384			
P value	0.089	0.451		0.094			

Table 2. Comparison of the follow up situations of two groups

Groups	Purulent exudate	Joint oblique slant	Joint stiffness	Total incidence
Group A (n=59)	2 (3.39)	4 (6.78)	0	6 (10.17)
Group B (n=41)	0	0	2 (4.88)	2 (4.88)
X^2 value	0.589	0.589	0.409	8.394
P value	0.004	0.002	0.003	0.000

Kirschner wire in transverse direction. At last, the nail was repaired and skin was sutured carefully to finish the surgery [6]. See **Figure 1**.

The operative method of Group B was modified based on that of Group A. A wedge incision was changed to perform in the direction of cutting from dorsal thumb towards the better developed finger. Moreover, the same operative procedures of Group A were carried on and ended with unequal sutures at the places of nail and nail bed to ensure joint stability. Finally, surgery was completed with the sutured skin [7].

Observation indexes

The patients were followed up after surgery. And the range of motion and stability of joints, angular deformity, appearance and recovery of thumb function of every patient were recorded.

Statistical methods

Statistical analysis was performed by the software SPSS 17.0 for the data and results of this study. The enumeration data were expressed as frequencies and percentages, and the comparison between groups was tested by χ^2 test. The measurement data were expressed as mean ± standard deviation, and the comparison between groups was tested by *t* test. The logistic regression analysis was used to analyze the potential factors affecting posto-perative recovery. When *P* value was lower than 0.05, the differences had statistical significance.

Results

General information of included patients

There were a total of 100 cases of children (59 males and 41 females). The age of patients ranged 2-13 years old with an average age of 1.62±2.36 years old. As for the deformity sites, there were 58 cases of right sides, 24 cases of left sides, 16 cases of bilateral and 2 cases of syndactylies. As shown in **Table 1**, the differences in general information between two groups had no statistical significance. All the *P* values were above 0.05, so the following data were comparable.

Comparison of the follow up situations of two groups

A total of 100 cases of patients were followed up with 59 cases in Group A while 41 cases in Group B. There was no wound infection, necrosis, internal fixation removal, bone nonunion, myasthenia or relapse etc. in both groups. The incidence of adverse events of Group A (10.17%) was higher than that of Group B (4.88%), the difference was statistically significant, as shown in **Table 2**.

Comparison of appearance and function situations of patients' thumbs in two groups

As shown in **Table 3**, the scores of range of motion and appearance of Group B were all higher than those of Group A. Nevertheless, the scores of joint stability and angular deformity of Group B were all lower than those of Group A with P<0.05 for all.

Comparison of the excellent and good rate of treatment effect of patients in two groups

As shown in **Table 4**, the excellent and good rate of Group B (95.12%) was apparently higher than that of Group A (83.05%). The difference was statistically significance.

Table 3. Comparison of appearance and function situations of patients' thumbs in two groups

Groups	SRM	SJS	AD	Appearance
Group A (n=59)	1.07±0.23	0.97±0.12	2.03±0.21	7
Group B (n=41)	1.78±0.34	0.78±0.11	0.89±0.14	9
t value	8.392	9.114	11.491	6.049
P value	0.012	0.009	0.005	0.003

Note: SRM, score of range of motion; SJS, score of joint stability; AD, angular deformity.

Table 4. Comparison of the excellent and good rate of treatment effect of patients in two groups

Groups	Excellent	Good	Bad	Excellent and good rate
Group A (n=59)	26 (44.07)	23 (38.98)	10 (16.95)	49 (83.05)
Group B (n=41)	22 (53.66)	17 (41.46)	2 (4.88)	39 (95.12)
X^2 value	0.020	0.024	5.587	9.783
P value	0.004	0.005	0.005	0.005

Table 5. Logistic regression analysis of excellent and good rate of surgery

Factors	RC	SE	X^2	F	OR	P	95% confidence interval	
							Upper limit	Lower limit
Age	-0.627	0.283	4.912	1	0.534	0.067	0.307	0.930
Sex	-0.394	0.239	3.185	1	0.634	0.089	1.656	5.316
DT	0.772	0.288	7.189	1	2.164	0.057	1.231	3.804
OM	1.067	0.450	5.632	1	2.906	0.018	1.204	7.014

Note: RC, regression coefficient; SE, standard error; X^2, Walds (X^2 value); F, degree of freedom; OR, OR value; *P*, *P* value; DT, deformity type; OM, operative method.

Results of the logistic regression analysis

As shown in **Table 5**, the analysis revealed that the operative method was the factor that affected the excellent and good rate of treatment effect, and the modified Bilhaut-Cloquet surgery was related to a higher excellent and good rate of surgery.

Typical cases

As shown in **Figure 2**, the patient who underwent the modified surgery had excellent therapeutic effects with good-looking appearance and achieved the surgery goal. The modified surgery was performed on this male patient who was 8-month-old, and after the surgery he obtained normal function of the finger and achieved satisfaction about the appearance.

