Surgical treatment of 125 cases with congenital diaphragmatic eventration in a single institution

Shengliang Zhao
Chongqing Medical University Affiliated Children's Hospital

Zhengxia Pan
Chongqing Medical University Affiliated Children's Hospital

Yonggang Li
Chongqing Medical University Affiliated Children's Hospital

Yong An
Chongqing Medical University Affiliated Children's Hospital

Lu Zhao
Chongqing Medical University Affiliated Children's Hospital

Xin Jin
Chongqing Medical University Affiliated Children's Hospital

Jian Fu
Chongqing Medical University Affiliated Children's Hospital

Chun Wu (✉ WuChun0312@sina.com)
Chongqing Medical University Affiliated Children's Hospital

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Abstract

Background: This study sought to investigate the clinical characteristics of congenital diaphragmatic eventration (CDE) and to compare the efficacy of thoracoscopy and traditional open surgery in infants with congenital diaphragmatic eventration.

Methods: We retrospectively analyzed the clinical data of 125 children with CDE (90 boys, 35 girls; median age: 12.2 months, range 1h-7 years; body mass 1.99-28.5 kg, median body mass 7.87±4.40 kg) admitted to our hospital in recent 10 years, and statistically analyzed their clinical manifestations and surgical methods.

Results: 108 children in this group underwent surgery, of which 67 underwent open surgery and 41 underwent thoracoscopic diaphragmatic Plication. 107 patients recovered well postoperatively, except for 1 patient with hiatal hernia died of respiratory distress after surgery. Followed up for 1-9.5 years, 107 patients had significantly improved preoperative symptoms. Eleven of the 17 children who did not undergo surgical treatment did not see a significant decrease in diaphragm position after 1-6 years of follow-up. In the thoracoscopy group, the lung function values of 13 children before operation and 3 years after operation were compared. The difference in percent of expiratory time to peak tidal time (tPTEF%tE) and percentage of tidal volume to peak tidal expiratory flow (VPEF%VE) was significant (P<0.05). In the thoracoscopy group, the index data on the operation time, intraoperative blood loss, chest drainage time, postoperative mechanical ventilation time, postoperative hospital stay, and postoperative CCU admission time were better than those in the open group. The difference between the two groups was statistically significant (P<0.05).

Conclusions: Clinical symptoms of congenital diaphragmatic eventration varied in severity. Patients with severe symptoms should be operated early. All kinds of surgical methods can achieve better results. The clinical symptoms of children after diaphragm Plication operation are better than those before operation. We believe that the continuous suture of barbed wire thread applied to thoracoscopy diaphragm Plication surgery has quick operation time, less trauma, and quick recovery should be the preferred treatment method for infants with congenital diaphragmatic eventration.

Background

CDE is considered to result from a congenital anomaly during the formation of the pleuroperitoneal membrane, as in Bochdalek diaphragmatic hernia, but occurring in a later stage during embryonal growth[1]. CDE is a rare pathology occurs in 0.02 to 0.07/1,000 births affecting mostly males in 60 to 80% of cases. It accounts for 5% - 7% of all diaphragm diseases[2]. Because the infant ribs are horizontal and the intercostal muscles are weak, the breathing movement mainly depends on the abdominal breathing of the diaphragm muscles moving up and down. Infants and children with CDE have abnormally elevated diaphragm muscles, which often lead to collapse of the affected alveoli or atelectasis, affecting lung ventilation and lung development. Therefore, infants and children with CDE
often have symptoms such as dyspnea, repeated respiratory infections, low weight, and stunting. Severe cases may manifest as respiratory distress syndrome, seriously affecting the quality of life of children. Traditionally, diaphragmatic plication has been performed by thoracotomy or laparotomy, particularly in symptomatic smaller children [3]. However, advancements in endoscopic surgery have allowed diaphragmatic eventration to be treated quickly and safely. Here, we present our experience with different surgical procedures to treat 125 cases with CDE. Presently reports as follows:

**Methods**

### 3.1 General Information

Inclusion and exclusion criteria for children with CDE in the surgical group: Children with CDE have dyspnea, repeated respiratory tract infection and other symptoms. Chest X-ray, CT or gastrointestinal radiography clearly diagnose diaphragmatic eventration. Exclude children with acquiring diaphragmatic eventration associated with surgery. Non-surgical group: children with no obvious clinical symptoms, but chest X-ray, CT or gastrointestinal angiography clearly diagnosed diaphragmatic eventration.

