Journal of General Surgery

ISSN: 2688-7479

Case report

Left Diaphragmatic Eventration in an Adult: A Rare Case Report

This article was published in the following Scient Open Access Journal:
Journal of General Surgery
Received December 01, 2021; **Accepted** December 07, 2021; **Published** December 09, 2021

Dr. Yash Patel[1], Dr. Hardik Makwana[2], Dr. Sandeep Kansal[3], Dr. Jay Chokshi[4]

[1]3rd Year Resident, Department of General Surgery, Government Medical College, Surat, India
[2]Assistant Professor, Department of General Surgery, Government Medical College, Surat, India
[3]Additional Professor, Department of General Surgery, Government Medical College, Surat, India
[4]Assistant Professor, Department of General Surgery, Government Medical College, Surat, India

Abstract

Diaphragmatic eventration is a congenital developmental defect of the muscular portion of the diaphragm. It has been attributed to abnormal myoblast migration to the septum transversum and pleuroperitoneal membrane. Diaphragmatic eventration is rare (incidence<0.05%), being more common in male. We present a rare case of 29-year-old male with congenital diaphragmatic eventration presenting an adult. The present case, though quite infrequent. The clinical features included intermittent dyspnea and abdominal pain for 3 months. The left hemithorax presented invasion of fundus of stomach and part of left lobe of liver. The treatment approach was laparoscopic diaphragmatic plication. The outcome was satisfactory. Eventration of diaphragm represents a clinical and diagnostic challenge; clinicians, radiologists and surgeons must be alert with a high degree of suspicion in order to correlate symptoms and imaging findings and understand the etiopathogenesis. In addition, they should plan an adequate and individualized surgical repair making use, as far as possible, of the minimally invasive procedures that are currently used.

Keywords: Eventration of diaphragm, Hemithorax, Diaphragmatic plication, Surgery

Introduction

There are four types of congenital diaphragmatic defects: posterolateral Bochdalek diaphragmatic hernia, Morgagni- Larrey parasternal diaphragmatic hernia, peritoneal-pericardial diaphragmatic hernia, and diaphragmatic eventration [1]. Diaphragm eventration refers to partial or total replacement of diaphragm muscle and fibroelastic tissue causing displacement of hemi diaphragm. The hemidiaphragm may have a normal appearance or variable degree of tissue degeneration, thus forming a translucent membrane without muscle fibres [2]. It consists of the elevation of one, or less likely, both hemidiaphragms causing protrusion of the intra-abdominal viscera to the affected hemithorax. The difference between diaphragmatic hernia and eventration is important; in the latter, there is no true defect. The incidence of diaphragmatic eventration is <0.05% [3].

Diaphragmatic eventration can be congenital or acquired. Congenital eventration is a developmental abnormality secondary to hypoplasia of the homolateral lung or diaphragmatic muscular aplasia leading to cardiorespiratory symptoms. In adults it is mostly because of diaphragmatic palsy through injury to the phrenic nerve that causes dyspnea. This incidence of this anomaly is 1 in 10000 live births. They commonly are presented with respiratory manifestations. Acquired diaphragmatic eventration occurs in older children and adults. There are also some patients without any explained cause of ED which is considered as idiopathic eventration. Idiopathic form frequently affects adults, is the result of a subclinical viral infection and presents commonly with unilateral involvement [25].

Diagnosis is delayed due to no symptoms or very mild ones and is generally done by imaging modalities [25].

The treatment must restore an active and effective contraction and improve the respiratory activity. This is achieved with diaphragmatic plication and can be performed vis thoracotomy or using a minimally invasive laparoscopic or using a minimally invasive laparoscopic or thoracoscopic route, with or without the interposition of a prosthetic material [4-6].

Case presentation

We describe the case of a 29-year-old male presented with chief complain of intermittent dyspnea and occasional epigastric discomfort after meals for 3 months.

***Corresponding author:** Dr Yash Patel, 3rd Year Resident, Department of General Surgery, Government Medical College, Surat, India.

