

## ORIGINAL ARTICLE

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## Funnel chest: treatment strategy and follow-up

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**Abstract** Although funnel chest is the most frequently seen deformity of the anterior chest wall in children, there is still considerable controversy regarding three major aspects, namely, the frequency of such deformities, their physiological importance, and the methods available for treatment. We retrospectively analyzed our experience with the 154 patients managed in our department. In 81 of these an operation was performed (OP), and the clinical findings for this group were compared with the 73 patients in whom an operation was not performed (NOP). Evaluation included subjective findings, especially the views of the patients' parents, and objective findings, including chest radiographs, computed tomography (CT), spirogram, electrocardiography, and echocardiography. In all patients the assessment included postoperative respiratory symptoms, appearance of the chest, and psychological aspects related to the deformity. Post-operatively, respiratory symptoms almost invariably subsided. The cosmetic result could initially be regarded as satisfactory or fair during the first 10 years following surgery, but over time there was frequently increasing concern regarding the scar. The NOP patients showed significantly less severity of the funnel index compared with OP patients. However, there was no spontaneous improvement in the deformity in older patients; most of the NOP patients continued to show a cosmetic deformity and 26.7% had psychological problems. This retrospective study confirms that our treatment strategy of objective criteria for operation (functional compression index  $>0.2$ , % vital capacity  $<80$ , and CT index less than 0.25) and timing of operation (between 4 and 6 years of age) provides good results. Based on the analysis of long-term follow-up, surgery is considered indicated in patients with severe deformity. However, in the interest of psychological

development, the indications for surgery may be extended.

**Keywords** Chest-wall deformity · Funnel chest · Funnel index · Sternotomy · Quality of life

### Introduction

Chest-wall deformities (CWD) can be classified according to the nature of the anatomic abnormality, and a preferable classification that may be adopted [1] enables recognition of three types of deformity: (1) depression deformities (funnel chest, FC); (2) protrusion deformities (pigeon chest); and (3) deficiency deformities. FC is the most frequently encountered deformity, but despite surgical endeavors during the past 50 years there are still considerable differences of opinion regarding the following: (1) the precise indications for operation; (2) the optimal age for such surgery; (3) the technique to be adopted; and (4) the results of operative correction.

In an attempt to answer some of these questions and evaluate results, a retrospective analysis of 154 patients treated at the Department of Pediatric Surgery, Kyushu University, was made and the results are presented in this article. The nature of the investigation is described and the importance of a follow-up study is emphasized.

### Patients and methods

A retrospective analysis was made of the 154 patients managed in our department during the 21-year period 1978–1999; in 81 patients an operation was performed (OP) and 73 were non-operated patients (NOP). The analysis involved two studies.

#### Study 1

A comparison was made of the clinical findings in the two groups and attention was also directed toward the family history,

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sex incidence, incidence of associated congenital anomalies, and recognition of such associations as Marfan's syndrome and Poland syndrome. Investigations performed included chest radiographs, computed tomography (CT), determination of the funnel index (FI) and CT index, spirometry, electrocardiography (ECG), and echocardiography (ECHO). The FI [2] is used to estimate the severity of the deformity (Fig. 1); it has two parameters referred to as the cosmetic compression index ( $F_1I$ ) and the functional compression index ( $F_2I$ ). The CT index [3] is measured as follows: the vertebral sternal distance at the apex of the depression is divided by the internal transverse diameter of the thorax. During the past 12 years ECHO has been performed, and in patients older than 5 years a spirometry was performed (this age was selected to enable appropriate co-operation). The results of these indices were statistically evaluated and Student's *t*-test was applied. *P* values less than 0.05 were considered to be significant.

Since 1978, our strategy for treatment of FC has been as follows: (1) We recommend surgical correction in those patients with a low percent vital capacity (%VC),  $F_2I$  more than 0.2, and strong wishes of the parents and/or patients; (2) The optimal age at operation is between 4 and 6 years; (3) The routine operative procedure is a sternotomy [4]; and (4) Patients who are not candidates for surgical correction at the first evaluation are followed regularly at the outpatient clinic every 6 months.

