Thoracoscopic Management of Bochdalek Hernia – Our Experience

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Abstract: Congenital diaphragmatic hernia (CDH) is a lethal birth defect with a reported incidence of 1:3000 live births. Herniation of abdominal contents occurs most often, in over 95% of cases, through the posterior foramen of Bochdalek, with 80% occurring on the left side. Diaphragmatic defects may be approached either through the thorax or the abdomen; this choice depends on the expertise and preference of the surgeon. Supporters of the laparoscopic approach refer to easier manipulation of the instruments and the ability to visualize the reduced viscera, thus avoiding unnoticed lesions. The present study was a prospective study that included patients with CDH presented at the Pediatric Surgery Unit and Neonatal Care Unit, Coimbatore Medical College Hospital, Coimbatore during the period from August 2015 to June 2018. They included 20 cases of Bochdalek diaphragmatic hernia managed by thoracoscopic repair. 15 cases presented in the neonatal period (neonatal group, 75%), and 5 cases presented after the neonatal period (delayed presentation, 25%). Conversion from a thoracoscopic to open repair occurred in 2 cases (10%). Recurrence of Bochdalek hernia occurred in 1 case. The utilization of MIS approaches have been suggested to be advantageous over traditional open surgery including less pain and incisional complications, avoidance of thoracotomy-related sequelae, as well as reduction of surgical stress.

I. Introduction

Congenital diaphragmatic hernia (CDH) is a lethal birth defect with a reported incidence of 1:3000 live births (Conforti and Losty, 2006). Herniation of abdominal contents occurs most often, in over 95% of cases, through the posterior foramen of Bochdalek, with 80% occurring on the left side. Less commonly, retrosternal herniation occurs, through the foramen of Morgagni (Robinson and Fitzgerald, 2007). CDH commonly presents with severe immediate cardiorespiratory distress with cyanosis, tachypnea and tachycardia. However, 10% of CDH may present later in life with a differing clinical picture. The most frequent presentations are respiratory (43%), followed by GI (33%), both respiratory and GI (13%) and asymptomatic (11%) (Yamamoto and Parikh, 2005). Surgical management of CDH traditionally entailed a thoracotomy or laparotomy with primary or patch closure of the diaphragmatic defect. With the increasing use of laparoscopy by pediatric surgeons, a number of more difficult problems are being approached with this modality. Included in this group are repairs of CDH originating from either the foramen of Bochdalek or foramen of Morgagni (Holcomb 3rd et al., 2005). Van der Zee and Bax (1995) described the laparoscopic repair of a congenital posterolateral hernia in a six-month-old infant, while Becmeur et al (2001) reported thoracoscopic repair of three late presenting diaphragmatic hernias in infants.

Diaphragmatic defects may be approached either through the thorax or the abdomen; this choice depends on the expertise and preference of the surgeon. Supporters of the laparoscopic approach refer to easier manipulation of the instruments and the ability to visualize the reduced viscera, thus avoiding unnoticed lesions (Krishna and Zargar, 2002). Supporters of the thoracoscopic route point out that, abdominal viscera are easily reduced with the CO2 insufflated into the thorax (Yang et al., 2005).

Aim of the work

The aim of this study was to evaluate our series of thoracoscopic repairs regarding initial results and preference of this approach in repair of congenital diaphragmatic hernia (CDH) in pediatric age.
Patients and Methods

The present study was a prospective study that included patients with CDH presented at the Pediatric Surgery Unit and Neonatal Care Unit, Coimbatore medical college Hospital, Coimbatore during the period from August 2015 to June 2018. They included 20 cases of Bochdalek diaphragmatic hernia. Their ages ranged from day 1 to 4 years (mean age of 6 months). A written informed consent obtained from child parents. Every parent was receiving explanation to purpose of study and had secret code number and photos applied only to part of the body linked to the research to ensure privacy to participants and confidentiality of data. Research results were only used for scientific purposes. Procedures were approved by both the Institutional and the Regional Ethical Committees. Any unexpected risk appears during the course of the research was cleared to the participants and ethical committee on time and proper measures was taken to overcome or minimize these risks. CDH cases which meet the inclusion criteria underwent thoracoscopic diaphragmatic hernia repair primarily

Study design:
A. Inclusion criteria:
1) Infants (> 1 month) and children diagnosed as having CDH with stable cardiovascular and respiratory conditions are offered a thoracoscopic repair.
2) Neonates presented by CDH are managed by thoracoscopic approach provided that:
   a. Stable cardiovascular and respiratory conditions for at least 24 h.
   b. Ventilator parameters:
      □ □ Oxygen saturation 88-92%.
      □ □ Peak inspiratory pressure (PIP) less than 24 cmH2O.
      □ □ Fraction of inspired oxygen (FiO2) less than 40%.
      □ □ Positive endexpiratory pressure (PEEP) of 3-4 cmH2O.

