Quality of Life After Gastric Transposition for Oesophageal Atresia

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Background/Purpose: A small proportion of infants born with oesophageal atresia in which the gap between the 2 ends of the oesophagus is too great for an end-to-end anastomosis will require oesophageal replacement. Since 1981 the author's procedure of choice for oesophageal replacement has been gastric transposition. The long-term functional outcome appears to be satisfactory, but the quality of life of these patients has not been investigated formally. This report assesses the health-related quality of life (QOL) of 2 groups of patients born with oesophageal atresia who have undergone gastric transposition.

Methods: The study group comprised 28 patients aged 2 to 22 years who resided in England. Group 1 (n = 13), comprised patients who had undergone cervical oesophagostomy and gastrostomy without attempt at oesophageal anastomosis; group 2 (n = 15), comprised patients who had undergone previous attempts at reconstruction or replacement. QOL was assessed using modified versions of the Gastrointestinal Quality Of Life Index (GIQLI).

Results: QOL scores based on patients' responses showed no significant differences between the groups (124 *v* 119). However, the disease-specific symptom scores showed that patients in group 1 experienced fewer symptoms compared with those in group 2. Additionally, based on parental responses, patients in group 1 had higher QOL scores than those in group 2. QOL scores for patients aged 2 to 4 years (n = 5) did not differ between the groups (81 *v* 92, not significant).

Conclusions: The quality of life for patients with oesophageal atresia undergoing gastric transposition was generally unimpaired by any side effects of gastric transposition. Patients undergoing gastric transposition as a primary procedure experienced fewer disease-specific symptoms in the medium term compared with patients who had undergone previous unsuccessful attempts at reconstruction or replacement of their oesophagus.

INDEX WORDS: Oesophageal atresia, gastric transposition, quality of life, Gastrointestinal Quality of Life Index.

UNTIL RECENTLY, the 2 most widely used procedures for oesophageal replacement were colonic interposition and gastric tube oesophagoplasty, both of which are attended by high complication rates. Anastomotic leaks and strictures develop in around 30% and 20%, respectively, in colonic interposition, whereas comparable figures for gastric tube oesophagoplasty are 50% and 30%.[1] In the long term, redundancy and slow transit are problems that occur in colonic interpositions,[2] whereas Barratt's oesophagitis has been documented after gastric tube oesophagoplasty after a latent period of 10 to 20 years.[3]

Since 1981[4] our procedure of choice for oesophageal replacement has been gastric transposition.[1,5,6] The stomach has the advantage of having a rich blood supply, and the procedure has a lower incidence of anastomotic complications. It is relatively simple and involves a single anastomosis between the oesophagus and fundus of the stomach. Potential disadvantages are the space-occupying bulk of the stomach within the thorax impeding respiratory function, delayed gastric emptying secondary to the total vagotomy, and the possibility of gastro-oesophageal reflux.[7]

It would appear that the long-term functional outcome of gastric transposition in the majority of patients is satisfactory,[7] but there have been no in-depth assessments of the quality of life of this group of patients. This study explores the health-related quality of life of a group of patients after gastric transposition.

MATERIALS AND METHODS

Patients

The study group comprised 28 patients (18 boys and 10 girls) between 2 and 22 years of age (mean 12.99 ± 5.59 years), who were born with an oesophageal atresia (OA) that required gastric transposition (GT) for oesophageal replacement. The patients were divided into 2 groups: group 1 (n = 13) in whom no attempt was made to anastamose the oesophagus. Four infants had OA with distal fistula (wide gap), 6 had isolated atresia with a gap of 4 to 8 vertebra (long gap), and 3 had atresia with a proximal fistula. The 4 infants with a distal tracheo oesophageal fistula had the fistula divided at thoracotomy, but no attempt was made at oesophageal astomosis, and the 3 with a proximal fistula had that divided at the time of fashioning the cervical

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Table 1. Patients' Characteristics

