Journal of Pediatric Surgery 49 (2014) 1343–1346

Contents lists available at ScienceDirect

Journal of Pediatric Surgery

journal homepage: www.elsevier.com/locate/jpedsurg

Original Articles

Measured gap length and outcomes in oesophageal atresia

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ARTICLE INFO

Article history:
Received 14 October 2013
Received in revised form 17 March 2014
Accepted 30 March 2014

Key words:
Oesophageal atresia
Gap-length
Anastomotic leak
Strictures
Reflux
Fundoplication

ABSTRACT

Aims: Oesophageal atresia (OA) with or without tracheoesophageal fistula (TOF) is the most common congenital anomaly of the oesophagus. There is limited literature suggesting a linear relationship between increasing gap length and the incidence of all major complications. We sought to assess whether measured gap length at the time of surgery was related to outcomes in our patients.

Methods: All patients with a diagnosis of OA +/− TOF who underwent repair under a single surgeon between 1983 and 2012 were included. The length between the oesophageal pouches was measured at the time of surgery. Patients were then divided into three groups; short ≤1 cm, intermediate >1–≤2 cm and long >2–≤5 cm. Outcome measures were anastomotic leak, strictures requiring dilatation, gastrooesophageal reflux disease (GORD) and need for fundoplication.

Results: 122 patients were included in the study. The outcomes for patients with short ($n = 53$), intermediate ($n = 51$) and long gaps ($n = 18$) were as follows: anastomotic leak — 1.9%, 2%, 5.5% (**P = 0.66**), strictures requiring dilatation — 32%, 33%, 50% (**P = 0.67**), GORD — 51%, 59%, 72% (**P = 0.58**) and need for fundoplication — 11%, 20%, 44% (***P = 0.02**). There were no deaths related to the repair.

Conclusions: Measured gap length at the time of surgery did not have a linear relationship with leak or stricture rate. Our experience suggests that when primary repair is possible absolute gap length is irrelevant to the development of post-operative complications. There is however a significant increase in the need for fundoplication in those with a long gap.

1. Background

Oesophageal atresia and tracheoesophageal fistula (OA/TOF) can present as a spectrum of anomalies with the most common subtype being OA with a distal TOF (85% of cases). The Waterston classification first proposed in 1962, was used as a means of assessing short-term outcome and was based on three variables — birth weight, the presence of a congenital anomaly and pneumonia [1]. This was then revised in 1994 as the Spitz classification taking into account changes in diagnostic capabilities and management of other congenital anomalies [2]. More recently however, the gap length in oesophageal atresia has also been reported to be an independent prognostic risk factor [1,3]. We sought to assess whether measured gap length at the time of surgery was related to outcomes in our single-surgeon series of patients treated at Great Ormond Street Hospital.

2. Methods

All patients with a diagnosis of OA +/− TOF who underwent primary repair under the care of a single surgeon between 1983 and 2012 were included in the study. Exclusion criteria were patients with long gap atresia requiring oesophageal substitution and in those whom the gap length was unknown.

At operation, an initial rigid oesophagoscopy was performed to confirm diagnosis, assess length of the upper pouch and detect the presence of an upper pouch fistula. The standard approach for surgery was an open extra-pleural right thoracotomy via the 3rd, 4th or 5th intercostal space, depending on upper pouch length. More recently, four infants underwent thoracoscopic repair.

The length between the oesophageal pouches was formally measured at the time of surgery using a forceps and ruler. Dissecting forceps were inserted into the chest and adjusted to measure the gap between the apex of the upper pouch and the fistula into the trachea or the apex of the lower pouch. The forceps were then removed and re-inserted a few times to ensure that the position held was correct. The gap was then measured with a ruler.

Patients were divided into three groups based on this length; short ≤1 cm, intermediate >1–≤2 cm and long >2–≤5 cm. Demographic data collected included gestation, antenatal diagnosis of polyhydramnios and VACTERL association.

Oesophageal repair was performed using polypropylene (Prolene) sutures — usually 6/0 but 5/0 and 7/0 on occasion. Routine chest drainage has not been utilised for over 20 years. This was abandoned as we found leaks to be rare and when they do occur, the first chest drain usually does not work requiring a second drain to be inserted.

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http://dx.doi.org/10.1016/j.jpedsurg.2014.03.021
0022-3468/

Table 1
Patient demographics as stratified by Gap Length.

Patient demographics	Gap length (as measured in cm)		
	Short (n = 53)	Intermediate (n = 51)	Long (n = 18)
	≤1	>1–≤2	>2–≤5 cm
Mean gestation	38	38	37
Gender (M:F)	29:24	27:24	13:5
Antenatal diagnosis of polyhydramnios	21%	41%	55%
VACTERL association	13%	27%	11%
Pure oesophageal atresia	*n = 0*	*n = 0*	*n = 5*

Table 2
Complications as stratified by Gap Length.