B. Exclusion criteria:
1) Bilateral diaphragmatic hernias, Morgagni hernias
2) Central, paraesophageal hernias
3) Patients requiring major ventilatory support with unstable cardiovascular and respiratory status.
4) Preterm babies.
5) Neonates with high PaCO2 (> 60 mmHg), acidosis (Ph < 7.25) and PPHT.
6) Sever associated malformations especially complex cardiac lesions.
7) Eventeration of the diaphragm.
8) Patients presented by acute intestinal obstruction with or withoutsuspected strangulation.

II. Results

Twenty cases of congenital diaphragmatic hernia (bochdalek) had been included in this study. Cases were operated upon using thoracoscopic technique at Pediatric Surgery Unit, Coimbatore medical college Hospital during the period from August 2015 to June 2018. The 20 cases included in the study had an age that ranged from day 1 to 4 years (mean age of 6 months), their body weight ranged from 2.8-20 kg (mean 5.6 kg).

Gender distribution included 12 males (60%) and 8 females (40%) (Table 1). Associated anomalies were detected in 3 cases out of 20 cases (15%). The anomalies included: cardiac anomalies in 3 cases in the form of ASD (n=1), patent ductus arteriosus (n=1), PFO (n=1). Cases of Bochdalek hernia were 20 in which 15 cases presented in the neonatal period (neonatal group, 75%), and 5 cases presented after the neonatal period (delayed presentation, 25%). Out of 20 cases, 3 cases of Bochdalek hernia had a sac (15%)
TABLE 1: GENDER DISTRIBUTION

Cases were grouped into 2 groups: group 1 included patients with delayed presentation of Bochdalek hernia (n=5); Group 2 included cases of Bochdalek hernia with neonatal presentation (n=15).

Conversion from a thoracoscopic to open repair occurred in 2 cases (10%), due to injury and perforation of the intestine due to manipulation by instruments, in the other case there was a large defect in a 1 day old baby with almost all abdominal contents in the chest and it was difficult to reduce them and accomplish the procedure. Through a left subcostal incision; laparotomy was done, and after solving the problem, contents reduced into the abdomen; Primary repair of the defect was done with a tube drain positioned in the left upper quadrant. The postoperative course was uneventful. The time needed to reach full enteral feeding (excluding cases converted to open repair) ranged from 6-30 hours (mean 14 hours). Length of hospital stay ranged from 1-4 days (mean 2.4 days). Four neonates in the series were operated while they were on mechanical ventilation that was maintained for 2 days postoperative. The complications were while reduction of contents with injury of the mesentery of small intestine, the second was injury and perforation of the intestine due to manipulation by instruments. Regular follow up of these neonates revealed that recurrence of Bochdalek hernia occurred in 1 case out of 18 cases completed thoracoscopic repair (5.5%). The recurrence occurred after 3 months and was diagnosed by plain x-ray of the chest and abdomen and contrast gastrointestinal radiography. It was repaired by open approach through a left subcostal incision. During laparotomy, partial dehiscence of the repair was found and repaired primarily.
III. Discussion

Congenital diaphragmatic hernia is an anatomical defect in the diaphragm that combines a muscle defect between the abdomen and the thoracic cavity with pulmonary hypoplasia. The muscle defect leads to herniation of the abdominal viscera into the thoracic cavity (Clugston and Greer, 2007). It usually arises from the left posterolateral part of the muscle and is called Bochdalek hernia or less commonly it emerges from the antero-medial retrosternal portion of the diaphragm and is called Morgagni hernia. CDH occurs in 1/2500 to 1/5000 live births with the Morgagni hernia less frequent and usually less urgent than the Bochdalek type (De Buys Roessingh & Dinh-Xuan, 2009). The traditional approach to repair diaphragmatic defects is via a subcostal incision on the ipsilateral side of the hernia. More than 90% of surgeons use this incision, whereas only 6% prefer the thoracic approach (Clark et al., 1998). A tension-free repair of the defective diaphragm is the key to reduce the recurrence rate. When the diaphragmatic defect is small, it can be repaired directly, however, when the defect is large, a prosthetic patch or autologous muscle flap can be used to achieve a tension-free repair (Gaxiola et al., 2009). Both open and minimally invasive thoracic or abdominal approaches to repair the diaphragmatic defects have been described and debate continues regarding the ideal technique. Laparotomy is used most commonly and has several well described advantages over thoracotomy, including easier reduction of intra-abdominal viscera, ability to mobilize the posterior rim of the diaphragm for closure, easier manipulation of the intestines in cases of rotational abnormalities, and avoidance of thoracotomy-associated musculoskeletal abnormalities (Clark et al., 1998).