	Group 1 (n = 13) Mean (SD)	Group 2 (n = 15) Mean (SD)
Age (yr)	13 (5)	13 (6)
Gestational age (wk)	36 (3.4)	35 (3)
Body weight (kg)	2.14 (.54)	2.39 (.89)
Number of patients (%) with associated anomalies	9 (69)	8 (53)
Age at GT (decimal yr)*	0.95 (0.6)	4.56 (5)
Time since GT (decimal yr)	12.26 (5.34)	8 (7)
Total No. of all operative procedures*	14 (15)	32 (25)
Related to oesophagus before transposition	1	19 (21)
Related to oesophagus after transposition	2 (2)	5 (7)
Body mass index: weight (kg)/length $(m)^2$ (z scores adjusted for age and sex)	−1.67 (0.98)	−1.70 (1.10)

*$P < .05$.

oesophagostomy. All 13 patients underwent cervical oesophagostomy and gastrostomy only before GT. Group 2, (n = 15) had undergone previous attempts at oesophageal repair including 6 replacements (4 colon, 2 Scharli type), 3 had recurrent fistula (one on 4 occasions), 4 underwent fundoplication (one on 3 occasions), one had a chronic leak and empyema after stricture resection, and one had an anastomotic disruption after delayed primary repair and suffered extreme failure to thrive.

Ethics

The study was approved by the Research Ethics Committee at our institution.

Design

Letters explaining the purpose of the study were sent to each patient and their family, and consent forms were given for each to sign if they were willing to participate in the study. The patients were assessed clinically (LS), and they and their parents were interviewed by the research psychologist (LL) using semistructured interviews adapted from previous studies of children born with surgically corrected anomalies.[8] Current psychological, emotional, and behavioural status was assessed, using data from the semistructured interviews and the Achenbach Child Behaviour Checklists. These results will be published in a separate report.

Quality-of-Life Measure

The subjective perception of well being or quality of life (QOL) of the patient was measured using a modification of the Gastrointestinal Quality of Life Index (GIQLI: English version) developed by Eypasch et al.[9] Our questionnaire included 21 (58%) of the original 36 GIQLI items (7 were extended to assess whether the symptom occurred during the day and also at night) and 9 new oesophageal-specific items. The questionnaire comprised items covering aspects of eating, disease-specific symptoms, and psychological, physical, and social functioning. Responses for each item, covering symptoms occurring in the "past 2 weeks," were scored as follows: 0 = all the time, 1 = most of the time, 2 = some of the time, 3 = a little of the time, 4 = never. The total possible score ranged from 0 to 144, the higher the score the better the quality of life. Internal consistency reliability estimates for the 36 items included in the total score of the modified GIQLI was high (Chronbach's alpha coefficient 0.93). Depending on the age of the patient, the questionnaire was completed by them and/or their parents. For the 5 patients aged 2 to 4 years, GIQLI items considered inappropriate for young children were excluded (total QOL score 0 to 120).

Statistical Analysis

For all continuous variables, group differences were analyzed using independent sample *t* tests, one-way analysis of variance, or the Mann-Whitney U test as appropriate. Chi-Square tests were used for categorical data. Body Mass Index standardized scores were calculated using the Child Growth Foundation diskette 1999. Agreements between patient and parent reports were assessed using statistical methods described by Bland and Altman[10] for measuring agreement between continuous variables.

RESULTS

The mean time since gastric transposition was 10.11 years (SD, 6.23) with a range of 0.84 to 19.53 years. Number of operations, including those performed in other institutions and those relating to procedures for associated anomalies, ranged from 2 to 91 (mean, 24 ± 22). Patients' characteristics within each group are summarized in Table 1, and those with associated anomalies in Table 2.

Four children (2 in each group) were below school age, and 8 patients (3 in group 1, 5 in group 2) had left school. One was at university, 3 were at college, 3 were employed, and one had recently given birth to a healthy boy.