The study included 125 children diagnosed with CDE admitted to the department of cardiothoracic surgery of Children's Hospital of Chongqing Medical University from January 2010 to January 2019. There were 90 males (72%) and 35 females (28%), aged 1h-7 years, median age: 12.2 months, with body mass of 1.99-28.5 kg (7.87±4.40 kg), 78 children (62.4%) on the right, 47 children (37.6%) on the left, and no bilateral children. In this group, there were 48 cases of simple CDE, 33 cases with intracardiac malformation, and 44 cases with other malformations. The types of malformations are shown in table 1.

### 3.2 Clinical Manifestations and Imaging Findings

The clinical symptoms of CDE were reported for 108 of 125 cases. The main symptoms of CDE in infants included the following: cough and asthma, dyspnea, recurrent respiratory tract infections, refuse milk and vomit, and arrhythmia. About 17 cases were found to asymptomatic or accidentally discovered on routine physical examination. The clinical symptoms of CDE are shown in table 2. All the 125 cases had positive manifestations on chest X-ray, among which 39 cases (31.2%) were diagnosed by combined chest CT, 32 cases (25.6%) were diagnosed by combined chest X-ray and digestive tract radiography, and 99 cases were found to have significant eventration of diaphragmatic shadow. All cases were confirmed as CDE after surgery, and the Position of CDE was presented in table 3.

### 3.3 Surgical Methods

Open surgery group: thoracotomy for the right diaphragm eventration and laparotomy for the left diaphragm eventration. Through the thoracoabdominal approach, we remove the weak diaphragm and intermittently sew with non-absorbable sutures to ensure that the cut diaphragm into a shingled shape to strengthen the weak area of the diaphragm. 41 underwent transthoracic diaphragmatic Plication, including 1 patient with congenital heart disease and 1 patient with isolated lung. 26 underwent transabdominal...
diaphragmatic Plication, among which 9 cases were diagnosed as CDE preoperatively. During the operation, the stomach, duodenum, spleen and part of the liver were herniated into the chest cavity, and the postoperative diagnosis of diaphragmatic hernia was found. Thoracoscopic group: using the three-hole method, 5 mm thoracoscopy is placed on the lower edge of the scapula tip trocar, and two operation holes are made in the fourth intercostal space on both sides of the trocar. Continuous barbed sutures from the outside to the inside are utilized to make the diaphragm into a shingled shape to strengthen the diaphragm. See Figure 1 for details. Non-surgical group: 17 decided to give up surgery or follow-up outside the hospital.

3.4 Lung function measurement methods

We selected children before 3 years old in the thoracoscopy group for preoperative and postoperative lung function tests. Because children before 3 years old cannot follow the instructions for active breathing, only tidal volume analysis. Lung function was measured when there were no obvious abdominal distension 12 hours after a meal, and the patients were in a natural sleep state during the measurement. Clean up the secretions of the patient's nasopharynx before operation to keep the airway clear. During the operation, patients should lie in the supine position, with the neck stretched back slightly, place the breathing mask on the mouth and nose properly, and hold up the lower jaw to avoid air leakage.

3.5 Statistical analyses

All the collected data were statistically analyzed using SPSS 22.0 software. The continuous variables were expressed as mean±standard deviation, and the classification variables were expressed as ratio columns. Preoperative and postoperative data were compared, using paired design Student's t-test, comparison between the two groups was expressed by independent sample t test, p value<0.05 was considered statistically significant.

Results

We analyzed the data of the relevant surgical indicators of the two groups. In the thoracoscopy group, the index data on the operation time, intraoperative blood loss, chest drainage time, postoperative mechanical ventilation time, postoperative hospital stay and postoperative CCU admission time were better than those in the open group. The difference between the two groups was statistically significant (p<0.05). There was no statistically significant difference between the two groups in descending distance of diaphragm (P>0.05). See Table 4 for details.