#### Study 2

In an attempt to evaluate the current status of quality of life in both groups (OP and NOP), a standardized questionnaire was sent to all patients except 14 in the NOP group who were aged less than 3 years, in whom decisions regarding operation had yet to be made. The following questions were asked: (1) How do they feel about the cosmetic result? (2) What comments do they have on psychological aspects? and (3) Are there any current residual problems?

## Results

### Study 1

These results are summarized in Table 1; there were 113 males and 41 females, a ratio of 2.8:1. This ratio was 3.5:1 in the OP patients and 2.2:1 in NOP patients. The deformity was noted at birth in one-third of the cases, while over two-thirds of the patients were identified by the age of 1 year. In the majority, the deformity was initially identified by the parents. In the remainder, recognition followed a visit to the general practitioner for minor ailments or during the course of a school medical examination.

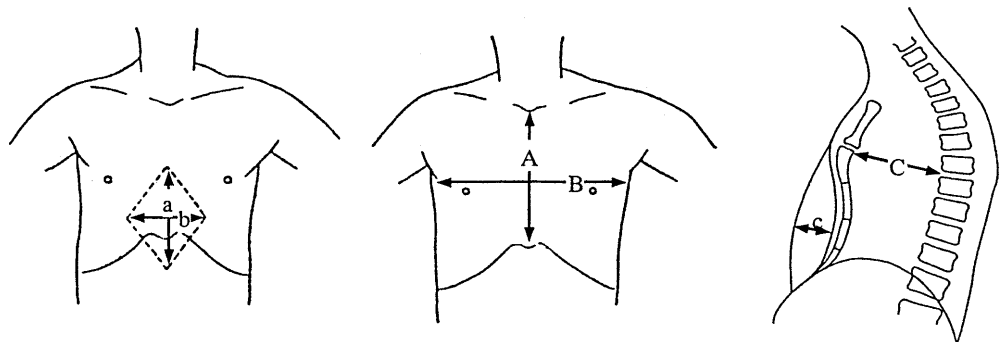
The mean age at consultation was 5.78 (range 2–16 years). There was no significant difference between the two groups. In 19 patients (12.3%) the family history was positive; thus a CWD was present in the parents of 7 patients and siblings of 2 patients, in grandparents in 6 and more distant relatives (cousins, uncles, and aunts) in 4 cases. The incidence of a family history was higher in OP (15/81, 18.5%) than NOP (4/73, 5.5%) patients.

