Title	Thoracoscopic repair of congenital diaphragmatic hernia: two centres' experience with 60 patients
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Citation	Pediatric Surgery International, 2014, v. 31, n. 2, p. 191-195
Issued Date	2014
URL	http://hdl.handle.net/10722/220763
Rights	The final publication is available at Springer via http://dx.doi.org/10.1007/s00383-014-3645-0

Introduction

With the marked improvements in endosurgical devices and technologies in the recent decade, minimal invasive surgery has gained much popularity across different surgical specialties. In specialized centres, laparoscopic surgery is now the standard approach for many elective conditions. The experience accumulated through laparoscopy has even been transferred to thoracoscopic surgeries, such as congenital diaphragmatic hernia repair.

Since the first report of congenital diaphragmatic hernia by Lazarus Riverius in 1679, this condition remains as a challenge for paediatric surgeons. Heidenhain performed the first successful repair via a midline laparotomy incision in 1902 [1]. The first transthoracic approach was reported 50 years later by Koop and Johnson in 1952, who suggested that this approach allowed closure of the defect under more direct vision [2]. However subcostal laparotomy has remained the approach of choice for most paediatric surgeons according to the Congenital Diaphragmatic Hernia Study Group 1997 report [3]. Advance in minimally invasive surgery in children has allowed for application of laparoscopy and thoracoscopy in various neonatal conditions, including congenital diaphragmatic hernia. The first successful laparoscopic repair of congenital diaphragmatic hernia in an infant was reported by van der Zee et al in 1995 [4]. In the same year Silen et al reported the first successful thoracoscopic congenital diaphragmatic hernia repair [5]. Since then the acceptance and ability to perform thoracoscopic repair of congenital diaphragmatic hernia within the field of paediatric surgery has showed substantial increase, to such an extent that it has become the routine practice in some large tertiary institutions [6]. The potential advantages of the thoracoscopic approach include better surgical field visualization, better wound cosmesis and the avoidance of thoracotomy associated musculoskeletal deformities.

Despite the growing interest in the field about this technique, only few series have been published in the literature, owing to the rarity of the disease [7-14]. In this study we aim to review the experience of our two centres on surgical techniques and post-operative outcomes.

Patients and methods

The University of Hong Kong first adopted the thoracoscopic approach for repair of congenital diaphragmatic hernia repair since early 2010. The other centre (Jiangxi) started the same operative approach two years later after the paediatric surgical team had received their training in Hong Kong.

A retrospective review of medical records of all patients who underwent surgery for congenital diaphragmatic hernia at these two centres between January 2010 and December 2013 was carried out. No patient was excluded from the study. All patients had chest radiography taken after birth. Computed tomography of thorax was only performed in patients with delayed or atypical presentation to confirm the diagnosis. Once diagnosed, patient was transferred to neonatal intensive care unit. Inhaled nitric oxide was applied only if there was evidence of severe pulmonary hypertension. No patient required extracorporeal membrane oxygenation in our series. Surgery was performed only when the patient was stable on conventional ventilation and weaned off from nitric oxide.

Patients' demographics, peri-operative and post-operative outcomes were collected.

Thoracoscopic repair was performed under general anaesthesia. Patient was placed in lateral decubitus position with the diseased side up. The ipsilateral arm was placed and fixed over the head. The monitor was placed at patient's foot side with operating surgeon standing over patient's head side. Single lung ventilation was not necessary.

Pneumothorax with insufflation of carbon dioxide at low flow (1 L/min) and low pressure (2-4 mmHg) was usually adequate for good exposure. Three 5mm ports were placed, one for the camera and two additional ports as working channels. The camera port was inserted in the third intercostal space at the mid-axillary line. The two working ports were inserted in the fourth intercostal space at the anterior and posterior axillary lines respectively.

The herniated organs were gently reduced into the abdominal cavity using blunt graspers. After reduction, the two diaphragmatic rims were closed with non-absorbable 2-0 Ethibond (Ethicon Inc, USA) interrupted sutures. In case of deficient posterior rim, the suture was tied around the ribs to provide stronger tension. For very large defects, prosthetic patch repair with Dualmesh (W.L. Gore & Assoc, Flagstaff, USA) was performed. No chest drain was inserted. All patients were kept intubated, sedated and paralyzed for at least three days post-operatively.

Statistical analysis of data was performed using SPSS (version 17; SPSS, Chicago, IL). Continuous variables were analyzed using Student's t test, ordinal variables were analyzed using Mann-Whitney U test and categorical variables were analyzed using chi-square test respectively. Data were presented as mean ± standard deviation and range. $p < 0.05$ was considered statistically significant.

Results

During the study period, 60 patients were admitted. The first thoracoscopic repair was carried out in 2010 and had become the standard procedure of both the centres since. There were 46 males and 14 females. The mean gestational age was 37.4 ± 5.6 days (range 34+2 to 39+6 weeks). The patients' age at the time of operation ranged from 1 day to 10 months, with 80% (n=48) of patients operated within the first week of life

commonly associated anomaly, with 7 of the patients in the series having atrial or

Table 1

(median 2 days) (Table 1). The mean body weight at operation was 3.03 ± 0.69 kg (range 2.1-6.0 kg). 50 patients (83%) suffered from left-sided diaphragmatic hernia. 8 patients were identified on antenatal ultrasound screening (all from Hong Kong centre). Other presenting symptoms included neonatal respiratory distress and feeding intolerance with vomiting in older infants (n=12). Cardiac anomalies were the most ventricular septal defects. One patient had coexisting high type anorectal malformation and one patient had significant congenital rib cage deformity. Patients from both centres shared similar demographics with no statistical significant difference (Table 2).