Because some patients were lost to follow-up, only 13 patients in the thoracoscopy group had pulmonary function values. Compared with the preoperative and postoperative pulmonary functions in the thoracoscopy group, the preoperative analysis tPTEF%tE and VPEF%VE were both significantly lower than the postoperative results. P<0.05 indicated mild to moderate small airway obstruction. The TEF50 as%TIF50 was significantly lower than that of postoperative (P<0.05), indicating mild large airway obstruction, and the PTEF as% VT, the TEF50 as%TIF50 were significantly lower than that of
postoperative (P<0.05), indicating light large airway obstruction. The PTEF as %TEF25 was significantly higher than that of postoperative, and P<0.05 suggested mild small airway obstruction, as shown in table 5.

Patients had been followed-up radiologically annually to demonstrate the position of the diaphragm, and symptoms if any were also evaluated. In open surgery group, 1 case died of respiratory distress after operation with esophageal hiatal hernia. Almost all respiratory and digestive symptoms disappeared within 1 month after the operation, and none had any symptom 3 years after surgery. See table 6 for details. Followed up for 1-9.5 years, 107 patients had significantly improved preoperative symptoms. Eleven of the 17 children who did not undergo surgical treatment did not see a significant decrease in diaphragm position after 1-6 years of follow-up, and 6 patients were lost to follow-up. The comparison of chest radiographs before and after operation is shown in Figure 2. The pathological examination of all open surgery prompts: diaphragmatic tissue congestion and edema, focal bleeding, pathologically visible focal diaphragm muscle is thin, fibrous tissue hyperplasia, in line with CDE, see Figure 3 for details.

Discussion

CDE is characterized by incomplete muscle regeneration. Subsequent abnormally elevated diaphragm muscles cause abnormal movement of the affected hemidiaphragm during respiration. It can occur locally or affect the entire diaphragm. In this study, there were 90 males (72%), 35 females (28%), 78 children (62.4%) on the right side, and 47 children (37.6%) on the left side. We observed that the incidence was higher in male children, and the incidence on the right side was higher than that on the left side. CDE can be associated with other developmental defects, and associated comorbidities include congenital hypoplastic lung, congenital heart disease, pectus excavatum, cleft palate, hypospadias, cryptorchidism, and congenital torticollis [4]. 77 patients in this group were combined with other malformations, complicated congenital heart disease (19, 15.2%) and congenital hypoplastic lung (16, 12.8%) were the main relevant abnormalities in this study. The above facts is difficult to determine whether CDE is accompanied by other malformations or other malformations with this disease. Its numerous accompanying malformations suggest that the cause of the teratology is difficult to explain with a single etiology, and may be similar to the cause of other congenital malformations.

The main symptom of CDE is the compression of the lower lobe of the lungs due to the increase of intra-abdominal organs. After compression, the mediastinum can also cause the mediastinum to move on the health side and reduce the health side lung function accordingly. In unilateral CDE, the lung capacity and total lung capacity is reduced by 20% - 30% [5]. Bilateral diaphragmatic eventration reduces the lung functions even more seriously, especially in the supine position [6]. The comparison of lung function before and 3 year after operation in this group of patients showed significant differences in the tPTEF%tE, VPEF%VE and other lung function values (P<0.05), suggesting Lung function shows signs of mild to moderate small airway obstruction. Therefore, we believe that children with respiratory symptoms should be performed diaphragm Plication as soon as possible, which is conducive to the recovery of lung
function. However, due to the small sample size of lung function and the lack of long-term follow-up results, the conclusions may be biased. Still waiting for further clinical research.

The treatment principle of CDE is to restore the normal anatomical position and tension of the diaphragm, the method is to strengthen the weak diaphragm, the goal is to maintain the normal volume of the lungs and the process of lung ventilation. Whether asymptomatic patients need surgical correction has been controversial for a long time. In this group of 17 children who did not undergo surgical treatment, 11 patients received 1-6 years of follow-up and did not see a significant decrease in diaphragm position. Therefore, we believe that symptomatic children need timely surgical treatment. Yazici M et al.'s study also considered symptomatic Children, usually require surgery [7]. Therefore, we believe that the indications for surgery are as follows: Relative to the normal position, the diaphragm is displaced upwards by 3 intercostals and above. The diaphragm eventration caused obvious compression on the affected side of the lung, and obvious shortness of breath, asthma and other respiratory distress symptoms. Frequent lung infections, hypoxemia, and even abnormal breathing exercise. During the follow-up, the diaphragm continued to rise and the eventration was aggravated.