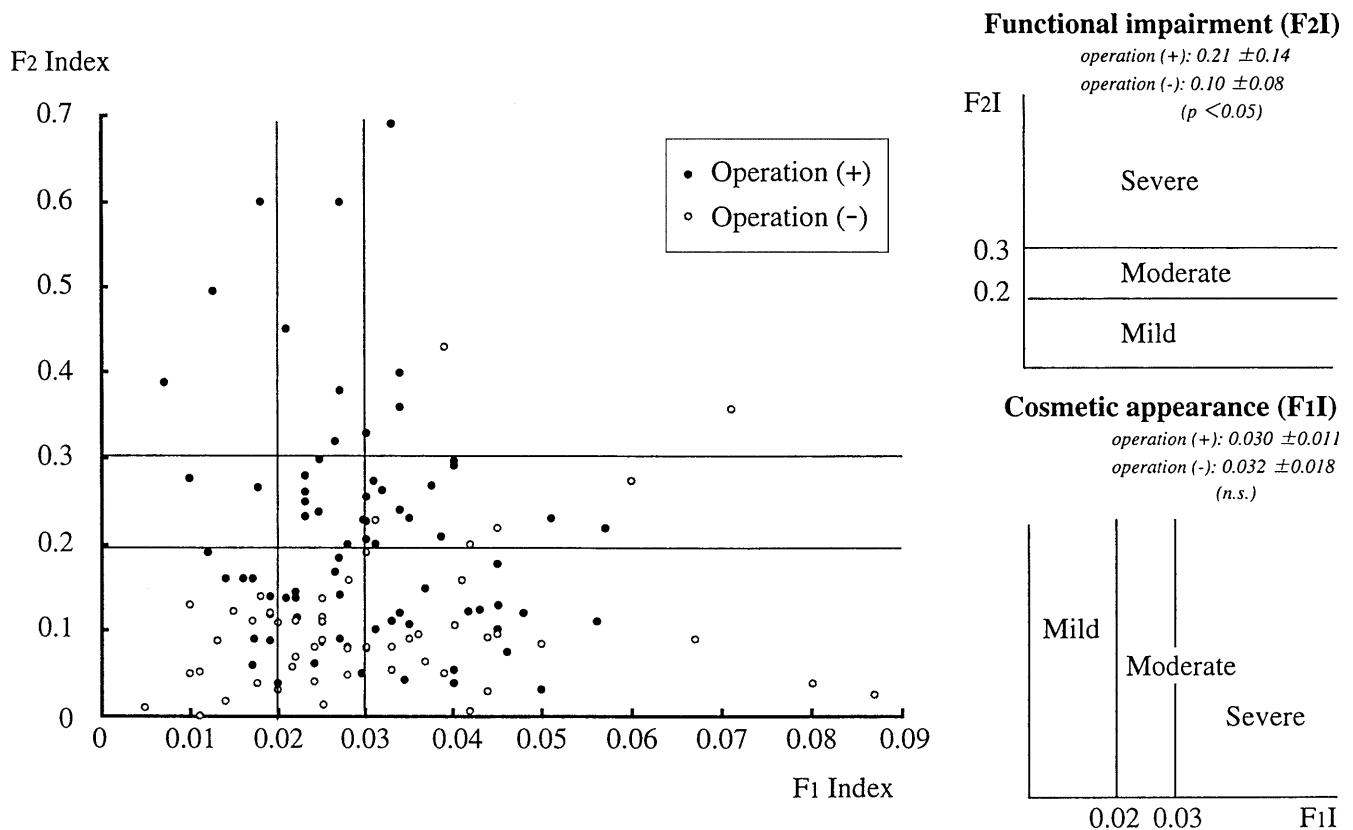
The incidence of associated congenital anomalies was 21.4%. Scoliosis was the most frequently associated finding and was present in 9.7% of cases. There was no diminution of exercise tolerance or shortness of breath in either group, but the OP patients had more frequent episodes of upper respiratory infection (RTI) (53.1%) compared to NOP patients (19.2%). The OP patients frequently commented on fatigability, but with few ex-

**Table 1** Clinical data in 154 patients with funnel chest

Parameter	Operated	Non-operated	Total
Number of patients	81	73	154
Sex incidence (m:f)	3.5:1	2.2:1	2.8:1
Age of presentation (years old)	0.81 ± 1.67	1.63 ± 2.95	1.14 ± 2.30
Age of consultation (years old)	5.50 ± 2.74	6.08 ± 4.49	5.78 ± 3.68
Family history	15/81 (18.5%)	4/73 (5.5%)	19/154 (12.3%)
Associated anomalies	12/81 (14.8%)	21/73 (28.8%)	33/154 (21.4%)
Scoliosis	6/81 (7.4%)	9/73 (12.3%)	15/154 (9.7%)
Marfan's syndrome	2/81 (2.5%)	3/73 (4.1%)	5/154 (3.2%)
Poland syndrome	1/81 (1.2%)	1/73 (1.4%)	2/154 (1.3%)
Frequent episodes of upper respiratory infection	43/81 (53.1%)	14/73 (19.2%)	57/154 (37.0%)

**Fig. 1** Measurement of funnel index: *a* longitudinal length of deformity, *b* width of deformity, *c* depth of deformity, *A* length of sternum, *B* width of chest, *C* minimum length between angle of sternum and anterior part of vertebra  
 $F_1I = c/(a \times b)$   
 $F_2I = (a \times b \times c)/(A \times B \times C)$





**Fig. 2** Funnel index of operated (OP) and non-operated (NOP) patients. Cosmetic compression index (F<sub>1</sub>I) of OP and NOP are  $0.030 \pm 0.011$  and  $0.032 \pm 0.018$ , respectively; functional compression index (F<sub>2</sub>I) of OP and NOP  $0.20 \pm 0.13$  and  $0.10 \pm 0.08$ , respectively

ceptions presentation was related to the cosmetic appearance of the chest wall. In the older children there was a feeling of "shame" in both the parents and patients, and comments from friends led to attempts to cover up the deformity.