Table 2

During operation, hernia sac was identified in 15 patients (25%). Intestine was observed to herniate into the thoracic cavity in all patients. Other abdominal organs present in the hernia content included spleen in 41 patients, stomach in 22 patients and liver in 6 patients.

Only one patient required the use of prosthetic patch for repair (Table 3). All other diaphragmatic hernias were repaired primarily. Mean operative time was 88.5 ± 39.0 minutes (range 31-194 minutes). No patient required blood transfusion and none required chest drain insertion post-operatively. There was no conversion to open thoracotomy nor was there any intra-operative death observed.

Table 3

As a standard protocol all patient except one received deep sedation, full muscle paralysis and kept intubated for at least three days. The mean time of ventilation was 3.24 ± 0.44 days (range 0-6 days). The mean intensive care unit stay was 7.13 ± 3.86 days (range 3-16 days).

Five post-operative complications were identified. Two patients developed atelectasis and two suffered from pleural effusion after the operation. Surgical emphysema over

the thoracoscopic wounds was observed in one patient. All of these complications were managed conservatively. No bronchopneumonia or persistent pneumothorax had been reported. The mean hospitalization duration, including the period since admission for physiological stabilization, was 14.6 ± 5.25 days (range 5-37 days).

There was no mortality up until out-patient follow up, with the mean follow-up time being 22.7 ± 8.65 months (range 1 to 37 months). Five patients had recurrence of the diaphragmatic hernia, all located on the left side. One patient presented with symptoms of intestinal obstruction and was admitted as an emergency. The other four patients were asymptomatic and picked up as incidental findings by routine chest radiography on follow-up. One of the recurrences was noted two months post-operation and another one three months. The other three recurrences were identified more than one year after the operation during the annual follow-up. The two early recurrences were repaired thoracoscopically and no patch repair was required. The remaining three recurrences were re-operated with open laparotomy approach, two of which required patch application. These were all late presenters and redo thoracoscopic repair was deemed difficult with relatively limited working space without single lung ventilation. Moreover, one of these patients presented as an emergency with symptomatic intestinal obstruction.

Discussion

Our centre started to perform paediatric thoracoscopic surgery since 2002 but it was not until 2010 that we carried out our first thoracoscopic repair of diaphragmatic hernia. For any new procedure, it was performed only after we were confident that we had acquired adequate knowledge and skills about the condition. We started with relatively simple diagnostic or elective procedures and progress to more complicated

neonatal conditions after we accumulated ample thoracoscopic experience [15-17].

Before the use of thoracoscopic means for congenital diaphragmatic hernia repair, our centre had two cases of laparoscopic repair during the transitional phase. Indeed, the laparoscopic approach was even more disadvantageous in our experience. As previously pointed out by Arca et al, reducing the spleen back into the abdominal cavity was difficult laparoscopically [18]. As the neonatal spleen is both smooth and fragile, it is technically demanding to pull it back into the abdomen without damaging its surface or mesentery which inevitably results in bleeding. On the contrary, pushing it down into the abdominal cavity under the thoracoscopic view is much easier. In addition, if the spleen is reduced as the last organ, it can actually act as a shield to cover up the intestines, which greatly facilitate the process of suturing. Another disadvantage about laparoscopic repair is the inferior surgical exposure. During the laparoscopic suturing of the diaphragm, the abdominal contents, mainly the intestine, will be obscuring the view of the hernia defect from time to time, unless an additional port is inserted for traction purpose. For thoracoscopic approach, the diaphragmatic defect is under clear visualization after all the hernia content has been reduced, then the sutures can be placed easily. This may explain the higher conversion rate of laparoscopic approach observed [11].

Thoracoscopic repair has the obvious advantage of wound cosmesis over laparotomy associated scar and thoracotomy related rib cage deformities [19]. Furthermore, without division of muscle means that post-operative pain is significantly reduced and there is earlier recovery of respiratory function as neonates are obligatory abdominal muscle breathers [20].

The high end tidal carbon dioxide content and intra-operative desaturation previously observed in some series can be prevented by the use of low pressure and low flow

carbon dioxide insufflation system, now a common practice among most authors [14, 18]. Single lung ventilation is not necessary as compared with other thoracic conditions. The pneumothorax created also facilitates the process of reduction of hernia contents into the abdomen [18].

The safety and feasibility of thoracoscopic repair was well demonstrated in this series with the no intra-operative complication and one hundred percent survival, when compared to around eighty percent in some study with open approach [21]. To our surprise no conversion to open thoracotomy or laparotomy was required. The mean operative time in our series was 88.5 minutes, which was similar to that reported by Liem et al (75 minutes) [14], Nam et al (118 minutes) [22] and Tanaka et al (194 minutes) [23]. In fact the operative time was the same if not faster than that of the open approach group in these comparative studies (88.3 and 161 minutes, Nam et al and Tanaka et al respectively [22, 23]). Chest tube drainage has been advocated by Liem et al in one of the largest published series in order to prevent pleural effusion [14], but we only encountered two such complication (3.3%) which later subsided spontaneously.

In the five patients with recurrence, none of them had large defect that required synthetic patch repair during the first operation. In retrospect, the posterior diaphragmatic rim in four of these patients were deficient albeit the abundant anterior muscle lip, resulting in the suture over the posterior pleura and muscle being too shallow and cutting through of stiches. We had since learnt from this and would place the sutures to include the ribs over the posterior side in similar situation. Thus far the result of the subsequent cases with deficient posterior rim had been satisfactory. Nonetheless surgeons should not hesitate to apply prosthetic patch repair in case of large hernia defect or excessive tension over the sutures.

In conclusion, thoracoscopic repair of congenital diaphragmatic hernia can be performed safely in experienced centre. It can be considered to be an effective alternative to open approach.

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