Of those at school, 5 children were in a special unit or in special schools, and 4 required special needs within normal schools (9 of 17, 53%); in addition, 2 of the older patients had been in a special unit, and 2 had moderate

Table 2. Patients With Associated Anomalies

	Group 1 (n = 13)	Group 2 (n = 15)
None	4 (31%)	7 (47%)
Significant*	7 (54%)	6 (40%)
Cardiac	2	3
VATER	4	2
Sensory deficit	2 (15%)†	2 (13%)‡

*Includes conditions such as Fanconi anaemia (group 1), Trisomy 21 (group 2).

†One with bilateral anophthalmos and cerebral palsy; one congenitally blind.

‡Two with profound deafness.

Table 3. Quality of Life: Patient Responses

	Group 1 (n = 10) Mean (SD)	Group 2 (n = 9) Mean (SD)	95% CI for Difference
Aspects of eating (0-16)	12 (2)	12 (3)	−1.95, 2.64
Disease specific (0-84)	77 (6)	72 (10)	−3.34, 12.47
Psychological (0-20)	16 (2)	16 (3)	−2.66, 2.17
Physical/social (0-24)	19 (5)	19 (4)	−4.69, 4.86
Total (0-144)	124 (13)	119 (17)	−9.44, 18.95

learning difficulties (13 of 28, 46%). With one exception, all the older patients, including the young mother, were still living with their parent(s).

Quality-of-Life Outcomes

Patient response (n = 19, age 10-22 years). The total mean QOL score for the 19 patients who were able to complete the questionnaire was 122 (SD 14) with a range of 94 to 141. The disease-specific symptom subscale score was somewhat higher for patients in group 1 compared with patients in group 2 (Table 3). The number of patients reporting symptoms and side effects are shown in Tables 4 and 5.

There was no relationship between total QOL scores, associated anomalies, gender, and the number of operative procedures since GT or time since GT.

Parent responses: Patients aged 9 to 18 years (n = 17). The total mean score, based on parental perception, of these 17 patients was 115 (SD 21; range, 74 to 144). The patients in group 1 had higher scores than those in group 2 (Table 6).

For patients in Group 1, the fewer post-GT operative procedures the patient underwent the higher the parental rating of QOL ($r = .772$; $P = .025$). There was no association between these variables in Group 2 ($r = .082$; $P = .84$). There was no association in either group between parental assessment of QOL and their gender or length of time since GT.

Agreement between patients and parents. Thirteen parent and patient pairs, 7 pairs in group 1 and 6 pairs in group 2 completed the GIQLI questionnaires. In group 1, 6 of the 7 parents rated their child's QOL higher than the patients themselves, whereas in group 2, 5 of 6 patients perceived their QOL as being better than their parents' perceptions.

Parental response: Patients aged 2 to 4 years (n = 5). The overall QOL mean score for the young patients was 87 ± 20 with a range of 61 to 108; (group 1 [n = 2] 81 ± 6 [77, 85]; group 2 [n = 3] 92 ± 27 [61 to 108]).

DISCUSSION

In this study we have compared the health-related quality of life of 2 groups of patients who had undergone gastric transposition. Based on patient responses, the only difference between the groups was on disease-specific symptoms. Examination of the individual items showed that a lower proportion of patients for whom gastric transposition was the primary reconstructive surgical procedure (group 1) experienced dysphagia (30% *v* 67%) or pain after eating (20% *v* 33%), compared with patients who had undergone previous unsuccessful attempts at reconstruction or replacement of their oesophagus (group 2). Similarly, a smaller proportion of patients in group 1 had gastro-oesophageal reflux symptoms such as heartburn or regurgitation during the day or at night compared with those in group 2 (40% *v* 67%). Some breathlessness was experienced during the day by more than half the patients in each group, but breathlessness at night was more frequent in group 2. Differences between the groups were not related to the length of time since gastric transposition. These data were supported by the parents' perception of the QOL of their children. Based on parental responses, patients in group 1 experienced fewer disease-specific symptoms