The FI (Fig. 2) revealed no difference in F<sub>1</sub>I between the two groups (OP  $0.030 \pm 0.011$ , NOP  $0.032 \pm 0.018$ ), while F<sub>2</sub>I showed a significant difference between OP ( $0.21 \pm 0.14$ ) and NOP ( $0.10 \pm 0.08$ ) ( $P < 0.05$ ). CT examinations have been performed since 1987. The CT index of the OP group was  $0.16 \pm 0.05$  while that of the NOP group was  $0.20 \pm 0.06$  (Fig. 3). The statistical significance of these findings was  $P < 0.05$ .

Abnormalities in ECG findings included incomplete right bundle branch block and axis deviation or an abnormal P wave, and were present in 51.2% of the patients. There was no significant difference between the OP and the NOP groups. No abnormalities were demonstrated on ECHO. The spirographic findings were as follows: average %VC  $79.4 \pm 14.1\%$  in the OP,  $83.7 \pm 17.7\%$  in the NOP group (Fig. 4).

In 41 of the 81 patients in the OP group (60%) (Fig. 5), the operation was performed between the age of 4 and 6 years; 77 (95.1%) had a sternoturnover and 4 had sternocostal elevation. Our sternoturnover tech-

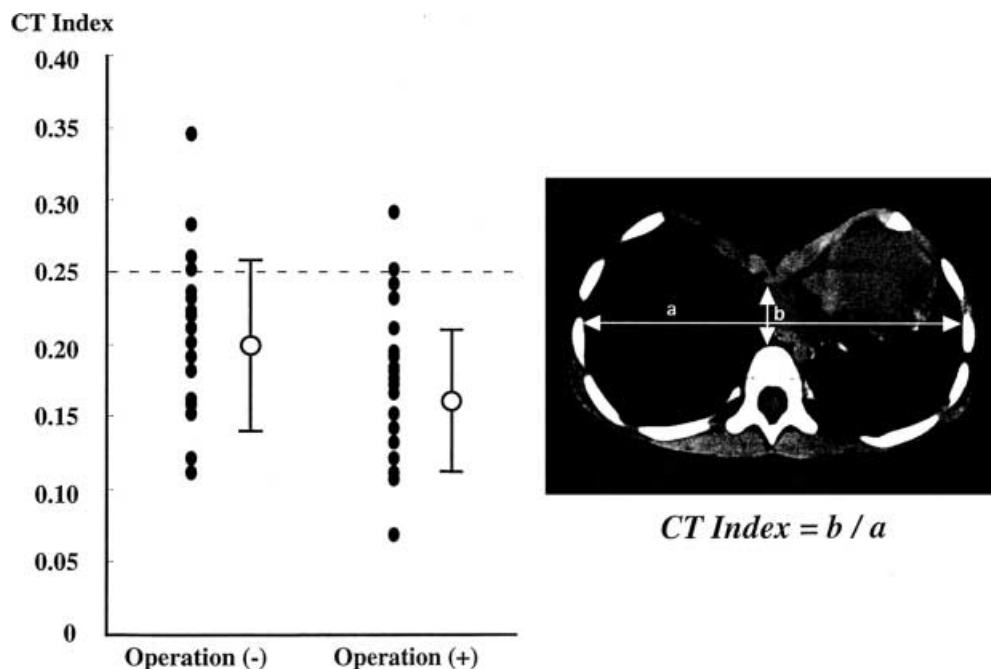
nique was basically the same as previously described by Wada [5] except that we preserved the internal thoracic and intercostal arteries with perichondrium of the sternum and costal cartilage to minimize ischemic changes.

Regarding the operative results, 97% of the patients with preoperative RTI showed a decrease in the frequency and severity of these infections following surgery and also had less fatigue, increased stamina, and more cheerful demeanor. Efforts to hide their chest under clothes tended to cease. In some cases a later problem was a hypertrophic scar.