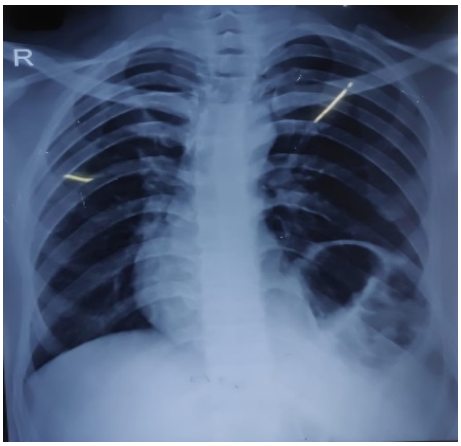

Figure 1: Pre-operative radiological view of both hemithoraces, showing elevation of left hemidiaphragm.

Patient had no previous history of trauma. His BMI was 20 with a blood pressure 132/88 mmHg, heart rate 98 bpm, and oxygen saturation 98% on room air.

Physical examination showed bowel sound involving left hemithorax. Imaging with chest X-ray, computed tomography scan showed the fundus of stomach and partial left lobe of liver inside the left hemithorax; Uniform and continuous diaphragmatic elevation throughout the hemithorax. Left lung was found to be collapsed.

Patient underwent plication of the diaphragm using the laparoscopy abdominal approach. Intra-operatively, the left diaphragm was attenuated, membranous and appeared like a cave. Laparoscopy revealed visceral displacement with elevation of the diaphragmatic dome and there was no diaphragmatic rupture, defined as eventration. Plication was done with multiple continuous strata fix sutures.

Incentive spirometry and deep breathing exercises were started. There were no post operative complications. Post-operative chest radiograph showed the expansion of the hemidiaphragm. The patient stayed in the hospital for 5 days with an adequate resolution of the respiratory symptomatology and the operative wound.

Discussion

Diaphragmatic eventration was first identified by Jean Louis Petit in 1774, the term was first used by Beclard in 1829, and the first surgical repair was described in 1923 by Morrison [2,4,7,8]. Unlike hernia (Bochdalek or Morgagni), there is no disruption of the pleural or peritoneal membrane; however, it is considered a “defect” since anomalous elevation allows invasion of the intraperitoneal viscera into the thoracic space. According to Christensen [2], diaphragmatic eventration is either unilateral or bilateral and complete or partial. Partial presentation is frequent in the anteromedial region of the right hemidiaphragm, whereas complete presentation is commonly found in the left hemidiaphragm [9]. In our patient, a partial eventration was seen in the left part of the thorax.

The pathology shows a translucent membrane of up to almost the normal muscle thickness, with the 3 layers of its structure

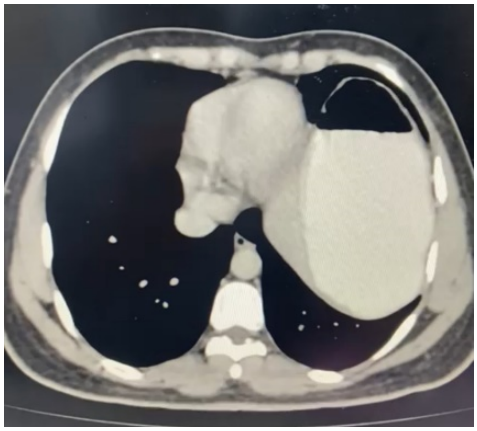

Figure 2: Axial section of both hemithoraces. Fundus of stomach and part of left lobe of liver occupying the left hemithorax.

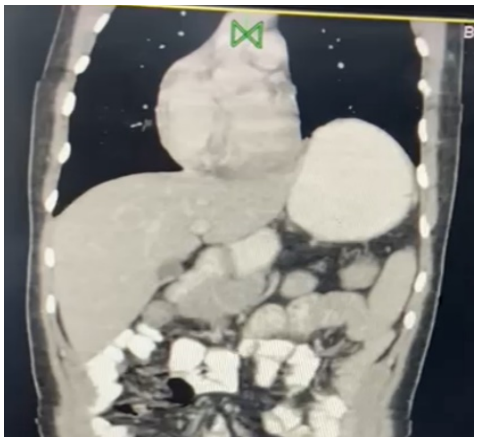

Figure 3: Contrast coronal section shows both hemithoraces. Lung parenchyma is collapsed in left hemithorax and occupied by stomach and left lobe of liver. Normal right hemithorax.

