Case Report

Late-presenting giant congenital diaphragmatic hernia with acute intestinal obstruction: A unique experience with individualized method of prosthetic mesh placement and staged abdominal wall closure.

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Abstract

Late presenting congenital diaphragmatic hernia is a rare entity, incidence of about 5-30%. In this case report, we discuss the management of a patient who underwent emergency surgery for Bochdalek hernia with intestinal obstruction, which led to a challenge in closure of laparotomy wound in order to accommodate extra loops of bowel that were reduced from left hemithorax. The purpose of this rare case report is to demonstrate our experience with thoraco-abdominal approach for giant Bochdalek hernia with acute bowel obstruction and an innovative staged abdominal wall closure technique applied to prevent complications of open abdomen and post operative abdominal compartment syndrome.

Keywords: Congenital diaphragmatic hernia, Bochdalek hernia, Loss of domain (LOD), Staged abdominal wall closure

Background:

Congenital diaphragmatic hernias comprise Bochdalek, Morgagni and hiatal hernia [1]. Bochdalek hernias result from the failure of fusion of postero-lateral diaphragmatic foramina between the eighth and tenth week of fetal life. Although it typically presents in neonatal period with severe respiratory distress, its delayed presentation in adults has also been reported, incidence of about 5-30% [1]. In this case report, we discuss the management of a patient who underwent emergency surgery for Bochdalek hernia with intestinal obstruction, which led to a challenge in closure of laparotomy wound in order to accommodate extra loops of bowel that

were reduced from left hemithorax. We have demonstrated a staged approach towards abdominal wall closure followed by definitive abdominal wall reconstruction as an alternative to open abdomen with successful outcome.

Case Report:

A 30 year old gentleman came with acute onset left sided chest pain followed by abdominal pain and vomiting. His chest X-ray revealed elevated left hemi-diaphragm and abdominal radiograph revealed distended bowel loops. There was no history of trauma over chest or abdomen. Resuscitation with intravenous fluids, Ryles tube insertion and continuous aspiration, empiric broad spectrum antibiotics were administered.

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Computed tomography of chest and upper abdomen revealed a giant left diaphragmatic hernia, with gastric herniation and multiple small bowel loops in left hemithorax (Fig. 1).

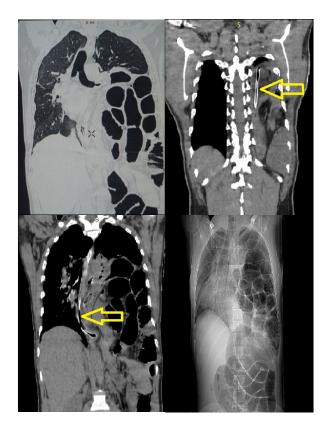


Fig.1 Longitudinal computed tomogram showing giant left sided diaphragmatic hernia with dilated bowel loops (yellow arrow pointing at Ryles tube into the stomach).

In the view of acute condition, patient underwent emergency left thoracotomy (Fig.2) through 8th intercostal space initially followed by separate laparotomy (Fig.3) which was later joined to make it a combined thoraco-abdominal approach (Fig.4). Total intravenous anaesthesia was administered along with single lung ventilation technique using left sided double lumen endo-bronchial tube and a thoracic

epidural catheter inserted prior to the operative procedure for maintaining analgesia during post operative period.

Intra-operative findings revealed a giant Bochdalek hernia with huge 10 x 10 cm defect in left posterior diaphragm with gastric herniation and multiple adhesive bands causing acute small bowel obstruction, with absent peritoneal sac and small left lung lower lobe. Stomach and bowel were found to be viable which brought abdomen, were into diaphragmatic defect was closed by a polypropylene mesh reinforced with omental flap on abdominal side (Fig.5). Anteriorly mesh being sutured to remaining portion of diaphragm and posteriorly anchored to periosteum of vertebral body. Thoracotomy wound closed with placement of two chest drains apical and basal.



Fig.2 Distented bowel loop popping out of thoracotomy incision.



Fig.3 Separate laparotomy incision to aid diaphragmatic repair and reduce the hernia into abdomen.

