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Early recurrence of congenital diaphragmatic hernia is higher after thoracoscopic than open repair: a single institutional study

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Abstract

Introduction—Experience in thoracoscopic congenital diaphragmatic hernia (CDH) repair has expanded, yet efficacy equal to that of open repair has not been demonstrated. In spite of reports suggesting higher recurrent hernia rates after thoracoscopic repair, this approach has widely been adopted into practice. We report a large, single institutional experience with thoracoscopic CDH repair with special attention to recurrent hernia rates.

Methods—We reviewed the records of neonates with unilateral CDH repaired between January 2006 and February 2010 at Morgan Stanley Children's Hospital. Completely thoracoscopic repairs were compared to open repairs of the same period. In addition, successful thoracoscopic repairs were compared with thoracoscopic repairs that developed recurrence. Data were analyzed by Mann-Whitney *U* and Fisher exact tests.

Results—Thirty-five neonates underwent attempted thoracoscopic repair, with 26 completed. Concurrently, 19 initially open CDH repairs were performed. Preoperatively, patients in the open repair group required more ventilatory support than the thoracoscopic group. Recurrence was higher after thoracoscopic repair (23% vs 0%; $P = .032$). In comparing successful thoracoscopic repairs to those with recurrence, none of the factors analyzed were predictive of recurrence.

Conclusions—Early recurrence of hernia is higher in thoracoscopic CDH repairs than in open repairs. Technical factors and a steep learning curve for thoracoscopy may account for the higher recurrence rates, but not patient severity of illness. In an already-tenuous patient population, performing the repair thoracoscopically with a higher risk of recurrence may not be advantageous.

Keywords

Congenital diaphragmatic hernia; Thoracoscopic repair; Recurrence

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Experience in thoracoscopic repair of congenital diaphragmatic hernia (CDH) has expanded in recent years [1–6]. Proponents of the operation argue not only that is it safe and feasible but also that it offers fewer postoperative ventilator days and less need for narcotics [4]. However, efficacy has not yet been proven. For thoracoscopic CDH repair, recurrent hernia has been reported to be between 0% and 25%, with most authors noting it to be higher than 15% [1–5,7,8]. Although no single-institution, published report has noted the recurrence rate to be statistically significant, the incidence is higher than the 10% recurrence rate after open repair that was reported in the largest series published to date [9].

We present a large single institutional experience with neonatal thoracoscopic CDH repair. The aim of our study was to examine its efficacy. We compared the thoracoscopic patients to our open CDH experience during the same period. In addition, we compared patients who underwent successful thoracoscopic CDH repairs to those patients who recurred to help identify factors, which may make patients prone to recur.

1. Methods

Study approval was granted by the Columbia University Institutional Review Board (IRB-AAAE7708). We reviewed the records of all patients diagnosed with a CDH between January 2006 and February 2010. This time frame was selected because it was the era when we performed thoracoscopic repairs. All repairs were completed at Morgan Stanley Children's Hospital—Columbia University Medical Center, New York, NY. Patients with lethal pulmonary hypoplasia, bilateral hernias, Morgagni hernias, significant cardiac disease, and whose repair was performed outside the neonatal period (>30 days) were excluded. In addition, patients supported by extracorporeal membrane oxygenation (ECMO) were excluded. At our institution, the diaphragmatic defect is repaired while on ECMO, and this was deemed a contraindication to thoracoscopic repair.

Neonates with CDH were managed using a strategy of pressure-limited, lung-sparing ventilation, allowing for permissive hypercapnea and spontaneous respiration [10]. Muscle relaxants were avoided preoperatively and postoperatively. Patients unresponsive to low rate conventional ventilation were transitioned initially to high-frequency positive pressure ventilation (100 breaths per minute). If hypoxia persisted, they were ultimately placed onto a high-frequency oscillator. Surgery was delayed until ventilator support had been minimized and when the preductal to postductal saturation gradient had decreased, indicating improving pulmonary vascular resistance.