## Study 2

Sixty of the 81 OP patients (74%) replied to the questionnaire; their average age was  $17.4 \pm 5.7$  years (range 6–29). The answers received are summarized in Tables 2 and 3. Complaints in the OP patients were as follows: scar 5, deformity 3, abnormal ECG 3, past history of blood transfusion 3, dyspnea 1, wound pain 1, frequent RTI 1. A major problem (71.4%) was the reaction to the scar, and some patients required scar revision at a later date. Significant data are summarized in Figs. 6–8; as time passed there was less satisfaction with the result of the operation (Fig. 6). However, there were no physical complaints or evidence of recurrence of the deformity except in a few patients with Marfan's syndrome. The patients operated upon at 4 to 6 years of age had highly satisfac-

**Fig. 3** CT index of operated (OP) and non-operated (NOP) patients: vertebral-sternal distance ( $b$ ) divided by internal transverse distance ( $a$ ) at most depressed portion. CT index of OP ( $0.16 \pm 0.05$ ) significantly lower than NOP ( $0.20 \pm 0.06$ )



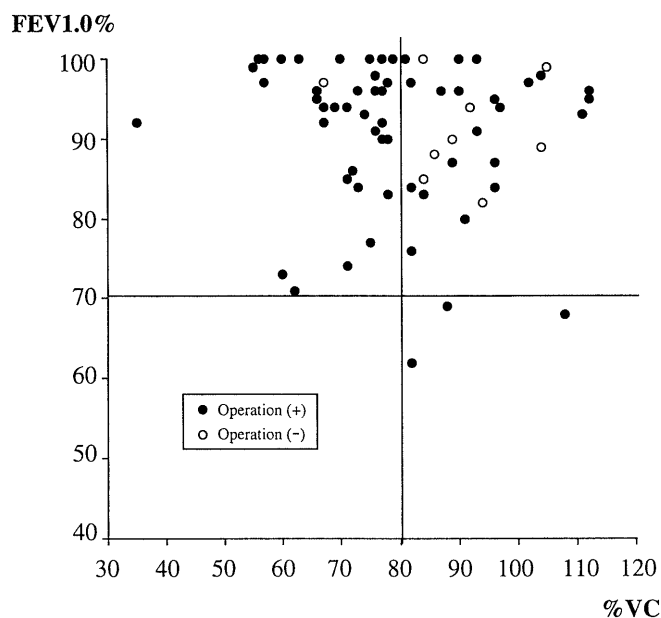
tory results (Fig. 7). Fig. 8 indicates the results in relation to the FL.

Of the 59 patients who had not been managed surgically, 46 (77.9%) replied to the questionnaire, but in 2 cases the answers were incomplete and these were excluded. The average age of these patients was  $16.7 \pm 5.9$  years (range 5–28). Details regarding the answers to the questionnaire in the NOP group are shown in Tables 2 and 3. The relationship between follow-up period and satisfaction is shown in Fig. 9. Many of these patients are

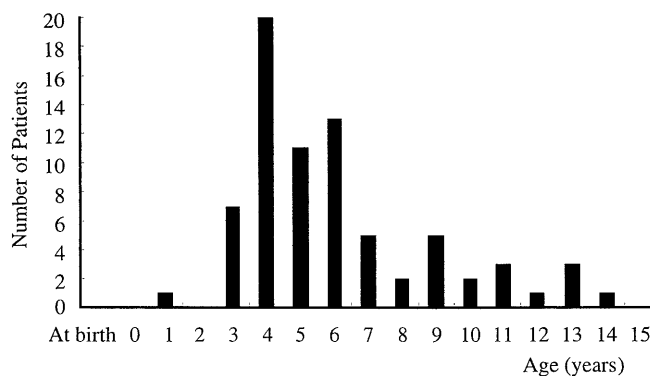
still considering surgical correction, and 4 have undergone surgery at another institution.