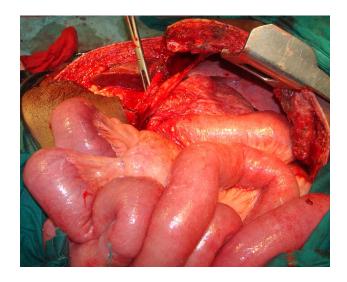


Fig.4 Both incisions joined (thoraco-abdominal incision) in view of multiple adhesions in the hernia.

Due to dilated bowel loops and additional bowel loops as hernia content, it was impossible to achieve primary abdominal wall closure (Fig.6 & 7).

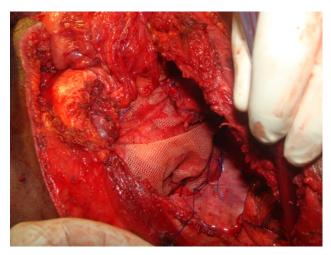


Fig.5 Omental flap over abdominal side reenforced with polypropylene mesh for diaphragm reconstruction.

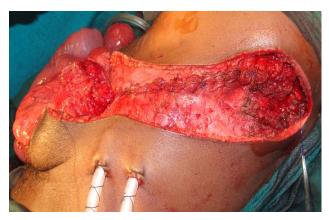


Fig.6 Dilated and additional bowel loops unable to fit in abdomen.

An empty plastic Urobag (the so-called Bógota bag, named for the Colombian surgeon who initially described its use) was opened on three sides to create a strong, non-adherent prosthesis that was sewn to the patient's skin (Fig.8) (thus preserving the patient's fascia unharmed for subsequent closure).



Fig.7 Open abdomen.

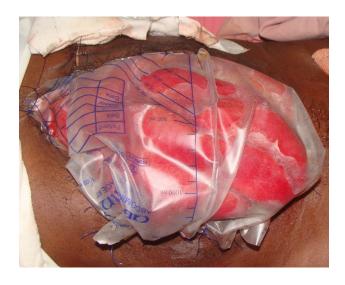


Fig.8 Bogota bag (Urobag sutured over) as temporary cover over oedematous exposed bowel.

This technique is inexpensive in resourcelimited settings, easy to perform, and uses materials that are readily available in any hospital. Patient was kept on ventilator support and epidural analgesia. Two days later, when the patient's visceral edema had subsided; the patient was returned to the operating theatre for primary skin closure. Later bilateral advancement of skin over midline by simple bilateral skin release incisions (as shown in Fig. 9) aided local skin cover over the exposed bowel. The skin was approximated in midline with help of 8 French multiple infant feeding tubes used like simple sutures, instead of sutures to prevent bow string injury of the underlying bowel (as shown in Fig. 10).



Fig.9 Flank skin release incision.



Fig.10 Midline approximation over silastic bag (Urobag) aided by multiple silastic tubes (infant feeding tubes).



Fig.11 Silastic bag removed.



Fig.12 Sequential midline approximation of skin.



Fig.13 Complete midline skin approximation.

Silastic bag was removed two days later followed by sequential approximation of skin by tightening the silastic (infant feeding) tubes (Fig. 11, 12 & 13). Skin release incision wounds received daily saline dressing over Vaseline gauze. Ventilator support was gradually weaned off. Patient was given abdominal binder and mobilized on 4th post operative day. Silastic tubes were removed after 6 weeks (Fig. 14), and skin wounds also contracted (Fig.15). Patient was called after 6 months for definitive abdominal fascial closure.

Post operative chest X-ray showed left lung expanded and one year follow-up showed no recurrence of hernia.



Fig.14 Removal of silastic tubes after 6 weeks.



Fig.15 Skin release incision healing by granulation.

Discussion:

Delayed presentation of diaphragmatic hernia can occur in congenital and secondary to past thoraco-abdominal trauma. Bochdalek hernia is the most common congenital diaphragmatic hernia [1]. Congenital diaphragmatic hernia occurs due to the failure in closure of pleuroperitoneal canal, which usually closes by 8-10 weeks of gestation. The right canal usually closes earlier than the left, leading to more

common occurrence of hernias on the left side. There is a male preponderance of 2:1. As such these hernias present in newborns as respiratory distress, a few cases may present in pediatric age group. There is about 5-30% incidence of the delayed presentation in adults [1].

