

Case report:

Unusal Presentation of Gastric Outlet Obstruction – A Rare Case Report

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Abstract:

Congenital Diaphragmatic hernia is seen in 1:2000 to 1:4000 per live births. It is seen more in neonates and children and is seen rarely in adults. Bochdalek's hernia which is left posterolateral is more common than right. We report a case of a 15 year old female patient with upper abdominal discomfort since 2 years and presented with acute gastric outlet obstruction. Computed Tomography of Thorax showed acutely distended luminal organ most probably stomach in the right hemithorax with collapse of right lung. Clinical suspicion, Timely diagnosis and treatment relieved the patient's gastric outlet obstruction and also allowed adequate expansion of the right lung.

Key words: Gastric outlet obstruction, Congenital Diaphragmatic Hernia, Clinical suspicion, timely diagnosis and treatment.

Introduction:

Congenital diaphragmatic hernias are seen in 1:2000 to 1:4000 live births. It is seen rarely in adults^{1,2,3}. Left sided posterolateral diaphragmatic hernias (Bochdalek hernia) are most commonly seen. Most of the adults are asymptomatic or mildly symptomatic with epigastric discomfort or few respiratory symptoms hence misdiagnosed often¹. However here we report a case of congenital diaphragmatic hernia in a young adult with acute gastric outlet obstruction, which is a rare mode of presentation and markedly decreased breath sounds in the right hemithorax making it a unique case which required high degree of suspicion and timely management.

Case Presentation:

15 year old female was referred to our department with symptoms of acute gastric outlet obstruction.

There had been mild Upper Gastrointestinal symptoms like epigastric fullness and had been treated with antacids in past. There was no history of trauma or any surgery in past. On examination, she was mildly dehydrated, had a ryles tube in situ, abdomen was soft and there was remarkable decrease in breath sounds on the right side. Rest of vitals were normal. Routine blood tests showed hyponatremia and hypokalemia. A computed tomography (CT) of abdomen and thorax revealed major part of stomach in the right hemithorax with shift of heart to the left side and right lung was partially compressed (Fig.i). She was hence referred to our department for further management. After adequate resuscitation and routine work up of the patient, she was taken up for surgery. With a double lumen endobronchial tube in situ, a standard posterolateral thoracotomy was done through right

5th intercostal space. During surgery a large part of stomach was found in the right thoracic cavity with dense adhesions to the right lung, pericardium, chest wall and diaphragm causing an acute kink (Fig.ii). Once adhesiolysis achieved, it was found there was no rent in the right diaphragm. On tracing the stomach, a defect was located on the left posterolateral side of the hemidiaphragm measuring approximately 4*4 cm (Fig iii). Stomach was delivered back in the abdominal cavity confirming no twist. Primary closure was done with no 1 non-absorbable monofilament(Fig iv). The procedure was ended with the right thoracic cavity drained by a 2 chest tubes (28F apical and 32 F basal).

Patient was electively ventilated for 24 hours. Patient had a uneventful postoperative recovery and was discharged with adequate expansion of lungs and symptom free. Patient was asymptomatic at 3 months followup.

Discussion:

Bochdalek described Bochdalek hernia in 1848, which is characterized by a congenital defect on the posterolateral region of the diaphragm without hernial sac. They are generally discovered in neonates, but is rarely reported in adults. ^{1,2} .No specific etiological factor for bochdalek hernia has been identified but we know that the occurrence of this disease is due to the failure of closure of the pleuroperitoneal canal during the ninth to tenth week of gestation ^{1,2}

Unlike infants who show with respiratory distress early, the most frequent symptom in adults is mild discomfort and 25% of adult patients are asymptomatic.

This makes the diagnosis of a Bochdalek hernia in adults difficult and hence it is commonly misdiagnosed ^{1,2,3,4}. Consequently, many patients are merely treated according to their symptoms. Similarly our patient had been Upper Gastrointestinal symptoms since 2 years and had

not been investigated prior to acute gastric outlet obstruction.

