Thoracoscopic repair of oesophageal atresia: Experience of 33 patients from two tertiary referral centres

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Abstract

Background: With advances in minimally invasive surgery, thoracoscopic repair of oesophageal atresia has become popular in many centres worldwide and indeed has been described as the pinnacle of neonatal surgery. Here, we report our experience in two tertiary referral centres.

Methods: Thoracoscopic technique was introduced in 2007. Thus, a retrospective review of all patients diagnosed with oesophageal atresia was carried out. Patients who had thoracoscopic repair were included, and those who had open repair due to co-morbidities were excluded. Patient demographics, operative data, complications, and associated anomalies were noted.

Results: A total of thirty-three patients underwent thoracoscopic repair during the time period. Thirty-one were successfully repaired thoracoscopically. Two patients had conversions due to intra-operative instability. The mean body weight of the neonates was 2.58 kg. The mean operative time was 146 min. Three patients suffered from minor anastomotic leaks, which healed on conservative management. Seven patients had anastomotic strictures, which responded successfully to endoscopic dilatation. Two patients died in the post-operative period due to pneumonia. One patient had a recurrent fistula 3 months after the primary repair, and he subsequently underwent a successful second repair.

Conclusions: In experienced hands, thoracoscopic repair of oesophageal atresia is at least as good as open surgery but with less surgical trauma. Standard of post-operative care contributes significantly to post-operative outcome. Thoracoscopic technique is now our preferred approach.

The use of minimally invasive surgery has seen major advances in recent years. In children, laparoscopy is now routinely performed and the technique has even translated to the management of neonatal conditions, such as oesophageal atresia.

Since Haight performed the first successful primary repair of esophageal atresia in 1941, the management of neonates with this condition has come a long way in terms of anaesthetic agents and monitoring, intensive care support, as well as surgical technique. In 1999, Lobe et al. reported the first successful thoracoscopic repair of oesophageal atresia [1]. Since then, there have only been a few case series published on this technique [2,3]. The theoretical advantages
of this procedure are excellent intra-operative visualization for both the surgeon and assistant, and also possibly decreased morbidity when compared to open thoracotomy. Although experience with this technique is growing worldwide, the clinical information in the literature is still sparse. Here we present the initial experience of our two centres to evaluate the safety and efficacy of oesophageal atresia repair using this approach.

1. Methods

Since July 2007, one unit (HKU) first adopted thorascopic approach for the treatment of babies diagnosed with oesophageal atresia. The other unit (Jiangxi) started the minimally invasive approach after the senior surgeon returned from his training in Hong Kong in 2009. The introduction of thorascoscopic repair was not taken lightly. Although the first report was published in 1999, it took us until 2007 when we felt that our minimally invasive skills had matured enough and after the publication of the multicentred study by Holcomb et al. in 2005 to finally convince us to embark upon this new endeavour [2]. With any new technique, the safety of our patients was always the top priority and many discussions with our anaesthetic colleagues were conducted well before the first case. We were also prepared to convert to open thoracotomy if there had been the slightest problem intra-operatively.

A retrospective review was carried out from July 2007 to Jan 2012. All babies with oesophageal atresia and tracheo-oesophageal fistula were treated thorascopically unless they were deemed unfit due to significant co-morbidities such as cyanotic heart disease. Patients with pure oesophageal atresia were also excluded because of the fact that these patients had long gap atresias most of the time and were deemed unsuitable for thorascoscopic repair. During the study period, all suitable cases for thorascoscopic repair in HK would involve either one of the two consultant surgeons who would perform thorascopscopic technique together with the remaining surgeon who initially had not learnt the technique. With experience, the remaining surgeon became competent to perform thorascoscopy subsequently. In Jiangxi, only one surgeon was in charge and he performed all the cases.

The demographic data, the operative time, operative outcome, and complications were recorded. Results were expressed in mean±standard error or range.

1.1. Operative technique

The procedure was carried out under general anaesthesia with tracheal intubation. All patients were placed in the 45° prone position to facilitate exposure of the oesophagus. Three ports were used for access to the thoracic cavity. A 5 mm camera port was inserted just below the tip of scapula at the posterior axillary line in the 5th intercostal space. 2 further ports (3 mm and 5 mm) were inserted above and below the first port in the mid-axillary line. Carbon dioxide insufflation at a pressure of 4 mm Hg was used to effect lung collapse. The azygous vein was identified and cauterized with diathermy. Distal fistula was then mobilized to as close to trachea as possible. The fistula was ligated close to the trachea with Hemolok® (Weck, USA), or by suture ligation. After the proximal end was identified, mobilized and opened, end-to-end anastomosis with the distal end was performed with 5-0 PDS (Ethicon, NJ, USA) either using extracorporeal or intracorporeal knot-tying techniques. A 6 Fr transanastomotic tube was placed routinely and a chest drain was placed at the end of the operation. These were removed after no leak was shown using contrast studies one week post-operatively.

2. Results

Since the start of the study period, there had been a total of 40 patients admitted with the diagnosis of oesophageal atresia. 4 patients were deemed unfit for thorascoscopic repair due to significant cardiac anomalies (Tetralogy of Fallot ×2; total anomalous pulmonary venous return ×1; coarctation of aorta ×1) and 2 patients had pure oesophageal atresias. Thorascoscopic repair have been attempted in 33 patients in our 2 centres. The mean gestational age at presentation was 38 weeks±6.5 days (range: 38 to 40±5 weeks). The mean birth weight was 2.58 kg (range: 1.4 to 3.3 kg). There were 21 males and 12 female.