These hernias may remain asymptomatic throughout life and diagnosed incidentally when abdominal organs are found in the thorax in a chest X-ray or may become symptomatic due to pressure secondary complications like bowel obstruction, incarceration, gastric volvulus and visceral perforation in the chest cavity. The liver, the kidney, and fat comprise right-sided hernias, whereas the left-sided hernias contain the stomach, bowel, the spleen, the pancreas, the kidney, or fat [2]. Chest X-rays with or without bowel contrast and contrast enhanced CT scans of the chest and the abdomen are the investigations of choice to diagnose this condition [2].

Absence of a peritoneal sac around the contents (its incidence varies from 10 to 38%) [3], and its long standing nature, leads to bowel adhesion onto the intrathoracic organs, may necessitate adhesiolysis, for which thoracotomy becomes a better access. Although thoracotomy is the preferred route in chronic traumatic or long standing cases of diaphragmatic hernia, a laparotomy may be needed to access both sides of the diaphragm if a visceral incarceration is suspected [4]. Interrupted non-absorbable sutures with or without using pledgets are used

to close small defects in diaphragm primarily but a larger defect will need a prosthetic mesh patch repair. The size of the mesh to be used will vary according to the defect size. Laparoscopic and thoracoscopic approaches have been used in selected cases [5, 6].

In this case, apart from the huge diaphragmatic hernia and the large defect, the difficult task was to fix the mesh and to accommodate the reduced bowel into the abdomen due to resultant loss of right of domain (LOD) within the abdomen. In such patients having LOD hernias, there is inability to fully accommodate the abdominal contents within the fascial boundaries of abdominal cavity. Closure of the fascia is either impossible, and if at all undertaken, it can lead to fascial dehiscence and high intra-abdominal pressures followed by abdominal compartment syndrome. Awareness on the development of such complications in correction of LOD hernias in emergency situation will warrant the surgeon to leave an open abdomen temporarily followed by staged closure [7, 8]. A good respiratory care, with mechanical ventilation if indicated or through chest physiotherapy and incentive spirometry exercises will prevent respiratory compromise and its complications.

The adoption of customized surgical strategy and aggressive postoperative management, led to a successful outcome in our case. In such situations, an immediate diagnosis to be made followed by emergent operative intervention to prevent further complication like bowel strangulation/gangrene/visceral perforation, associated with high mortality. Although thoracic approach is utilized for delayed presentation of congenital diaphragmatic hernias due to possibility of dense intrathoracic adhesions, an abdominal or a combined thoracoabdominal approach may also be necessary as per the situation. A staged abdominal wall closure allows all reduced bowel to be placed within the abdomen, without the need to sacrifice any viable bowel loop and also avoids complications of an open abdomen like enteroatmospheric fistulas or compartment syndrome following forceful primary closure in such LOD hernias.

The surgical team needs to individualize its management plan while it deals with such type of rare situations that may present when it treats adult diaphragmatic hernias associated with complication, anticipating the possible postoperative challenges that each case may offer.

Conclusion

In our case, hypoplastic left lower lobe, the location of the defect and the absence of a history of trauma in the presence of clinical and radiological findings supported the diagnosis of Bochdalek hernia. In an emergency situation, while treating such LOD hernias when there is inability to accommodate reduced hernia contents into abdomen a staged approach towards abdominal wall closure followed by

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reconstruction should be adopted. This simple, indispensible and innovative method of staged closure of abdomen prevents complications of forceful closure and sequel of open abdomen.

The staged management technique consisted of the following:

Stage I: Bogota bag technique.

Stage II: Bilateral flank skin release incision, partial midline approximation over silastic bag (urobag) with help of silastic tubes. Stage III: Removal of silastic bag followed by sequential tightening of silastic tubes acting as sutures.

Stage IV: Definitive reconstruction after 6 to 12 months (allowing for inflammation and dense adhesion resolution) using the modified components separation technique or retro-rectus PTFE mesh placement.

Limitations:

This is a single case report.

Conflict of Interest: None

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