Among all radiological investigations, a computed tomography (CT) scan allows the highest accuracy for a correct diagnosis. It provides a precise assessment of the anatomical relationships between the viscera and status of the lung in the concerned hemithorax ^{1,2}. Diaphragm of a patient with a Bochdalek hernia is interrupted and has a defect on it ^{1,2}. In our patient there was a 4*4cm defect in the left posterolateral part of the hemidiaphragm confirming it to be a bochdalek hernia intraoperatively. This is rare presentation of a bochdalek hernia as the contents of hernia were on the right side..

The principal management of Bochdalek hernias include reducing the abdominal organs and repairing the defect ^{1,2,4}. Various approaches have been advocated for the repair of these hernias i.e transthoracic approach or transabdominal approach ^{1,2,3,4}, while others advocate video-assisted thoracoscopic or laparoscopic techniques ^{3,4}

Controversy continues with which type of repair would be the best. Surgeons who prefer thoracotomy approach justify with convenience of separating adhesions between thoracic contents and the hernia sac. Though 62% to 90% of Bochdalek hernias do not have hernial sac, there are many a times dense adhesions between the abdominal contents and diaphragm due to the prolonged duration of disease. Surgeons who prefer laparotomy justify that its easier to identify, prevent and deal with possible complications such as malrotation, obstruction, strangulation and perforation of abdominal viscera ^{1,4}. Our patient underwent a thoracotomy due to possibility of adhesions between thoracic contents and pleura and adhesions with diaphragm due to long standing complaints. Small defects can be closed primarily with non absorbable sutures ³. However for larger

defects primary closure is difficult. In these patients, a prosthetic patch is used to achieve closure³. It is sutured to the rims of the orifice with interrupted sutures to avoid excessive tension. For repair of congenital diaphragmatic hernias; Prosthetic materials, including polypropylene mesh (Marlex), polytetrafluoroethylene (PTFE) patch (Goretex), expanded polytetrafluoroethylene (ePTFE) and polyethylene terephthalate mesh (Dacron) have been used quite successfully⁴. Also dura, bovine pericardium, autologous tissues such as fascia or muscle flaps² have been used⁴. We performed primary closure as defect was small and as the content of hernia was on the opposite side there were not many adhesions with left hemidiaphragm.



Fig i: CT Scan : Major part of stomach in Right hemithorax with compression of Right lung

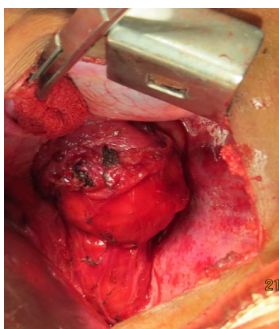


Fig ii: Large part of stomach in Right hemithorax with dense adhesions

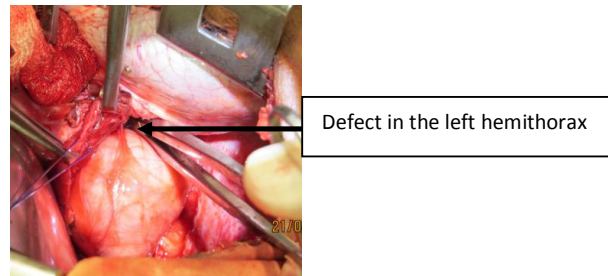


Fig iii: Stomach in Right hemithorax with underlying defect

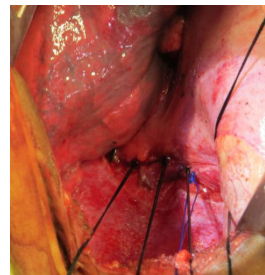


Fig iv : Primary closure of defect with interrupted no 1 non absorbable monofilament sutures

Conclusion:

Bochdalek hernia is a rare entity in adults and a gastric outlet obstruction is a rare presentation. High degree of suspicion needed for timely diagnosis and management. Timely surgical correction with a aggressive postoperative management has a reasonable chance of cure of the patient without additional morbidity.

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