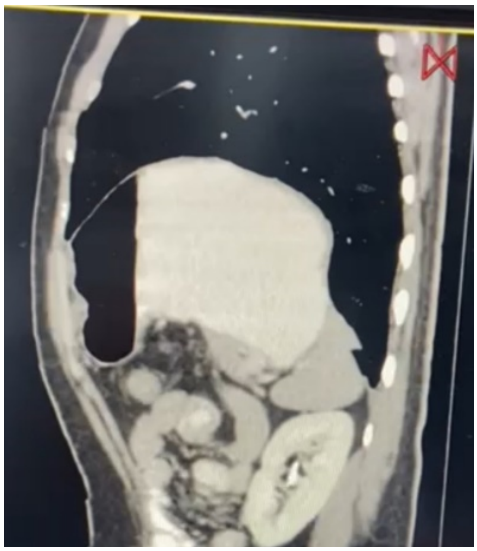

Figure 4: sagittal section of left hemithorax showing occupation by stomach.

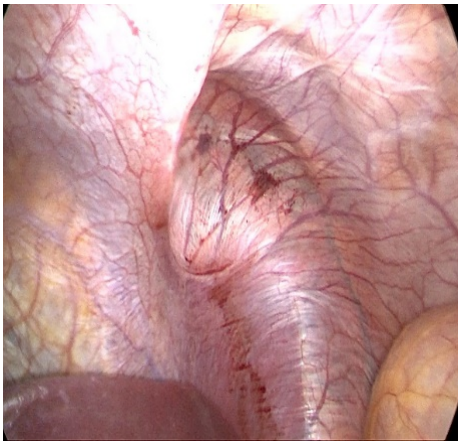

Figure 5: The left hemidiaphragmatic eventration.

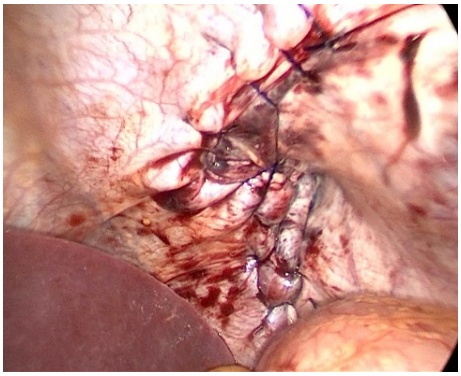

Figure 6: Suture of left hemidiaphragmatic defect.

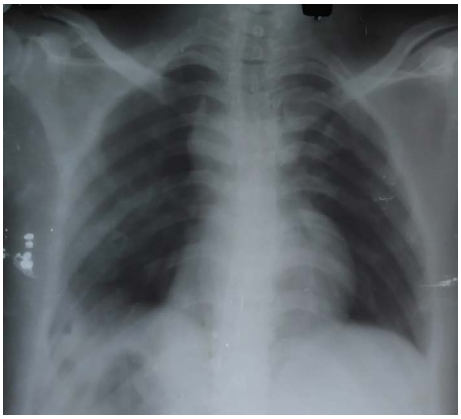

Figure 7: Post-operative radiological view of both hemithoraces, showing left lung expansion.

and normal diaphragmatic inserts. The high diaphragmatic level varies and is not a specific criterion to define the condition. The result is decreased respiratory mobility or paradoxical movement, causing poor lung expansion and hypoxia [10]. The prevalence is 5 per 10,000 births (<0.05%), mostly affecting the male sex, and it is frequent in the left hemidiaphragm [7]. It is a relatively rare anomaly in adults [11]. Groth and Andrade [3] differentiated the congenital etiology of true eventration from the acquired one. The embryological changes involve abnormal migration of myoblasts from the upper cervical somites to the transverse septum (4 weeks of gestation) and pleuroperitoneal membrane (8 to 12 weeks); microscopy shows diffused fibroelastic changes and lack of fibers. Eventration is detected at birth or later according to its symptoms and is associated with prematurity, chromosomal anomalies, and develop- mental defects [12,13].