Table 4. Patient Reported Symptoms: Eating Habits and Swallowing Ability

	Group 1 (n = 10) No. (%)	Group 2 (n = 9) No. (%)
Unrestricted diet	6 (60)	4 (44)
Size of meals similar to family/peers	4 (40)	2 (20)
Liquids with meals similar to family/peers	3 (30)	4 (44)
Slow eating	4 (40)	3 (33)
Dysphagia	3 (30)	6 (67)
Number meals per day	Median 5 (1-5)	Median 4.5 (2-6)

Table 5. Patient Reported Side Effects of Gastric Transposition

	Group 1 (n = 10) No. (%)	Group 2 (n = 9) No. (%)
Regurgitation	2 (20)	5 (56)
Heartburn	3 (30)	2 (22)
Vomiting	2 (20)	2 (22)
Halitosis	2 (20)	3 (33)
Dumping symptoms (diarrhoea, sweating, dizzy)	4 (40)	6 (67)
Respiratory tract infections	2 (20)	2 (22)
Breathlessness	6* (60)	5 (56)

*One (10%) breathless at night versus 3 (33%) in group 2.

Table 6. Quality of Life: Parent Responses

	Group 1 (n = 8) Mean (SD)	Group 2 (n = 9) Mean (SD)	95% CI for Difference	*P* Value
Aspects of eating (score 0-16)	13 (4)	10 (3)	.122, 7.30	.044
Disease specific (score 0-84)	76 (9)	64 (12)	1.10, 22.26	.033
Psychological (score 0-20)	17 (4)	15 (4)	−1.89, 5.59	.311
Physical/social (score 0-24)	21 (5)	15 (6)	−.094, 10.65	.054
Total (0-144)	127 (18)	104 (19)	3.37, 41.63	.024

such as dysphagia, dumping symptoms, and pain after eating. In addition, with the exception of psychological and physical/social symptoms, parents in group 1 perceived the health-related QOL of their children to be significantly better than parents of patients in group 2.

Based on parental reports the overall QOL of the young children, especially for those in group 2, was affected adversely by difficulties relating to all aspects of eating—their enjoyment of food, restrictions in types of food they could eat, and the amount they were eating. However, these problems are often reported about healthy children of this age. Examination of the disease-specific symptoms showed that one patient in each group (50% *v* 33%) experienced dysphagia, and one child in group 1 (50%) compared with 2 in group 2 (67%), experienced pain after eating. Both patients in group 1, and 1 patient in group 2 (33%), were reported to have gastro-oesophageal reflux symptoms. Similar proportions experienced some breathlessness during the day. Almost half of this small group of young children (44%) had associated anomalies, and this was an important factor affecting their lives.

The physical growth of the patients showed that, with one exception in each group, all the patients were below the 50th percentile for weight, but 5 in group 1 (41%) and 2 (12%) in group 2 were above the 50th percentile for height. When the standardized Body Mass Index (BMI; z scores), adjusted for age and gender, was calculated, all the patients had a BMI below zero, ranging from −0.10 to −3.91.

As far as we are aware, there are no other studies that have examined the medium-term outcome for patients with OA requiring gastric transposition. Ure et al[11] examined QOL more than 20 years after repair of OA using an earlier version of the GIQLI (scores 0 to 128). Eight of the 58 patients who underwent follow-up (aged 20 to 31 years) had long gap OA, and all had a colon interposition. These 8 patients had significantly worse specific symptoms scores compared with 50 primary anastomosis patients whose QOL scores were comparable with a group of healthy controls assessed by Eypasch et al (n = 150; aged 18 to 74 years).

Direct comparisons cannot be made with the validation study (phase III) of the GIQLI (patients aged 25 to 60 years) since we modified the questionnaire to include more oesophageal-specific symptoms and had excluded items not appropriate for our age group. However, the mean score of our patients in group 1, 124, is similar to the healthy individuals (125.8 [95% CI 121.5, 127.5]) whereas that of Group 2, 119, is outside the 95% CI.

With one exception, all the patients and the families in this study reported that they were extremely satisfied with the outcome after GT. The patients without debilitating conditions led normal lives, although they tended to be less socially and emotionally independent than their peers. Many enjoyed sporting activities. One teenage boy had achieved success in competitive sporting activities, and a teenage girl had been awarded a bronze Duke of Edinburgh award.