## Discussion

The role of surgery in the management of patients with FC remains controversial. Today, the indications for surgery are mainly based on cosmetic and psychological factors. Like many other series [5–8], our cases demon-



**Fig. 4** Pulmonary function tests: percent vital capacity (%VC) of non-operated patients  $>80$  except in 3 cases,  $<80$  in about 60% of operated patients ( $FEV 1.0\%$  forced expiratory volume at 1 s)



**Fig. 5** Age at operation

**Table 2** Number of patients who answered questionnaire (OP period after operation, NOP period after first evaluation)

Period	OP	NOP
Less than 5 years	10	9
5–10 years	10	13
10–15 years	23	22
Over 15 years	17	0
Total	60	44

**Table 3** Results of questionnaire

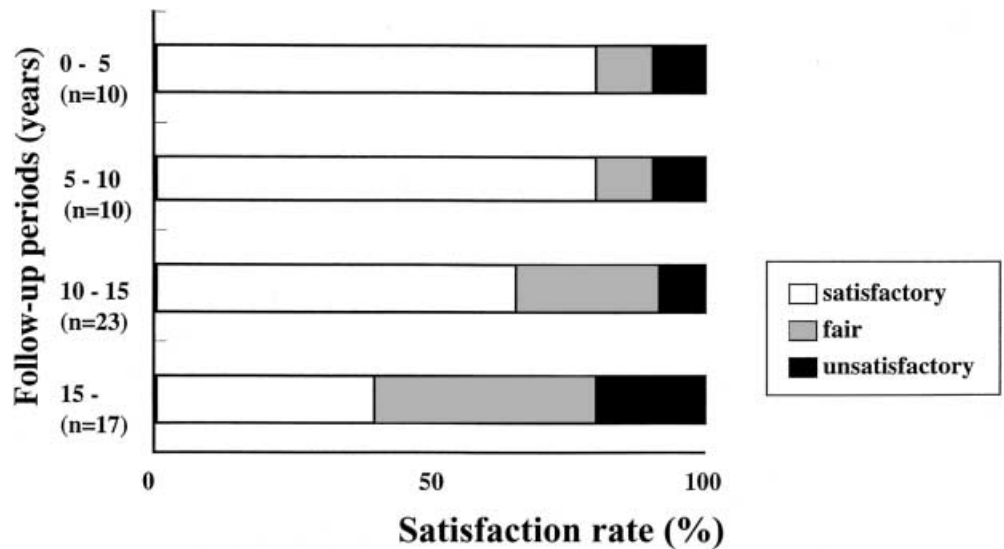
Answer	Operated	Non-operated
Satisfactory	37	2
Fair	14	24
Unsatisfactory	7	18
No answer	2	0
Total	60	44

strated that preoperative clinical symptoms were infrequent apart from embarrassment at having the chest exposed to view when swimming, being easily fatigued, and frequent episodes of upper RTI. To make an accurate and objective assessment of the severity of FC, it is important to identify patients who warrant surgical repair. We thus used the low %VC and FI F<sub>2</sub>I. However, even patients with an F<sub>2</sub>I of less than 0.2 sometimes underwent an operation because of parents' insistence for cosmetic reasons. The FI cannot always show the degree of asymmetry, and for that reason, we

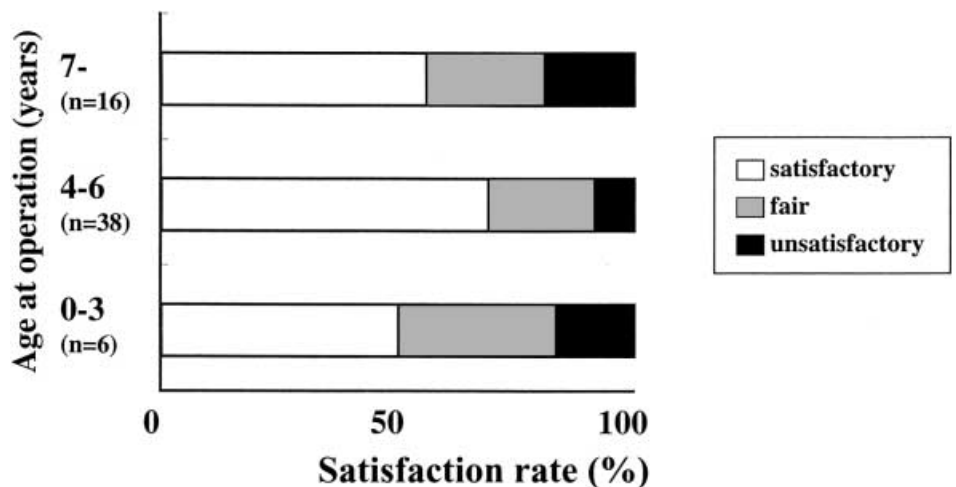
have used the CT index since 1987. The degree of both severity and asymmetry can be shown by the CT index, which demonstrated a significant difference between the OP and NOP groups [3]. In addition, a systolic ejection murmur is thought to be a good indication for operation [9]. We have included this parameter in our criteria for the past 6 years, however, no such cases have been identified so far. Exercise cardiac function tests using radionucleotide angiocardigraphy have also been reported to be useful in evaluating the cardiac function of patients with FC [10].

