

TITLE: Thoracic versus abdominal approach to correct diaphragmatic eventration in children

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Abstract

Background

Plication of diaphragm (DP) for eventration (DE) can be done using thoracic or abdominal approaches. The purpose of our study was to compare outcomes between these approaches.

Methods

Retrospective records of children <16 years who underwent DP (single-centre, 2004-2018) were recorded and analysed. Data are reported as median (range).

Results

Eighty-nine cases were identified in thoracic (Congenital=5, Acquired=84) and 13 (Congenital=10, Acquired=3) in abdominal group aged 5.88 (0.36-184.44) and 10.0 (0.12-181.8) months. Improvement in diaphragm level post-DP was significantly higher in abdominal [2(0-4)] than chest [1.5(0-5)] group ($p=0.04$). On Cox regression analysis, there was no difference in time to extubation ($p=0.2$) or time to feed ($p=0.18$) between the 2 groups. Patients operated transthoracically left intensive care unit after a significantly longer time ($p=0.04$). All 16 recurrences were in the chest group although this was not statistically significant due to skewed patient numbers ($p=0.19$).

Conclusion

This is one of the largest reports on outcomes of children undergoing DP for DE. There was no significant difference in recurrence rate, even though all recurrences in our series (15.7%) were in the acquired cases operated using a thoracic approach.

Keywords

Diaphragmatic eventration, plication, thoracotomy, thoracoscopy, laparotomy, laparoscopy

Level of evidence

Level III (Treatment retrospective comparative study)

Diaphragmatic eventration (DE) is not very common among paediatric patients. However, successful diaphragmatic plication (DP) can be life changing for the affected children. DE can either be 'congenital' as a result of defective embryological development of the diaphragmatic muscle or 'acquired' secondary to phrenic nerve damage as a result of birth trauma or mediastinal surgery [1,2]. Congenital DE is rare with an incidence of 0.02-0.07 per 1000 births [3]. Phrenic nerve palsy has been observed in 0.3-12.8% of children undergoing cardiac surgery [4,5]. As the consequences of DE from either aetiology are similar, they are usually discussed and reported together.

Surgical repair of DE in children essentially involves plicating the redundant central portion of the diaphragm. Different routes have been employed for surgical access, beginning with a laparotomy in 1927 followed by open transthoracic, thoracoscopic and laparoscopic approaches [6,7].

While proponents of a thoracic approach claim the advantage of better visualisation of the phrenic nerve and inherent circumvention of the peritoneal cavity, proponents of an abdominal approach proclaim a liberal working space, avoidance of single lung ventilation and clear visualisation of adjacent structures, especially bowel [8-10]. In the minimally invasive surgery (MIS) era, similar plea have been advocated for thoracoscopic and laparoscopic repairs in addition to the common tenets of quicker recovery, smaller incisions and decreased pain [11].

Personal experiences of various surgeons with each of these approaches has been reported in literature, but there is no defined 'best practice' to date. We report here our experience with the use of both routes for DP and compare outcomes, especially recurrence, between transthoracic and transabdominal approaches.

1. Material and methods

1.1 Patients

Following institutional audit approval (Ref No. 2509), case records of patients (< 16 years age) who underwent surgical repair (DP) of DE at a single tertiary referral centre between January 2004 and December 2018 were retrospectively reviewed. Patient characteristics, diagnosis, management and outcomes were recorded and analysed. Outcomes included improvement in the level of diaphragm, as assessed by the difference in number of rib levels on a plain chest X-ray before and after the operation; time to extubation; length of intensive care unit (ICU) stay; time to feed and recurrence.

1.2 Statistical analysis

We divided our patients into 2 groups – those operated via a thoracic or an abdominal approach. Data were analysed applying standard statistical tests using software IBM SPSS Version 25 (SPSS Inc., Chicago, IL, USA). Patient characteristics were compared using Mann-Whitney U test for continuous variables. Fisher's exact test was used for dichotomous variables. Outcomes between the two groups were analysed using multivariate Cox-regression and the log-rank test for univariate analysis. P value of ≤ 0.05 was considered significant.

2. Results

A total of 102 patients (65 boys, 37 girls) underwent DP for DE. Eighty-nine patients (87%) underwent transthoracic (Open=86, MIS=3) and 13 (13%) transabdominal (Open=4, MIS=9) plication of the diaphragm (**Table 1**). The groups were comparable with regards to gender distribution, age, weight at surgery and laterality. Most children had an acquired DE (87/102) secondary to phrenic nerve injury related to cardiac (n=80) or other mediastinal surgery (n=7) and were operated using chest approach in 84/87 (97%). On the other hand, a significantly higher proportion of children had congenital DE in the abdominal group.

Median improvement in the level of diaphragm was significantly greater in the transabdominal as compared to the transthoracic group (2 rib levels vs. 1.5 rib levels respectively; $p=0.04$) (Figure 1).