1.1. Technique for thoracoscopic repair

After obtaining informed consent from parents, the patient is placed in the lateral decubitus position with the affected side elevated. The back of the patient is positioned on the edge of the table, which allows 1 operating hand the most degree of movement. The patient is placed on a stack of towels with the position of the head slightly lower on a cushion. This prevents the other operating hand from contacting the endotracheal tube as well as allowing for more movement. An axillary roll is used. Respirations are held, and a 5-mm port is placed subscapularly. A pneumothorax is created by gradual insufflation of carbon dioxide to a pressure of 5 mm Hg. A 4-mm, 30° telescope is inserted through the subscapular port. Two

additional 5-mm working ports are placed medially and laterally under direct vision. Viscera are gently reduced into the abdomen through the defect using endopeanut cotton tip instruments. If reduction is difficult, a fourth port is placed to facilitate this. If a hernia sac is identified, it is excised using hook cautery. After excision of the hernia sac is completed, the carbon dioxide insufflation often aids in the reduction of the viscera. Repair is performed using 2-0 braided polyester suture. Knots are intracorporeally tied or secured by using the Ti-Knot device (LSI Solutions, Victor, NY). The posterolateral portion of the defect is closed by passing nonabsorbable sutures around the ribs and tying them extracorporeally. In cases where there is excessive tension on the diaphragmatic closure, a 1-mm-thick polytetraflouroethylene patch is placed thoracoscopically. The patch is marked before placing it into the thorax for easy orientation. A thoracostomy tube is not placed after completion of the repair.

1.2. Technique for open repair

The patient is placed supine on the operating table. Appropriate monitors and padding are placed. An ispilateral-subcostal incision is made, and dissection is carried down until the abdomen is entered. The translocated viscera are gently reduced into the abdomen. If present, a hernia sac is excised. Repair of the diaphragmatic defect is performed using nonabsorbable suture. In cases where there is excessive tension on the diaphragmatic closure, a 1-mm-thick polytetraflouroethylene patch is placed. The abdomen is then closed in the standard fashion.

1.3. Data collection and statistical analysis

The records of the patients who underwent a thoracoscopic and open CDH repair were reviewed for sex, side of hernia, birth weight, gestational age, lung-to-head ratio (LHR) value, APGAR score (1 and 5 minutes), site of liver on prenatal imaging, preoperative maximum ventilator, age at repair, day of repair ventilator settings, and arterial blood gas. In addition, conversion to open repair, operative time, type of repair (patch vs primary), maximum postoperative ventilator requirement, postoperative ventilator days, length of stay, recurrence, and survival were studied. Patients are followed post discharge at a multidisciplinary CDH clinic.

Completed thoracoscopic repairs were compared with patients who had an initially open procedure during the same period. In addition, successful thoracoscopic repairs were compared with thoracoscopic repairs with recurrence. Continuous variables were reported as medians with ranges and compared using Mann-Whitney *U* tests. Differences in categorical variables were assessed using χ^2 and Fisher exact tests. Statistical significance was assumed at $P < .05$. Data analyses were performed using the Statistical Package for the Social Sciences (SPSS 15.0; SPSS, Chicago, IL).

2. Results

We identified 89 patients diagnosed with a CDH between January 2006 and February 2010 at Morgan Stanley Children’s Hospital of New York Presbyterian. Patients supported by ECMO (n = 17), lethal pulmonary hypoplasia (n = 9), bilateral hernia (n = 1), Morgagni

hernias (n = 2), significant congenital cardiac disease other than ventricular septal defect (n = 3), and age at repair greater than 30 days (n = 3) were excluded.

Thirty-five neonates underwent attempted thoracoscopic CDH repair. Twenty-six patients (74%) were successfully completed thoracoscopically. Of the 9 patients converted to open, 7 were owing to a large defect and the need for a patch repair. All of these occurred early, in the first 18 months of our experience. In the remaining 2 patients, 1 was converted to open because of persistent desaturation and the other because of early ischemic changes of the reduced bowel.

2.1. Thoracoscopic repair compared to open repair

Comparisons of prenatal variables between the thoracoscopic group (TG; n = 26) and open group (OG; n = 19) are presented in Table 1. The TG had a similar number of male infants as the OG (53.8% vs 42.1%; *P* = .55). There was an equal number of right-sided hernias (TG, 11.5%; OG, 15.8%; *P* = .686). Birth weight (3.29 vs 3.25 kg; *P* = .535) and gestational age (39 vs 38 weeks; *P* = .102) were similar in the TG vs OG. APGAR scores at 1 and 5 minutes were similar in both groups. Lung-to-head ratio value (1.4 vs 1.3; *P* = .58) and percentage of patients with liver in the chest on prenatal imaging (56.3% vs 70%; *P* = .683) were not different between the TG and OG.