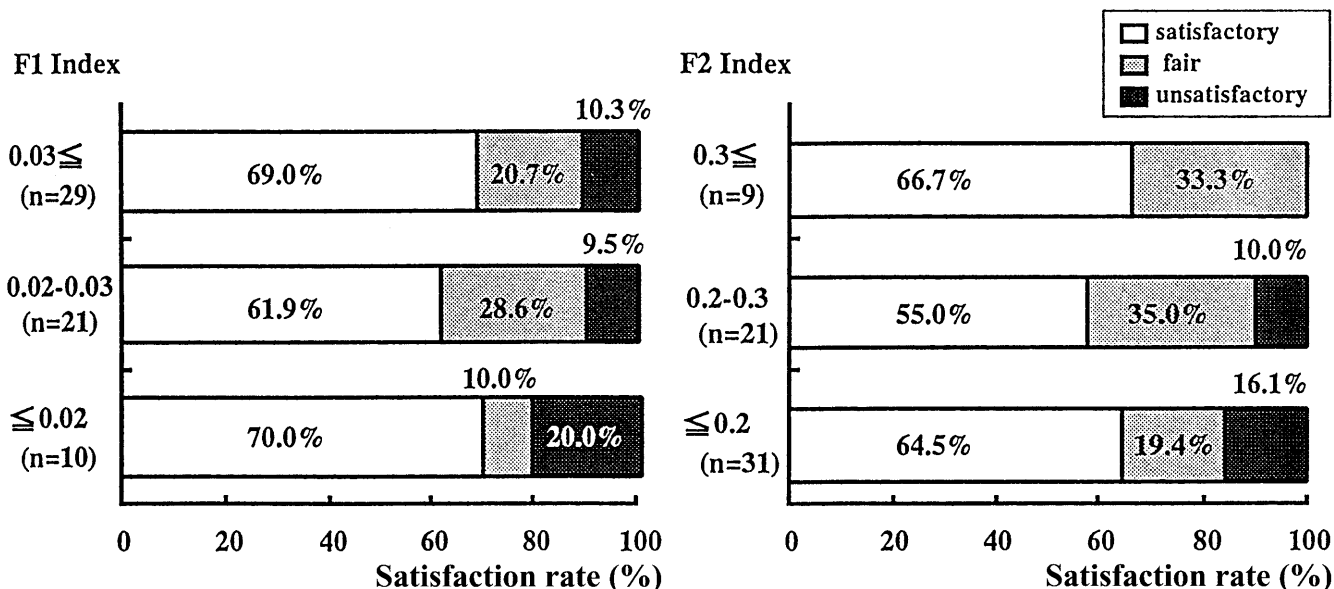
Various opinions exist about the value of early or late operation [6–9], but we believe that in patients less than 3 years of age this deformity sometimes disappears or becomes insignificant. Therefore, following a final decision for surgical correction when the patient reaches 3 years of age, we prefer to perform correction between the ages of 4 and 6 years in all children with a moderate to severe deformity. From the standpoint of the cosmetic result, patients operated upon between 4 and 6 years of age showed the highest satisfaction (Fig. 7).