Minimally invasive surgery (MIS) for infants and children continues to grow. It was first introduced for the treatment of CDH in 1995; Silen et al. used thoracoscopy, while Van der Zee and Bax used laparoscopy. However, most pediatric surgeons have hesitated to apply MIS in CDH because of the associated fragile respiratory status and pulmonary hypertension. MIS appears to be gaining more acceptance for CDH repair thus minimizing the strict selection criteria for achieving this goal (Yang et al., 2005). Some surgeons supported the laparoscopic route, referring to easier manipulation of instruments, less probability of injury to the reduced viscera, avoiding unnoticed lesions, and the possibility of diagnosing and correcting intestinal malrotation associated with CDH (Krishna and Zargar, 2002) (Kohno et al., 2007). Others supported the thoracoscopic approach referring to easier reduction of the herniated viscera with CO2 insufflation into the thoracic cavity, wider space available for manipulating instruments, and easier repair of the defect as the reduced viscera doesn’t obscure the operative field (Yang et al., 2005) (Becmeur et al., 2007). As minimally invasive approaches continue to gain popularity in the pediatric surgical community, the pediatric surgeons should report not only their successes but also their failures. Report of honest information will allow the pediatric surgical community to learn from each member’s experiences and to apply the growing technology appropriately to infants and children. Attempting repair of a CDH using minimally invasive techniques is not an easy task that requires careful planning, patient selection, and cooperation between surgeon, anesthesiologist and an efficient ICU team (Arca et al., 2003). The utilization of MIS approaches have been suggested to be advantageous over traditional open surgery including less pain and incisional complications, avoidance of thoracotomy-related sequelae, as well as reduction of surgical stress, but the open approach have been associated with shorter operative time and a lower recurrence rate. In addition patients who are not candidates to MIS are managed by open approach. Moreover, the use of either laparoscopic or thoracoscopic approaches in the repair of such patients is still a matter of controversy; so the utilization of MIS in the repair of CDH still needs further evaluation and meta-analysis (Cho et al., 2009) (Tsao et al., 2011). For such reason, 20 patients with CDH have been studied in this work, and all of them were operated using thoracoscopic technique. Such patients were divided into the following 2 groups:

**Group (1)** included infants with late-presenting Bochdalek hernia (N.° 5)

**Group (2)** included cases with neonatal presentation of Bochdalek hernia (N.° 15).

In the current study, all cases were Bochdalek hernia. Al-Salem et al (2002) and Yang et al (2006) reported that Bochdalek hernia is the most common type of CDH (90-95%), while Morgagni hernia represents about 3-6 % of patients. In the present study, gender distribution included 12 males and 8 females. It comes in agreement with Baglaj (2004) and Crankson et al (2006) who reported a male predominance in CDH in a ratio of 2:1. A higher male predominance in a ratio of 5:1 had been reported by Pandey et al (2008) and was attributed to more attention being paid to male children in the male-dominated society of the low to medium socio-economic status in Northern India, and so more male children were brought for treatment in preference to female children.