Complications (%)	Gap length (as measured in cm)		
	Short (n = 53)	Intermediate (n = 51)	Long (n = 18)
	≤1	>1–≤2	>2–≤5 cm
Anastomotic leak	1 (1.9%)	1(2.0%)	1(5.5%)
Strictures requiring dilatation	17 (32%)	17 (33%)	9 (50%)
GORD	27 (51%)	30 (59%)	13 (72%)
Need for fundoplication*(P = 0.02)	6 (11%)	10 (20%)	8 (44%)

Outcome measures were anastomotic leak, strictures requiring dilatation, gastro-oesophageal reflux disease (GORD) and need for fundoplication. Patients requiring at least two dilatations for proven strictures were classified as having multiple dilatations. Objective evidence in the form of any pH or contrast studies assessing for reflux +/− strictures was also recorded.

Post-operative paralysis and ventilation were employed in all patients who had an anastomosis constructed under tension.

Feeding through the oral enteral route is commenced on the second post-operative day in those who are not ventilated. All those on a ventilator are fed by naso-gastric tube, commencing on the second day after operation. Transanastomotic tubes were used only when oral feeding was impossible due to ventilation or prematurity.

Routine contrast studies were not performed unless there was clinical suspicion of anastomotic leakage. After discharge, infants were then reviewed in the outpatient clinic within 6 weeks of discharge and then regularly thereafter as per clinical need. Patients were followed up to the age of 16 years for post-operative complications including reflux and strictures.

Statistical analysis for the data was performed using the Chi Square test. P-values <0.05 were considered to be statistically significant.

3. Results

122 patients who underwent primary repair of OA +/− TOF were included in the study. Six patients were excluded due to gap length not being recorded at the time of surgery. Two children died pre-operatively from cardiac failure and four patients died post-repair also from congenital cardiac disease.

Twelve patients had a delayed primary repair. Five patients had pure oesophageal atresia of which four had a delayed primary anastomosis. Details of their outcomes are described separately in Table 3.

Patient demographics as stratified by the gap length are shown in Table 1.

4. Complications

Table 2 illustrates the overall complication rate for the outcomes assessed in each of the three groups.

- **Anastomotic leak**

There were three anastomotic leaks (one in each group).

Patient 1 Born at 39 weeks of gestation, OA + TOF, open repair and tension-free primary anastomosis (gap length 0.9 cm). 36 h post-op clinically deteriorated with sepsis. Contrast imaging showed evidence of a leak. Immediate re-exploration was performed. The leak had occurred where a suture had cut through the lower pouch. The perforation was closed with 6/0 Prolene sutures and the baby was subsequently paralysed and ventilated for 5 days. Recovery was uneventful after this operation.

Patient 2 Born at 40 weeks of gestation, OA + TOF, open repair and primary anastomosis under moderate tension at day 1 of life (gap length 1.5 cm). Second day post-op noted to have saliva in the chest drain. The infant then clinically deteriorated and was taken to theatre after resuscitation. Severe disruption of the anastomosis was noted and in view of significant systemic illness the decision was made to fashion a cervical oesophagostomy and gastrostomy.

Patient 3 Born at 42 weeks of gestation, OA + TOF, open repair and primary anastomosis under severe tension at day 1 of life (gap length 5 cm, trans-anastomotic tube sited). Second day post-op noted to have saliva noted in the chest drain. Contrast imaging suggested a small leak and this was managed conservatively.

There was no statistical difference in the incidence of anastomotic leaks between the groups (P = 0.66).

- **Strictures requiring dilatation**

All patients clinically suspected of having a stricture were further investigated with either a contrast swallow or oesophagoscopy and subsequently underwent dilatation. The median time to first clinical suspicion of a stricture was 33 (1–465) weeks post-operatively. The incidence of strictures requiring at least two dilatations was 32%, 33% and 50% respectively in the three groups (P = 0.67).

- **Gastro-oesophageal reflux disease and need for fundoplication**

Once clinically suspected, GORD was initially investigated with a contrast swallow in most cases. A pH study was conducted in 33% of all suspected cases. The median time to clinical suspicion of reflux was 13 (1–775) weeks post-operatively.

The incidence of reflux proven objectively either on a contrast and/or pH study was 51%, 59% and 72% respectively across the three groups (P = 0.58). These patients were all managed initially with conservative treatment in the form of oral anti-reflux medication. Of those patients who went on to fail medical management, an anti-reflux fundoplication procedure was performed with increasing frequency across the three groups; 11%, 20% and 44% respectively (***P = 0.02**). The median age at fundoplication was 71 (6–696) weeks.

5. Discussion

Oesophageal atresia is a complex congenital anomaly with significant associated short and long-term morbidity. The pre-operative risks for this condition have traditionally been assessed using the Waterston, Montreal and Spitz classifications [4]. More recently however, gap length as an independent predictor of outcome has been postulated in a few studies in the literature. We sought to assess patient outcomes in our single-surgeon series extending from 1983 to date.

Upadhyaya et al. reported an increased incidence of all complications with increasing gap length in a series of 50 patients [1], with Brown and Tam also reporting a similar association in 66 consecutive patients [3]. The most significant complication associated with repair of oesophageal atresia is an anastomotic leak. There were three leaks in our series of 122 patients (overall 2.5%) (one in each group), which is lower than estimates of 7%–26% reported in large single-centre studies [5]. In the two studies above, leak rates of 31% (>3 cm gap) [3] and 80% [1] (>3.5 cm gap) were reported respectively. Risk factors which are considered to predispose to anastomotic leakage include gastro-oesophageal reflux, ischaemia at the anastomotic site,