Discussion

Thumb duplication is one kind of the congenital deformities of hand, but the final conclusion about its pathogenesis has not been achieved yet. Clinically, the most accepted pathogenesis was about the influence caused by external environment. For example, the mothers took some medicine during their pregnancies or got the impact of ray or disease, which led to the gene mutation of children and their manifestation is polydactyly [8-10]. The goals of the treatment of polydactyly were not only recovering finger function, but also requiring aesthetic appearance. However, the therapeutic effects were closely related to the operation timing and operative methods [11]. For the perspective of operation timing, scholars around the world have made quite a lot of in-depth studies. They all agreed that the operation timing of children' s CDTD need to be determined according to the time of ossification center of thumb, which represented as serious deformities, large variation of thumb bone structure and so forth [12]. The operation timing of every included object in this study was determined by completely following this standard and the practical situation.

The modified Bilhaut-Cloquet surgery by our hospital can solve the disease directly, without the need for secondary correction. In theearly reconstruction of the resection of the duplication of the thumb ligament and posterior osteotomy, there was a good appearance and function of the thumb. At present, the best time for Bilhaut-Cloquet surgery is 12-18 months after birth. During this period, the development of duplication of the thumb deformity is immature, therefore, this surgery is more widely promoted in the early stage.

This study focused on the operative method, as a result, the thumbs which underwent the modified surgery have preserved its original function as well as its aesthetic appearance in this study. The modified Bilhaut-Cloquet sur-

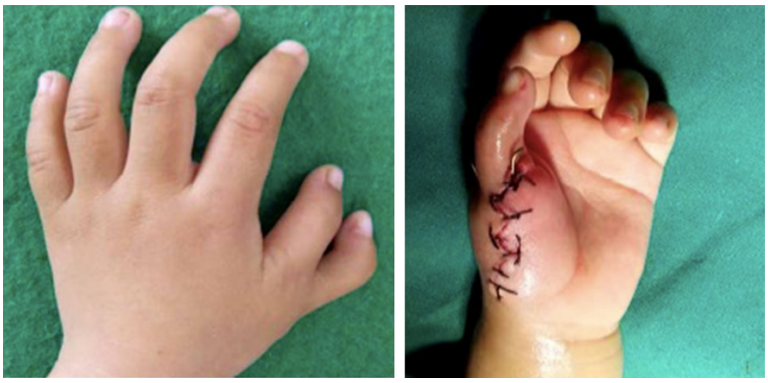

Figure 2. Comparison between the preoperative and postoperative condition of deformity of thumb duplication. The left photo was shot one day before the surgery, which indicated that the patient represented deformity of thumb duplication (one well-developed thumb and the other hypoplastic thumb); the right photo was shot right after the surgery, which indicated that the well-developed thumb was preserved after the surgerywith both good function and aesthetic appearance.

gery could preserve the better developed thumb and then reconstruct or repair the joint capsule or accessory ligament in terms of patients' need [13]. In that way, the remained thumb could preserve its best appearance and function. Particularly, the V-shape incision would not have any impact on the nail bed, finger bones, joints or anything else [14-17]. What's more, hypoplastic thumb was not completely removed; instead, its skin flaps on side were preserved [18]. With the help of its own skin flaps, the incision could be repaired better and at the same time aesthetics of appearance could be kept to the greatest extent. Thereby, the satisfactory postoperative effects were achieved [19, 20]. The study also found that the incidence of adverse events in the traditional group was higher than that in the modified group.

The influence of traditional Bilhaut-Cloquet on joint stabilization system was less than that of the modified Bilhaut-Cloquet surgery. The postoperative effect of modified Bilhaut-Cloquet on the stability of the joint was improved. In cases of traditional surgical treatment adopted by Tonkin et al, the reconstructed thumbs look small with joint deformity and joint instability. In this study, the scores of joint range of motion and appearances of the modified group were better than those of the traditional group, while the scores of stability of joint and angular malformation of the modified group were lower than those of the traditional group, but the improvement rate of the modified group was significantly higher than that of the traditional group.

Logistic regression analysis showed that modified Bilhaut-Cloquet surgery was associated with excellent postoperative recovery. Its possible reason may be that the modified Bilhaut-Cloquet surgery can reduce the occurrence of nail deformity and ankylosis.

Although the application of the modified Bilhaut-Cloquet surgery in clinical surgery is very simple, the treatment effect is quite different. We should strictly grasp the indications of children before the surgery, and confirm the size of the thumb, appose the phalanges, release the collateral ligament and reduce the scar tissue during surgery. The above notes on the postoperative rehabilitation training play vital roles. As for symmetrical thumbs which was bigger than contralateral 2/3 of the Wassel type I thumbs, we recommend that doctors should use Bilhaut-Cloquet surgery; the IPJ activity of Wassel type II thumbs is poor, so it is recommended to carefully select the surgical approach; if children are belonging to cicatricial diathesis or thumb bone asymmetry, the surgerymethod should be carefully selected.

In conclusion, the modified Bilhaut-Cloquet surgery can decrease the incidence of adverse events, improve the scores of range of motion and appearance and enhance the overall therapeutic effects, which is a recommended surgery.

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Disclosure of conflict of interest

None.

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