In the current study, associated anomalies were detected in 3 cases (15%). The anomalies included: cardiac anomalies in 3 cases in the form of ASD (n=1), patent ductus arteriosus (n=1), PFO (n=1). Associated anomalies occur commonly in CDH, in as much as 40% of cases in different series. Anomalies identified most commonly include, in descending order: cardiac, renal, central nervous system and gastrointestinal (Graham & Devine, 2005). The association of congenital heart disease with CDH has been described with an incidence of
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10- 35% (Graziano, 2005). Chromosomal anomalies have been described in 10% of affected individuals. CDH is present in conjunction with trisomies 13, 18 and 21 in 8% and has been described in many other syndromes (Garne et al., 2002) (Waag et al., 2008). In the present study, cardiac defects were the most commonly encountered anomalies(15%). It is to be of lower compared to data seen earlier like in Bedoyan et al (2004) who reported a total rate of associated anomalies in 43% of patients, with cardiac anomalies being the most common among them (52%). McHoney et al (2010) reported associated cardiac anomalies in 2 cases (15.4%) out of 13 cases with Bochdalek hernia repaired by thoracoscopic technique. Liem (2003) reported 1 case (14.3%) with anorectal malformation out of 7 cases with Bochdalek hernia repaired by thoracoscopic approach. The most common associated anomalies include congenital heart defects (ASD, VSD, and tetralogy of Fallot) which are reported in 80% of cases, Down’s syndrome, intestinal malrotation (26%), omphalocele and chest wall defects. In the current study, no familial cases were detected among the 20 patients with CDH. Tibboel & Gaag (1996) reported that the majority of CDH cases are sporadic with only 2% occurring with a familial association. In the current study; cases of Bochdalek hernia were 20 which includes 5 cases with late-presenting Bochdalek hernia (group 1= 25%) and 15 cases with neonatal presentation (group 2= 75%). Late presentation of CDH is not common reaching from 5-25% in different series (Weber et al., 1991); cases of CDH with delayed presentation in current series constituted 25% which almost comes with agreement with others. Elhalaby & Abo Sikeena (2002) reported an incidence of 45.5% of late-presenting CDH that was explained by the patient referral pattern in rural locality, which differs significantly from urban areas in Egypt. On the other hand El-Zohiri et al. (2013) reported an incidence of 77% of cases of CDH with delayed presentation which was attributed to the strict selection criteria that have been followed in his study.

In the present study, total mean operative time to perform the thoracoscopic repair in this group of patients was 81.25 minutes. It comes in agreement with results reported by Ferreira et al (2013) who reported a mean operative time of 82 minutes. A shorter mean operative time reported by Liem (2003) who reported repair of 2 neonates with time of 68 minutes. Liem & Dung (2006) who reported repair of 19 neonates with Bochdalek hernia with a mean operative time of 54 minutes, El-Zohiri et al (2013) reported mean operative time 67 minutes for 10 neonates. In the current study, four neonates (20%) were operated while they were on mechanical ventilation that was maintained for 2 days postoperative and then weaned. The mean time needed to reach full enteral feeding was 14 hours.

Recurrence was noted in one neonate in group 2 on follow up, which underwent open repair. No recurrence was noted in group 1. Okazaki et al (2011) reported a single recurrence (20%) among 5 neonates that completed thoracoscopic repair of their Bochdalek hernia. It occurred after 20 days and was managed by open repair. Gander et al (2011) reported 6 recurrences (23%) after successful thoracoscopic repair of 26 neonates. The exact timing of recurrence was not reported, but 2 recurrences occurred during initial admission and 4 cases after discharge.

The technique had been found to be feasible and safe, associated with reduction in surgical stress, quicker recovery, fewer postoperative ventilation days, decrease in postoperative pain and consequently less need for narcotics, improved cosmesis and shorter hospitalization. Moreover, thoracoscopy had also the advantages of easy access, improved visualization and excellent exposure of the herniated viscera with a wider working space.

IV. Conclusion

Congenital diaphragmatic hernia is a complex congenital anomaly that consists of incomplete formation of the diaphragm coupled with herniation of the abdominal viscera into the chest that results in some degree of pulmonary hypoplasia in the ipsilateral lung and to a lesser extent in the contralateral lung. Bochdalek hernia is the most common hernia type of CDH. Bochdalek hernias usually present immediately after birth with respiratory distress while late-presenting hernias represent about 5-20% of cases and are characterized by mild-to-moderate symptoms with a relatively stable general condition. Morgagni hernias are usually diagnosed late due to non-specific nature of symptoms with very few cases detected and managed in the neonatal period. Following a period of preoperative stabilization and optimization; patients with CDH are traditionally repaired surgically via a laparotomy or thoracotomy. Recently, MIS has been applied in the field of CDH utilizing both laparoscopic and thoracoscopic techniques. In the current study, 20 patients with Bochdalek CDH had been treated surgically by Thoracoscopic approach, were divided into 2 groups:

Group (1): included infants with late-presenting Bochdalek hernia (N=5)

Group (2) included cases with neonatal presentation of Bochdalek hernia (N=15)

The study had shown easiness and feasibility of the thoracoscopic route. Thoracoscopic route is much more preferable in treating Bochdalek hernia in infants and children because of easy reduction of hernia contents, short operative time, low conversion rate, short time to reach full enteral feeding, short hospital stay and low rate of complications and recurrences. The same results have been obtained in group (2) patients with
neonatal presentation of Bochdalek hernia, provided that the strict exclusion criteria have been followed; thus minimizing the complications and improving the outcome of thoracoscopic route. Limitations would be lesser number of study population needs elaborate number to derive further significance. Need to compare with other type of diaphragmatic hernia and their outcomes

References

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