A higher number of patients required high-frequency or oscillatory ventilation preoperatively in the OG than the TG (63.2% vs 19.2%; *P* = .005) (Table 2). Age on the day of repair was not different between TG and OG (3 vs 4 days; = .925). There was no difference in arterial blood gas values or preductal oxygen saturation on the day of repair between the 2 groups (Table 2). In comparing ventilator settings on the day of repair, the TG required a lower rate of ventilation (30 vs 40 breaths per minute; *P* = .018) and lower fraction of inspired oxygen (FIO_2) (0.21 vs 0.26; = .003). There was no difference in peak inspiratory pressure (PIP) or positive end expiratory pressure (PEEP) settings.

Table 3 compares operative and postoperative data between the TG and OG. Operative time was significantly longer in the TG (148.5 vs 113.5 minutes; *P* = .012). There was a lower frequency of patch repairs in the TG (46.2% vs 84.2%; *P* = .013). There was no difference in need for high-frequency or oscillatory ventilation postoperatively (TG, 26.9% vs OG, 42.1%; *P* = .347). Postoperative ventilator days (1.7 vs 4.5 days; *P* = .001) and length of stay (21 vs 41 days; *P* = .007) were significantly shorter in the TG.

Six patients recurred in the TG (23.1%) compared to none in the OG (*P* = .032). The location of these recurrences was variable. Three were posterolateral, 2 were posteromedial, and 1 was anterolateral on the diaphragm. In the TG, 3 (25%) of 12 infants with patch repairs and 3 (21%) of 14 infants with primary repairs developed hernia recurrence. Four TG recurrences presented after initial discharge. There was no difference in survival. Median follow-up time was 14 months for both groups. The number of cases and recurrences by surgeon for both the thoracoscopic and OG is listed in Table 4.

2.2. Recurrence-related factors

Comparison of variables between the successful TG and recurrent TG is presented in Table 5. Of the 26 completed thoracoscopic repairs, there were 6 recurrences. No appreciable differences were found between the 2 groups.

3. Discussion

Since the first published report of thoracoscopic CDH repair in 2003, there have been over 200 cases published in the literature [11]. This minimally invasive technique has the potential advantages of improved visualization, less need for narcotic postoperatively, and shorter length of intubation [4]. Despite these perceived advantages, one of the most important outcomes for a CDH is a secure, long-lasting repair. Although not reaching statistical significance, there has been a reported higher recurrent hernia rate after thoracoscopic CDH repair seen in other institutional reports [1–5,7,8]. Recently, a meta-analysis reported a statistically higher recurrence rate in the thoracoscopic repair group [12]. However, this was a heterogeneous patient population treated with varying ventilator strategies at several institutions. Our institutional approach to medically and surgically managing neonatal CDH has remained constant since 1982. This allowed us to investigate a homogenous patient population to compare thoracoscopic CDH repair and open repair.

We attempted 35 neonatal thoracoscopic CDH repairs between January 2006 and February 2010. Twenty-six patients were successfully completed. During that same time, we performed 19 initially open neonatal CDH repairs, exclusive of our ECMO population. To examine the efficacy of thoracoscopic CDH repair, we compared the 26 completed thoracoscopic repairs to the 19 initially open repairs. There was no difference between the groups about sex, hernia side, birth weight, gestational age, APGAR score, LHR value, or liver in chest. This suggests that they were a similar patient population at birth.

A significantly larger percentage of patients in the open repair group required high-frequency or oscillatory ventilation preoperatively. This suggests that the open patients were more ill. Despite this, the 2 groups were similar in the amount of days until hernia repair. In addition, arterial blood gas analysis and ventilator settings on the day of repair were essentially similar. The small differences in FIO_2 and respiratory rate likely have no clinical significance. Taken together, this demonstrates that although the open patients may have been somewhat less stable initially, the 2 groups were equally stable on the day of repair.