Paresis or diaphragmatic paralysis is an acquired condition resulting from certain abnormalities that affect the neuromuscular axis between the cervical spinal cord and the diaphragm. This induces a progressive amyotrophy and stretching of all muscular fibers of the dome. Phrenic nerve injury is one of the frequent causes, especially at delivery, followed by neoplastic infiltration, compressions, or surgical interventions [4,14,15]. The lesion of this nerve can be a consequence of neurological pathologies such as myelitis, encephalitis of diverse viral origins, postherpetic neuralgia, polio, tetanus, or diphtheria [4,7].

Diaphragmatic eventration causes loss of compliance of the chest wall and lack of the normal caudal movement of the diaphragm, which are necessary for inspiration, thus altering the ventilation/perfusion ratio. In adults, it is asymptomatic or associated with respiratory symptoms, such as progressive dyspnea, orthopnea, chest pain, palpitations, or severe respiratory distress due to cranial displacement of the diaphragm. The high dome without contraction causes pulmonary collapse causing atelectasis, bronchial or parenchymal infections, or alterations in myocardial excitability due to mediastinum deviation [16]. In infants, the symptoms are severe due to the poor development of the thoracic cage and weakness of the intercostal muscles that cause paradoxical breathing and require the use of mechanical ventilation [3,10]. Digestive symptoms, such as nausea, vomiting, flatulence, abdominal pain, epigastralgia, constipation, anorexia, difficulty in gaining weight, and varying degrees of gastroesophageal reflux, may coexist [17]. The presented case came to our centre with abdominal pain and dyspnea. Pulmonary function tests (PFTs) are important for a dyspneic patient with an elevated hemidiaphragm for highlighting the forced vital capacity and forced expiratory volume in 1 second, showing a restrictive pattern. It is useful to perform the tests in the standing and supine positions. In the supine position, the volumes can decrease to up to 20–50%; nevertheless, these findings do not correlate with the severity of dyspnea, and the tests are mainly used after surgical treatment [3,14].

However, Bochdalek hernia can also be present in the males. Thus, appropriate diagnosis between diaphragmatic hernia and eventration should be made for treatment. The majority of Bochdalek hernia patients presented as acute surgical emergencies and a lack of awareness of this condition lead to an incorrect diagnosis [26]

A chest radiograph shows the elevation of the affected hemidiaphragm; a shadow of the left eventrated hemidiaphragm should be at least one intercostal space raised with respect to the right and 2 intercostal spaces if it is the oppo- site side [10]. Foci of pulmonary condensation, atelectasis, or mediastinal deviations can be observed. Bowel loops can be seen in the thorax if a radiopaque contrast agent is used [7]. Ultrasonography has also been used to visualize the PM of the diaphragm. Both chest radiography and ultrasonography are not useful in the differentiation of eventration and diaphragmatic hernia.

Computed tomography and magnetic resonance imaging can be used to accurately determine the elevation of the dome, the viscera in the intrathoracic position, and associated injuries; tumors can be seen in the base of the pulmonary, cervical, or renal ectopia. Reconstructions in coronal and sagittal planes are useful [11,18]. It is possible to show uninterrupted continuity of the diaphragm to differentiate eventration from hernia, a fact that determines the surgical criteria in case the patient is asymptomatic. Our patient presented this diaphragmatic continuity on computed tomography, which was initially overlooked and required laparoscopic intraoperative detection, as described by Mantoo and Mak [18].

In asymptomatic patients, there are no data comparing the surgical treatment and the conservative approach or the adequate surgical time from the onset of symptoms, especially in the group with phrenic nerve injury; patients with phrenic injury after cardiac surgery may show improvement in the first or second year. Each surgical team establishes its own criteria for surgery. The post-surgery improvement encourages its selection in the majority of patients [19], especially in children, taking into account the lung growth that occurs up to about 10 years. Extreme caution is necessary in patients with morbid obesity or who have neuromuscular disorders [3].