The traditional treatment method of CDE is diaphragmatic plication performed either by laparotomy or thoracotomy. However, with the development of minimally invasive technology, thoracoscopy is gradually applied in the treatment of CDE [8-10]. We believe that children with right diaphragm eventration and intrapulmonary malformation need to be corrected through the thoracotomy approach as the first choice, because it is not affected by the intestinal canal, full exposure, easy to operate, can see the phrenic nerve and reduce postoperative intestinal paralysis. The laparotomy is suitable for children with left diaphragmatic eventration, inability to distinguish between diaphragmatic eventration and diaphragmatic hernia, and considering gastrointestinal malformation. Because the left chest is the heart, there is a high risk of thoracotomy. The use of subcostal incision is conducive to the repair of the herna and the discovery of possible intestinal malformations. The preoperative diagnosis of 9 children in this group was unknown, and diaphragmatic herna and other gastrointestinal tract malformations were found during the operation, so the choice of preoperative approach was particularly important. We resect the weak diaphragm in the diaphragm via the thoracoabdominal route and sutured the diaphragm intermittently with non-absorbable sutures to make the cut diaphragm imbricate to strengthen the weak area of the diaphragm. The advantage of this technique is that it increases the tension of the diaphragm to evenly distribute the tension throughout the repair area.

With the development of minimally invasive technology, thoracoscopy is gradually used in the treatment of CDE. We compared the effect of open surgery and thoracoscopy in the treatment of CDE in children. The operation time, chest drainage time, postoperative mechanical ventilation time, postoperative hospital stay and postoperative CCU admission time in the thoracoscopy group were shorter than those in the open group, and the difference between the two groups was statistically significant (P<0.05). We consider the possible reasons as follows: Thoracoscopic surgery adopts three hole method, which is less traumatic and less prone to bleeding. The recovery of children is faster after operation. The technique of thoracoscopy is skilled, and the operator and assistant cooperate with each other. We used barbed wire
to sew continuously without knot, which greatly shortens the operation time and is obviously better than
the open surgery.

In this group of 41 children without other thoracoabdominal malformations that need to be corrected, we
used thoracoscopic diaphragm plication. We used barbed wire to suture the diaphragm from the outside
to the inside in a continuous imbricated fashion to strengthen the diaphragm. Combined with the literature
and our experience, compared with ordinary absorbable suture, continuous suture of the diaphragm with
barbed wire has the following advantages: starting from the second stitch, it is not easy to slip after
tightening the suture. One stitch is sewn to tighten one stitch, and no knot is needed during the suture
process, which greatly shortens the operation time. The diaphragms were sutured continuously by
barbed wire to make the diaphragms stretch evenly from the center to all directions, and the tension
distribution was uniform, so that the movement of the diaphragms was more coherent, and the
diaphragms would not be ischemic due to over tight suturing, nor would the suture relax to cause
reurrence. The barbed wire suture is close, less bleeding, wireless knot, absorbable, wireless knot
reaction and residual suture. There is a view that continuous suture may compromise the safety of the
suture and the loosening of the knot may affect the folding of the entire diaphragm, but there is no
evidence to support this view [11]. A. Parlak, et al., and others adopted double-purse suture method to
strengthen the diaphragm, achieving better clinical effect [12]. The usual advantages of thoracoscopy,
such as reduced postoperative pain, satisfactory appearance and rapid recovery, are also applicable to
our surgery, and should be the preferred treatment for CDE. There are some complications in the
treatment of diaphragmatic plication by endoscopic surgery, such as tissue and organ bleeding, fluid
pneumothorax, chylothorax, atelectasis, liver injury, and gastrointestinal perforation, especially in
newborns and infants [13].