Patients with OA, for whom gastric transposition was the primary reconstructive surgical procedure, experienced fewer disease-specific symptoms in the medium term compared with patients who had undergone previous unsuccessful attempts at reconstruction or replacement of their oesophagus. The overall quality of life of both groups of patients, excluding the young children, was generally unimpaired by any side effects of gastric transposition.

REFERENCES

1. Spitz L: Gastric transposition for esophageal substitution in children. J Pediatr Surg 27:252-257, 1992

2. Rode H, Cywes S, Millar AJW, et al: Colonic oesophageal replacement in children—Functional results. Z Kinderchir 41:201-205, 1986

3. Lindahl H, Louhimo I, Virkola K: Colon interposition or gastric tube? Follow-up study of colon-esophagus and gastric tube-esophagus patients. J Pediatr Surg 18:58-63, 1983

4. Ahmed A, Spitz L: The outcome of colonic replacement of the esophagus in children. J Pediatr Surg 21:22-25, 1986

5. Spitz L: Gastric transposition via the mediastinal route for infants with long-gap esophageal atresia. J Pediatr Surg 19:149-154, 1984

6. Spitz L, Kiely E, Sparnon T: Gastric transposition for esophageal replacement in children. Ann Surg 206:69-73, 1987

7. Spitz L: Gastric transposition for oesophageal replacement in children. South Afr J Surg 39:9-13, 2001

8. Ludman L, Spitz L, Tsuji H, et al: Hirschsprung's disease: functional and psychological follow-up comparing total colonic and rectosigmoid aganglionosis. Arch Dis Child 86:348-351, 2002

9. Eypasch E, Williams JI, Wood-Dauphinee S, et al: Gastrointestinal Quality of Life Index: Development, validation and application of a new instrument. Br J Surg 82:216-222, 1995

10. Bland JM, Altman DG: Statistical methods for assessing agreement between two methods of clinical measurement. Lancet 1:307-310, 1986

11. Ure BM, Slany E, Eypasch EP, et al: Quality of life more than 20 years after repair of esophageal atresia. J Pediatr Surg 33:511-515, 1998

Discussion

From the Floor: If you use the same scale in normal children, what would be their score?

L. Ludman (response): The questionnaire was developed originally for patients between 18 and 86 years of age. I modified the questionnaire to exclude items that were inappropriate for our age group. The mean score of the patients in group 1, which was 124, was very similar to that of the normal controls in the validation study, which was 126. I would add, however, that it is very difficult to compare the patients in the study with normal healthy individuals, for many reasons.

A. Coran (Ann Arbor, Michigan): This is a very nice and important study because there have been many questions about primary versus delayed gastric transposition. How many of the patients in either group 1 or group 2 were pure atresias? Do you have any data about what the actual gap was between the 2 ends? We have done more than 60 gastric transpositions and have found that in the baby with pure esophageal atresia after 10 to 12 weeks of stretching of the upper pouch, a primary gastric transposition without esophagoscopy works much better. Do you have any patients in the latter category in your series?

L. Spitz (response): Of the 6 patients in the group, one had pure atresia (long gap), 3 had atresia with proximal fistula, and 4 had distal fistula (wide gap). The extent of the gap could not be measured because most patients were referred from other centres having already undergone surgery. We have not had occasion to perform a primary gastric transposition after the 6- to 12-weeks waiting period, because all were amenable to primary anastomosis.

A. Winthrop (Milwaukee, WI): We also, as Dr Coran mentioned, have been using this as our primary oesophageal reconstructive procedure in long gap or pure oesophageal atresia and have been working on patients at a much younger age than the median age of group 1. I would like to know what the difference was between group 1 and 2 in terms of the age at which they established full role feeding. One of the difficulties with this group of children is the length of time it takes to develop feeding skills, particularly when they are on tube feeding for a long period before establishing oesophageal continuity.

L. Ludman (response): I have no data on the delay in establishing oral feeding.