Operative time was longer in the TG. This has been demonstrated in other series as well [5]. The number of patients requiring high-frequency ventilation or the oscillator postoperatively was similar in both groups. Postoperative ventilator days and length of stay were higher in the OG. These data likely are reflective of the open patients being more ill than the thoracoscopic patients.

Recurrence rate was significantly higher in the TG (23%) than the OG (0%). As was demonstrated, the TG did not appear to be as ill as the OG. The TG also did not have factors that have been shown to independently predict recurrence: longer postoperative length of

stay and abdominal wall patch use [9]. Thus, the higher recurrence rate after thoracoscopic repair cannot be attributed to patient severity of illness.

Given the higher recurrence rate, we compared our recurrent thoracoscopic hernia repairs to our successful thoracoscopic hernia repairs. This was performed to see if certain factors can predict which patients were more likely to recur and, thus, should not be offered thoracoscopic repair. There were no significant differences in perinatal variables between the 2 groups, but the number of patients is small (Table 5). Given these results, we currently have no predictive models of thoracoscopic recurrence.

The cause of the higher recurrence rate is not known, but certain technical factors may contribute to it. As in other minimally invasive operations, the operative field is magnified. This may lead to overestimating the space between sutures on the diaphragm or patch. When one of our thoracoscopic recurrences was explored, bowel was found herniating between sutures on the diaphragm. A second explanation may be the tension placed on the suture. Diaphragm muscle is prone to tearing, and the correct amount of tension needs to be applied when tying a suture during the repair. Tension is difficult to disperse among several sutures when tying knots thoracoscopically, and an excessive amount may be placed on each suture during the repair. Another recurrence occurred when the suture tore through the diaphragm. A third explanation may be inadequate mobilization of the rim of diaphragm. During the open procedure, the diaphragm is routinely “unfurled,” exposing a larger area of posterior diaphragm to allow suture placement for repair. This may not be thoroughly performed during thoracoscopic repair because of limited visibility from the chest. It is possible that this also contributes to inadequate bites of tissue or increased tension on the repair.

Another possible explanation for the higher recurrence rate is the learning curve of this new operation. However, recurrences occurred throughout the period, not just early in our experience. It is possible that the learning curve is much steeper than previously anticipated. Given that CDH occurs in 1 in 2000 to 4000 births and that 25% of patients require ECMO, it may take a much longer time to accrue an adequate number of patients to become proficient in this operation [13]. In addition, our multidisciplinary CDH follow-up clinic captures even asymptomatic recurrences by radiograph or echocardiography. It is possible that lower recurrence rates reported in other studies may be attributable to less vigilant follow-up in the absence of symptoms.

Limitations of our study are its retrospective, nonrandomized nature and small number of patients. In addition, there was selection bias because the type of operation was at the individual surgeon’s discretion.

Minimally invasive surgery offers certain advantages over open surgery, such as quicker recovery and improved cosmesis. However, it should provide equal outcomes to the standard operation. We have seen in our 4-year experience, early recurrence rates are higher in thoracoscopic CDH repair than open CDH repair. We have also demonstrated that it cannot be completely explained by patient severity of illness. In an already-tenuous patient population of CDH, performing the operation thoracoscopically appears to be associated with a higher risk of early recurrence and, therefore, may not be advantageous.

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Table 1

Prenatal variables (thoracoscopic vs open)

	Thoracoscopic (n = 26)	Open (n = 19)	*P*
Male sex	53.8%	42.1%	.55
Right-sided hernias	11.5%	15.8%	.686
Birth weight (kg)	3.29 (1.3–4.2)	3.25 (1.79–3.95)	.535
Gestational age (wk)	39 (34–41)	38 (36–41)	.102
LHR value	1.4 (1–2.6)	1.3 (0.8–2.4)	.58
APGAR score, 1 min	7 (2–9)	5 (1–9)	.063
APGAR score, 5 min	8.5 (5–9)	8 (4–9)	.172
Liver up	56.3%	70%	.683

kg, kilograms; LHR, lung to head ratio.