Conclusion

Clinical symptoms of CDE varied in severity. Patients with severe symptoms should be operated early. All
kinds of surgical methods can achieve better results. The clinical symptoms of children after diaphragm
Plication operation are better than those before operation. We believe that the continuous suture of barbed
wire thread applied to thoracoscopy diaphragm Plication surgery has quick operation time, less trauma,
and quick recovery should be the preferred treatment method for infants with congenital diaphragmatic
eventration.

Abbreviations

CDE (congenital diaphragmatic eventration), ASD (atrial septal defect), VSD (ventricular septal defect)

PDA (patent ductus arteriosus), TAPVC (total anomalous pulmonary venous connection), CCU (coronary
care unit), CT (computed tomography), RR (respiratory rate), TV/kg (tidal volume per kilogram),
tPTEF%tE (percent of expiratory time to peak tidal time), VPEF%VE (percentage of tidal volume to peak
tidal expiratory flow), PTEF as%TEF 2 (The ration of the tidal expiratory flow at 25% of the remaining tidal
volume to peak tidal expiratory flow\(^{28}\) PTEF as\%VT\(^{29}\) the ratio of peak tidal expiratory to tidal volume

\(^{26}\) TEF50 as\%TIF50\(^{27}\) (the ratio of mid-expiratory to mid-inspiratory flow) 

\(^{28}\) TIF (the ratio of mid-expiratory to mid-inspiratory flow) 

\(^{29}\) Ratio ti to tE\(^{30}\) (the ratio of inspiratory time to expiratory time) 

\(^{31}\) PEF (peak expiratory flow) 

\(^{32}\) TEF75\% (expiratory flow in 75\% tidal flow) 

\(^{33}\) TEF50\% (expiratory flow in 50\% tidal flow) 

\(^{34}\) TEF25\% (expiratory flow in 25\% tidal flow)

**Declarations**

**Ethics approval and consent to participate:**

Medical Research Ethics Committee of Children's Hospital Affiliated to Chongqing Medical University. This study obtained written informed consent from all participants.

**Consent for publication:**

Not applicable

**Availability of data and materials:**

The datasets used and analysed during the current study are available from the corresponding author on reasonable request.

**Competing interests:**

The authors declare that they have no competing interests

**Funding:**

Not applicable.

**Authors' contributions:**

ZSL analyzed and interpreted the patient data regarding the congenital diaphragmatic eventration. JX, FJ, ZL was responsible for data collation. PZX, AY, LYG was responsible for part of the design of the paper. PZX, AY, LYG, WC performed the continuous suture of barbed wire thread applied to thoracoscopy diaphragm Plication surgery. WC was a major contributor in writing the manuscript. All authors read and approved the final manuscript.

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References


Table 1. Associated malformations with congenital diaphragmatic eventration (n = 125)

Associated malformation rate meant the probability of one specific malformation appearing in 125 patients. *1 (including two or more ASD, VSD, PDA, TAPVC) *2 (including one of ASD, VSD, PDA)

<table>
<thead>
<tr>
<th>Types of CDE with malformation</th>
<th>No. of cases</th>
<th>Rate(%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Complex congenital heart disease *1</td>
<td>19</td>
<td>15.2%</td>
</tr>
<tr>
<td>Congenital hypoplastic lung</td>
<td>16</td>
<td>12.8%</td>
</tr>
<tr>
<td>Single congenital heart disease *2</td>
<td>14</td>
<td>11.2%</td>
</tr>
<tr>
<td>Pectus excavatum</td>
<td>8</td>
<td>6.4%</td>
</tr>
<tr>
<td>Hiatal hernia of esophagus</td>
<td>4</td>
<td>3.2%</td>
</tr>
<tr>
<td>Right shift heart</td>
<td>3</td>
<td>2.4%</td>
</tr>
<tr>
<td>Chicken breast</td>
<td>3</td>
<td>2.4%</td>
</tr>
<tr>
<td>Congenital malrotation of intestine</td>
<td>2</td>
<td>1.6%</td>
</tr>
<tr>
<td>Congenital pyloric stenosis</td>
<td>1</td>
<td>0.8%</td>
</tr>
<tr>
<td>Chylothorax</td>
<td>1</td>
<td>0.8%</td>
</tr>
<tr>
<td>Cryptorchidism</td>
<td>1</td>
<td>0.8%</td>
</tr>
<tr>
<td>Isolated lung</td>
<td>1</td>
<td>0.8%</td>
</tr>
<tr>
<td>Auricular deformity</td>
<td>1</td>
<td>0.8%</td>
</tr>
<tr>
<td>Congenital microphthalmos</td>
<td>1</td>
<td>0.8%</td>
</tr>
<tr>
<td>Bilateral inguinal hernia</td>
<td>1</td>
<td>0.8%</td>
</tr>
<tr>
<td>Meckel's diverticulum of ileum</td>
<td>1</td>
<td>0.8%</td>
</tr>
</tbody>
</table>