Time to extubation was compared between chest and abdominal approaches by Cox regression analysis, with age at diagnosis, open/MIS and congenital/acquired as co-variables and this was not significant (hazard ratio=0.54 [0.21-1.40] $p=0.2$). Patients operated by the chest approach left ICU after a significantly longer time (hazard ratio=0.34 [0.12-0.97] $p=0.04$). There was no difference between time to feed between the 2 groups (hazard ratio=1.80 [0.76-4.25] $p=0.18$).

There were 16 short term recurrences in the chest group and none in the abdominal group but this was non-significant (log-rank test, hazard ratio=0.32 [0.06-1.68], $p=0.19$). Of 16 recurrences in the transthoracic group, 13 underwent a redo transthoracic operation, whereas 3 underwent transabdominal procedure for their second repair. There were 17 post-operative complications in addition to the recurrences, these are listed in Table 2; there was one post-operative mortality from necrotising enterocolitis.

3. Discussion

The diaphragm is perhaps the only organ in the human body that is so strategically placed so as to allow access from two completely different directions. Paediatric surgeons, over the years, have taken advantage of this fact and successfully approached it through the chest as well as the abdomen, beginning with open surgical procedures and moving on to minimally invasive techniques as technology has advanced. Majority of the literature on paediatric DE repair describes personal or single institutional experiences using one approach or the other [12-24] with limited number of multicentric studies [25,26]. Pros and cons of each of these approaches have been clearly defined by various adult and paediatric studies but there is no consensus regarding 'best practice'. Whether outcomes between the two approaches are different or not was the driver for us to review our own experience using both approaches.

The primary outcome of clinical interest is the recurrence rate after surgery. We found all our recurrences in the transthoracic group only (16/102), although a comparison with the abdominal group is limited by the shorter follow-up. It is notable that the recurrences can be grouped into 'early' (<1 year) or 'late'; although the abdominal group appears to have no early recurrences, we do not yet know whether there may be late recurrences. On aetiological segregation, 15/16 recurrences were in children with 'acquired' and 1/16 in 'congenital' DE. The majority of the studies previously published in literature have reported nil recurrence rates except for two, one describing outcomes after the thoracoscopic approach (25% recurrence) and one reporting on laparoscopic repair (46% recurrence) [21,25]. In the first study, the thoracoscopic technique involved use of an endostapler to resect the redundant diaphragm in one of the patients with recurrence [25]. This method is not standard practice and the authors have given a word of caution against it. The other patient experiencing a recurrence underwent a thoracoscopic DP both as primary and redo procedure. The second study reported recurrences in 6 of 13 children after laparoscopic repair and all underwent redo laparoscopic repair with no further recurrences reported [21].

Difference in the level of the highest point of diaphragm can be calculated using a pre and post surgical chest radiograph. This could be an objective indicator of 'effectiveness' of the procedure, although post-operative requirement of respiratory support defines 'success' clinically. We found a higher degree of improvement in the diaphragmatic level after the abdominal as compared to the chest approach. This might be related to better visualisation of the entire diaphragm, including costophrenic recesses, especially when laparoscopic artificial pneumoperitoneum balloons it out like a dome. This allows the surgeon to take stitches as peripherally as needed to make the diaphragm taught, keeping the bowel safely retracted. On the contrary, this visualisation of the entire periphery is compromised in a thoracotomy, especially if done after previous cardiothoracic surgery, which can lead to dense adhesions. Additionally, in thoracoscopic operations, an artificial pneumothorax may keep the diaphragm relatively flat due to the increased intrathoracic pressure, thereby making it difficult to gauge the effectiveness of the repair intra-operatively. Previous studies have assessed improvement in level of diaphragm post plication, but none has compared this parameter between different approaches [14,24].

Time to extubation is an indirect evidence of improvement in respiratory function. We found a longer time to extubation and intensive care requirement in patients operated using a transthoracic versus a transabdominal approach. This may be attributed to post-operative pain and splinting of diaphragm after a thoracotomy; or single lung ventilation, hypercarbia and artificial pneumothorax causing impaired venous return after a thoracoscopic procedure causing haemodynamic and respiratory compromise [27,28]. Previously published studies have reported successful median time to extubation between 0-2 days after transthoracic plication, erring on the lesser side for MIS as compared to open technique [15-18,22]. In a review of 18 cases of acquired DE operated laparoscopically, median time to extubation was 0 (0-2) days [14]. However, no difference was noted in another study comparing thoracoscopic and laparoscopic repairs [25].

Another important surgical outcome measure is the time to commence feeds. We found comparable time to commence feeds in the chest and abdominal groups. An abdominal operation with handling of bowel can cause a brief period of post-operative ileus that is clearly avoided in a thoracic surgery. However, with laparoscopic procedures, bowel handling is usually less than in an open abdominal operation. Median time to feeds in a previously reported study was 1.6 days (1-4) in both groups and this was not significantly different [25].

Our study has several limitations. Our own experience is a retrospective review with limited data on surgical decision-making and limited follow-up, especially in the abdominal group. We found no significant difference in recurrence rates when comparing approach (chest or abdomen), aetiology (congenital or acquired) or technique (open or minimally invasive). We did, however, note a higher recurrence rate in trans-thoracically repaired, acquired DEs plicated using open technique, yet asymmetric patient distribution between groups in all 3 categories (approach, aetiology and technique) limits the validity of the analysis. Which of the three is the single most important predictor of outcome remains to be proven using a prospectively conducted study.