Table 2

Preoperative data (thoracoscopic vs open)

	Thoracoscopic (n = 26)	Open (n = 19)	*P*
Preoperative HF/HFO vent requirement	19.2%	63.2%	.005
Age at repair (d)	3 (2–22)	4 (1–10)	.925
pH	7.375 (7.33–7.45)	7.38 (7.3–7.51)	.917
Pco_2	42.5 (30–51)	42 (29–55)	.147
Po_2	70 (46–121)	64 (39–109)	.147
HCO_3	24 (19–34)	25 (21–32)	.898
Preductal oxygen saturation (%)	97 (90–100)	97 (83–100)	.57
Ventilator respiratory rate (breaths per min)	30 (12–100)	40 (12–100)	.018
PIP	19 (14–22)	20 (16–23)	.451
PEEP	5 (1–5)	5 (1–5)	.133
Fio_2	0.21 (0.21–0.35)	0.26 (0.21–0.4)	.003

HF vent indicates high-frequency ventilator (100 breaths per minute); HFO vent, high-frequency oscillatory ventilation; PIP, peak inspiratory pressure; PEEP, positive end expiratory pressure; FI02, fraction of inspired oxygen.

Table 3

Operative and postoperative data (thoracoscopic vs open)

	Thoracoscopic (n = 26)	Open (n = 19)	*P*
Operative time (min)	148.5 (74–273)	113.5 (68–220)	.012
Patch repairs	46.2%	84.2%	.013
Postoperative HF/HFO vent requirement	26.9%	42.1%	.347
Postoperative ventilator days	1.7 (0.75–17.25)	4.5 (1.25–22)	.001
Length of stay (d)	21 (8–128)	41 (14–101)	.007
Recurrence	23.1%	0%	.032
Survival	100%	94.7%	.422
Follow-up (mo)	14 (1–35)	14 (1–47)	.59

Min, minutes; HF vent, high frequency ventilator (100 breaths per minute); HFO vent, high frequency oscillatory ventilation.

Table 4

Cases by surgeon

Surgeon	Thoracoscopic (recurrences/total)	Open
1	3/14	1
2	2/4	1
3	0/3	1
4	1/2	5
5	0/1	7
6	0/1	3
7	0/1	0
8	0	1

Table 5

Thoracoscopic recurrence comparison

	Recurrence (n = 6)	No recurrence (n = 20)	*P*
Male sex	50%	55%	1.0
Right-sided hernias	0%	15%	1.0
Birth weight (kg)	3.59 (3.35–4.22)	3.21 (1.30–4.00)	.003
Gestational age (wk)	38.5 (37–41)	39 (34–41)	.533
LHR value	1.1 (1.01–2.00)	1.45 (1.00–2.60)	.234
APGAR, 1 min	6.5 (3–8)	7 (2–9)	.744
APGAR, 5 min	9 (8–9)	8 (5–9)	.295
Liver up	50%	58%	1.0
Preoperative HF/HFO vent requirement	0%	25%	.298
Age at repair (d)	3 (2–4)	4 (2–22)	.176
pH	7.36 (7.34–7.44)	7.395 (7.33–7.47)	.268
Pco_2	42.5 (39.0–50.0)	42 (30.0–51.0)	.700
Po_2	78.5 (56–91)	68.5 (46–121)	.176
HCO_3	24 (23–34)	24 (19–31)	.882
Preductal oxygen saturation (%)	96.5 (93.0–99.0)	97 (90.0–100.0)	.744
Ventilator respiratory rate (breaths per min)	34.5 (16–40)	25 (12–100)	.555
PIP	19 (14–20)	19 (15–22)	.877
PEEP	5 (4–5)	5 (1–5)	1.0
Fio_2	0.22 (0.21–0.30)	0.21 (0.21–0.35)	.882
Operative time (min)	159 (74–236)	143 (90–273)	.882
Patch repairs	50%	45%	1.0
Postoperative HF/HFO vent requirement	17%	30%	1.0
Postoperative ventilator days	2 (0.75–3.75)	1.13 (0.75–17.25)	.494
Length of stay (d)	37 (20–70)	20 (8–128)	.069
Survival	100%	100%	1.0

kg, kilograms; LHR, lung to head ratio; Min, minutes; HF vent, high frequency ventilator (100 breaths per minute); HFO vent, high frequency oscillatory ventilation; PIP, peak inspiratory pressure; PEEP, positive end expiratory pressure; FI02, fraction of inspired oxygen.