**Fig. 6** Relationship between postoperative follow-up period and satisfaction



**Fig. 7** Relationship between age at operation and postoperative satisfaction





**Fig. 8** Relationship between preoperative funnel index and postoperative satisfaction: patients with  $F_1I < 0.02$  or  $F_2I < 0.2$  showed relatively less satisfaction while no patients with  $F_2I > 0.3$  were not satisfied

Fonkalsrud and Bustorff-Silva reported that the operation in adults was more difficult than in children, although the results were similar [11]. Haller et al. reported 12 children and teenagers in whom severe cardiorespiratory symptoms had developed due to failure of chest-wall growth after extensive operations at less than 4 years of age, and recommended delay in operative repair in small children until at least 6 to 8 years of age [12]. Operative correction in the very young age group (< 4 years) is thought to be harmful. Our satisfactory outcomes indicate that it is desirable to operate on children aged between 4 and 6 years. In this range, the operation is technically easier than in older patients and children have no need to be absent from school.

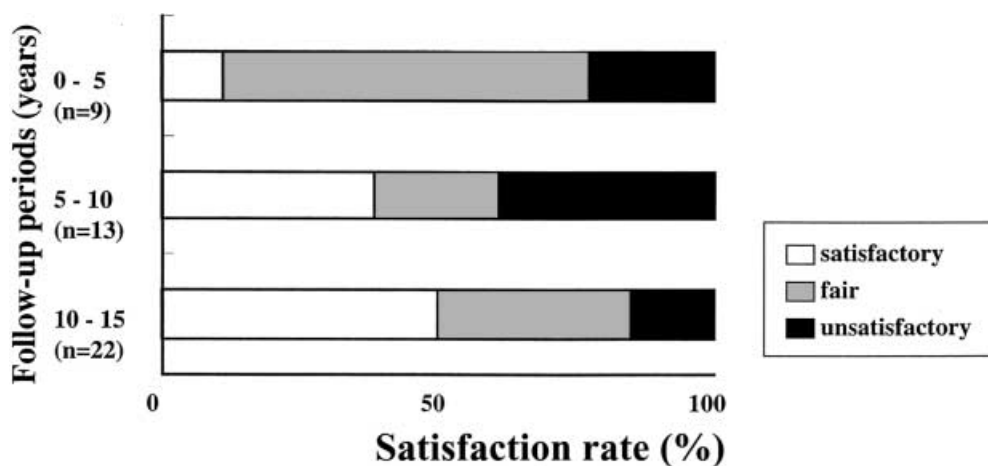
A variety of surgical techniques are available, and no one method is universally accepted as the optimal procedure [2, 5, 13–17]. Recently, endoscopic procedure has

been introduced [18, 19]. The choice of operative procedure depends solely on the surgeon's personal preference, and we have thus used sternoturnover for most of our cases because it is simple and easy to perform. As mentioned above, as we try to preserve the blood supply to the perichondrium and sternum, it is very rare to see patients with bone necrosis and/or recurrent depression of the sternum.

There was nearly 90% cosmetic satisfaction soon after the operation but as time elapsed the unsatisfactory rate increased. However, no patients reported any physical complaints after the operation. Most of the unsatisfied patients developed excessive scar formation, while the older patients did not remember the preoperative deformity since they were young at the time of operation. The more severe the preoperative deformity, the higher the satisfaction in the OP group. In the NOP group, as time elapsed, the satisfaction increased, probably because muscle or fat mass increased in the body and improved the appearance of the deformity.

NOP patients showed significantly less severe of FIs, compared with OP patients. Thus, patients without a

**Fig. 9** Relationship between follow-up period and satisfaction in non-operated patients: satisfaction was very low in first 5-year period of follow-up, tended to increase thereafter



severe deformity are not considered to be suitable for operative correction. However, there was no spontaneous improvement of the deformity in older patients; most NOP patients continued to show a cosmetic deformity and 26.7% had psychological stress. Therefore, in the future the indications for surgery may be extended if informed consent can be obtained even when the deformity is less severe.

In summary, this retrospective study confirms that our treatment strategy provides good results. An  $F_2I$  more than 0.2, %VC less than 80, and CT index less than 0.25 appear to be good objective criteria for operation. Based on the analysis of long-term follow-up, the indications for surgery are considered to be present in patients with severe deformity. However, in the interest of psychological development, these indications may be extended.

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