4. Conclusion

Our experience of 102 patients who underwent DP for DE is the largest paediatric series reported to date. We found a higher recurrence rate with transthoracic as compared to transabdominal repair. Further evidence is required to define the optimal approach.

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Table legends

Table 1. Patient characteristics of transthoracic and transabdominal diaphragmatic plication groups

Table 2. Post-operative complications as per Clavien-Dindo classification

Figure legends

Figure 1. Median preoperative as well as postoperative rib levels of the diaphragmatic dome were lower by 2 rib levels (8th vs. 6th rib) or 1.5 rib levels (9.5 vs. 8th rib) in the chest group compared to the abdominal group, respectively ($p < 0.001$ both comparisons). However, median improvement in diaphragm level was greater after abdominal approach compared to the chest approach [2 (0-4) rib levels vs. 1.5 (0-5) rib levels; $p = 0.04$].

Figure 2. Although all 16 recurrences were in the chest group, but this was not statistically significant due to skewed patient numbers in both the groups (Hazard ratio=0.32 [0.06-1.68], $p = 0.19$).

Table 1. Patient characteristics and outcomes between transthoracic and transabdominal diaphragmatic plication groups

	Chest	Abdomen	P value
Number of patients (Total, n=102)	89	13	
Sex [M/F]	57/32	8/5	>0.99
Age at surgery [months] ◇	5.88 (0.36–184.4)	10.0 (0.1-181.8)	0.601
Weight at surgery [kg] ◇	5.86 (1.3-41.7)	8.45 (3-50)	0.282
Laterality [Right/Left/Bilateral]	45/43/1	6/7/0	0.803
Aetiology [Congenital/Acquired]	5/84	10/3	<0.001
Surgical technique [Open/minimally invasive]	86/3	4/9	<0.001
Follow-up [months] ◇	3.8 (0-137)	3.2 (0-26.2)	0.372

◇ Data reported as median (range)

Table 2. Post-operative complications as per Clavien-Dindo classification

Complications	n	Clavien-Dindo grade
Chest group		
Haemothorax	1	III-b
Lung collapse/consolidation	4	I
Necrotising enterocolitis	3	II (n=2) V (n=1)
Pneumothorax	1	I
Pyopneumothorax	2	I
Chylothorax	3	II
Cardiac arrhythmia	1	I
Abdominal group		
Adhesive bowel obstruction	2	III-b
Lung consolidation	1	I

Figure 1

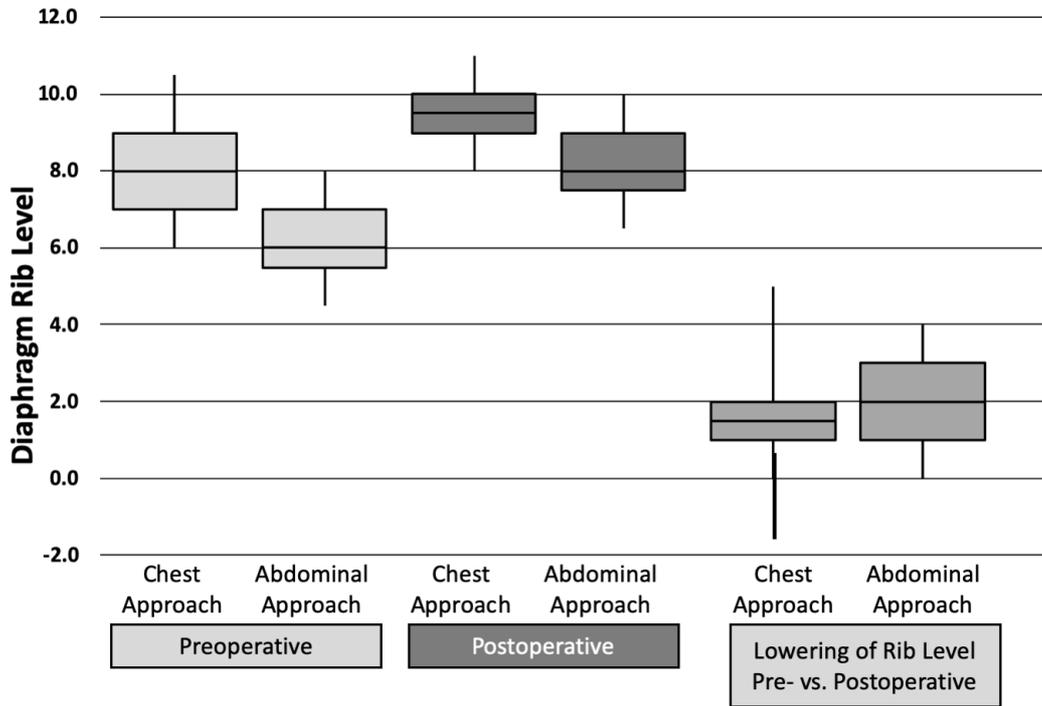


Figure 2