All patients included this study had oesophageal atresia with distal tracheo-oesophageal fistula (Gross type C). Associated VACTERL anomalies were seen in 10 patients (Table 1). The mean gap distance between the two oesophageal ends was 1.76±0.3 cm. 26 of the 33 patients had short gap atresias (less than 1 vertebra). 7 patients of these had gap distance of the two oesophageal ends ≥ 3 cm (more than 3 vertebrae). All patients were operated on a mean of 3 days after birth (range: 1–11 days). Any delay in operation was due to the need of stabilization of the neonate.

Three patients who had long gap distance (3.2 cm; 3.5 cm; 3.5 cm) received staged procedure in which the first stage involved ligation and division of the fistula and approxima- tion of the two ends with sutures. The second stage of definitive anastomosis took place thorascopically one week later.

<table>
<thead>
<tr>
<th>Table 1</th>
<th>Coexisting congenital anomalies in the series of patients.</th>
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<tbody>
<tr>
<td>Associated VACTERL anomalies</td>
<td>Number of patients</td>
</tr>
<tr>
<td>Vertebral (Hemi-vertebra)</td>
<td>1</td>
</tr>
</tbody>
</table>
| Anal (Cloacal anomaly) | 1 *
| Cardiac (ASD; VSD) | 8 * |
| Renal | 0 |
| Limb | 1 |

* One patient had both cloacal anomaly and cardiac anomaly.
3. Discussion

Open repair of oesophageal atresia has been the standard operation for years. Since the introduction of thoracoscopic repair of oesophageal atresia in 1999, some large tertiary centers have reported their results with this method. Indeed, most of them have drawn the conclusion that the minimally invasive technique was a feasible alternative to open repair [1-4]. With the improvement in technique and intensive care support, the overall survival rate of patients with oesophageal atresia is now more than 90%. Most patients died because of prematurity and underlying cardiac anomaly [5]. In our combined series, we observed two post-operative mortalities, both were operated in Jiangxi. One patient was a 1.4 kg male with no underlying cardiac anomaly while the other was also a boy weighing 1.7 kg. He did have a small ventricular septal defect but was not in cardiac failure. Based on Spitz’s risk classification, one patient thus belonged to group I and the other one in group II. In a recent review by the unit in Great Ormond Street, the overall survival rates for these groups were 98% and 82% respectively [6]. Thus, one would not have predicted a high mortality for these two patients. As both were stable intra-operatively and the operative times were not any longer than the average, we could not attribute long operative time to the development of pneumonia. One possibility is that sub-clinical anastomotic leaks occurred in the post-operative period, as both patients had long gap atresias. The leaks would lead to pneumonia. Furthermore, poor oromotor skills in these infants and relatively less intensive nursing care in the mainland unit would contribute to the poorer outcomes. Taking these into account, our data would certainly point out the difference and the importance of the standard of post-operative care for these patients in developed and developing countries.

Early in our series, we found that nearly half of our patients developed anastomotic stricture post-operatively. In other reported case series, the stricture rates after thoracoscopic repair ranged from 14% to 45% [2-4, 6-8]. Although gastro-oesophageal reflux disease is known to be one of the factors for anastomotic stricture and the incidence of gastro-oesophageal reflux disease among oesophageal atresia patients was up to 48% [9, 10], we could not attribute our higher stricture rate to reflux disease alone, as only two patients had documented reflux from 24 pH studies and only one required fundoplication. The other factors which may explain the high anastomotic stricture rate could be excessive manipulation, placement of too many sutures and inadequate opening of the proximal stump due to the early experience. Indeed, since we changed to excising a short segment of the proximal pouch instead of simply opening a small hole, the recurrence of anastomotic stricture decreased dramatically to an overall of 22.6%. Despite the occurrence of strictures in our early patients, these are easily amenable to balloon dilatations, which are relatively non-invasive and safe [11]. For recurrent and resistant strictures, one case report suggested the use of high dose steroid to suppress the inflammation process around the stricture site, while a few others suggested the use of topical mitomycin C with success [12-14]. The recurrence of tracheo-oesophageal fistula in one of our patients was unexpected as this occurred 3 months after the primary surgery. Anastomotic tension could not be attributed to as there was only 1 mm gap distance between the two oesophageal ends. Conversely, the recurrent TOF might have been due to the proximity of the anastomosis to the fistula and we hypothesized that very minor post-operative leak might have played a part also. To help prevent recurrent fistulas, one recent report suggested not to divide the azygos vein as is the usual practice for most surgeons, as this might separate the oesophageal anastomosis from the site of fistula ligation [4]. However, whether this is really true will depend on a properly conducted prospective trial.

The intended benefit of thoracoscopic repair is the avoidance of musculoskeletal sequelae of thoracotomy. Co-

<table>
<thead>
<tr>
<th>Number of oesophageal dilatation required</th>
<th>Number of patients</th>
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<tbody>
<tr>
<td>0</td>
<td>17</td>
</tr>
<tr>
<td>1</td>
<td>7</td>
</tr>
<tr>
<td>2</td>
<td>3</td>
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<td>6</td>
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morbidities including rib fusion, chest-wall deformity have been reported [15]. In thoracoscopic repair, it can give an excellent cosmesis. The scar is almost invisible around 3 months after operation. Apart from this, we also have excellent visualization of anatomy during thoracoscopic repair. For open repair, the assistant struggles for the operative view, thereby making learning to perform the operation very difficult.

Nonetheless, performing thoracoscopic surgery is much more demanding than the open operation, both in terms of technique, as well as the need of excellent neonatal anaesthesia backup. As a result, surgeons who would like to carry out thoracoscopic repair should have not only advanced skills in minimally invasive surgery as a pre-requisite, but also surgeons who have previous thoracoscopic experience as mentor to attain a good result.

In conclusion, thoracoscopic repair of oesophageal atresia is a safe alternative procedure to open repair. It has become our preferred approach.

References