Table 2. Symptoms of congenital diaphragmatic eventration in children (n = 125).

Rate meant the probability of symptom appearing in 125 patients.
Clinical symptoms | No. of cases | Rate(%) |
--- | --- | --- |
Cough and asthma, difficulty breathing | 61 | 48.8% |
Recurrent respiratory tract infection | 15 | 12.0% |
Shortness of breath, cyanosis | 17 | 13.6% |
Refuse milk and vomit | 9 | 7.2% |
Arrhythmology | 6 | 4.8% |
Asymptomatic chest X-ray findings | 17 | 13.6% |

Table 3. The Position of congenital diaphragmatic eventration found during operation (n = 108)

Rate meant the probability of the Position of congenital diaphragmatic eventration appearing in 125 patients.

<table>
<thead>
<tr>
<th>Diaphragmatic position</th>
<th>No. of cases</th>
<th>Rate(%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Third front rib</td>
<td>18</td>
<td>16.67%</td>
</tr>
<tr>
<td>Fourth front rib</td>
<td>67</td>
<td>62.04%</td>
</tr>
<tr>
<td>Fifth front rib</td>
<td>11</td>
<td>10.19%</td>
</tr>
<tr>
<td>Sixth front rib</td>
<td>3</td>
<td>2.78%</td>
</tr>
<tr>
<td>Concomitant esophageal hiatal hernia and other malformations were not clearly identified during operation</td>
<td>9</td>
<td>8.33%</td>
</tr>
</tbody>
</table>

Table 4. Comparative analysis of operative related indexes between open group and thoracoscopy group (n = 108)
Table 5. Preoperative and postoperative pulmonary function comparison of children with congenital diaphragmatic eventration in thoracoscopic group (n =13)

The continuous variables were expressed as mean±standard deviation

*Preoperative and postoperative data were compared. P value of <0.05 was considered statistically significant.
Table 6. Clinical symptoms of congenital diaphragmatic eventration pre- and post-operation (n = 107)

<table>
<thead>
<tr>
<th>Clinical Symptoms</th>
<th>Pre-operation (N)</th>
<th>Post-operation (1 month) (N)</th>
<th>Post-operation (3 year) (N)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cough and asthma, difficulty breathing</td>
<td>60</td>
<td>4</td>
<td>0</td>
</tr>
<tr>
<td>Recurrent respiratory tract infection</td>
<td>15</td>
<td>3</td>
<td>0</td>
</tr>
<tr>
<td>Shortness of breath, cyanosis</td>
<td>17</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Refuse milk and vomit</td>
<td>9</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Arrhythmology</td>
<td>6</td>
<td>2</td>
<td>0</td>
</tr>
</tbody>
</table>

Figures

**Figure 1**

Comparison of diaphragmatic before and after thoracoscopic diaphragmatic plication (n=41) The diaphragm muscle weakness was shown before the diaphragm plication (Fig1a). After the diaphragm plication, the diaphragm was reinforced by continuous suture of barb line (Fig1b).
Figure 2

Preoperative chest radiograph of a child with CDE shows a raised left hemidiaphragm (Fig2a). A radiograph taken in the postoperative recovery period shows the left hemidiaphragm in normal position (Fig2b).

Figure 3
Pathological examination of congenital diaphragmatic eventration showed that the diaphragm was thin(Fig3a) and fibrous tissue hyperplasia(Fig3b).