Surgical approaches for eventration include thoracotomy, laparotomy, and thoracoscopy or laparoscopy [1,8]. Total intravenous anesthesia is preferred due to lower cardiovascular and respiratory compromise and minimal effect on hypoxic pulmonary vasoconstriction [20]. The surgical treatment pursues the improvement of ventilation by minimizing dysfunctional diaphragmatic excursion in the process of inspiration [3]; Yalcinkaya et al. [6] achieved this improvement in 97% of the cases. Plication of the diaphragm [3,4,14] improves respiratory mechanics by increasing tidal volume and maximal respiratory capacity. This results in the immobilization of the plicated diaphragm and reduction of the paradoxical movement and contralateral mediastinal involvement [21]. Plication by posterolateral thoracotomy is achieved through the sixth, seventh, or eighth intercostal space, using U points, mattress stitch, continuous suture, or stapling [3]. Resection of the redundant diaphragm and reapproach with overlapping edges have been described, similar to the Mayo technique for umbilical hernia.

Evman et al. [17] reported 42 patients with accordion plication, consisting of several rows of nonabsorbable suture, which are then adjusted to cause "wrinkling" of the diaphragm. The author compared this procedure with the "Mayo technique" that forces the diaphragm to impinge and imbricate the two sheets ("double breasted"). The incision of the diaphragm allows avoiding an intra-abdominal viscera injury below the diaphragm by direct vision. It is advisable not to incise the diaphragm, but there are situations in which it can be done safely and situations in which it is required, such as visceral adhesions under the diaphragm, as per Shah et al. [8].

Thoracoscopy can be performed with two ports and minimum thoracotomy (Gel Port) or using three or four ports; the detailed procedure is described by de Andrade Cordeiro et al. [10]. Yalcinkaya et al. [6] successfully reported on 36 patients who underwent thoracoscopic plication with less pain, shorter stay, shorter operative time, and even less cost, resulting in a better pulmonary compliance of the patient in the postoperative period. It is useful in situations such as pregnancy as it is associated with less morbidity [5]. The disadvantage is the reduction of the operative field, although in a recently published case, a thoracoscopic uniportal access was used for the plication with the help of Endo stitch [22].

Laparoscopy is associated with less pain as it avoids intercostal nerves; both lungs are ventilated for a better visualization and amplitude for manipulation at the time of suturing. Laparoscopic plication is carried out from the posterior to the anterior position and then from the medial to the lateral, of which a detailed description is made by Groth and Andrade [3, 14]. The prosthetic mesh is used in cases of extreme amyotrophy [4] by placing it on the surface of the muscle fixing it in the peripheral diaphragmatic inserts, and preventing recurrence at the ends of the plication; some authors are reluctant to use it due to the possibility of infection and increased cost of surgery, although these criteria are relative, if properly managed. Clifton and Wulkan [23] report the use of PTFE in patients with hernias and eventrations with an excellent description of the operative technique in neonates. The success of plication clinically improves dyspnea, with favorable changes in PFTs of up to 20% according to Evman et al. [17] and Özkan et al. [24] and a favorable follow-up, which is then maintained for up to 2 years. The image shows the abolition of the paradoxical movement, and although the diaphragm can remain motionless, there is no recurrence of symptoms [21]. Plication reports complications with low incidence, pleural effusions, compartment syndrome, and venous thrombosis.

In the present case, the laparoscopic approach was used for diagnosing eventration and not diaphragmatic hernia. Plication was performed by interposing diaphragm with help of strata fix, causing an adequate tension of the hemidiaphragm.

Conclusion

Eventration of diaphragm can have different manifestations. So, the relevant differential diagnosis is very important. Non-traumatic diaphragmatic eventration is rarely diagnosed in adults. In those who have severe signs and symptoms and do not show any response to conservative treatment then the aim of treatment is expansion of intrathoracic space which is done by plication of the diaphragm. Diaphragmatic plication decreases intrathoracic pressure, compression force on the heart and lung, as well as allowing for increased lung function and capacity.

Declarations

- Funding: None
- Conflict of interest: None
- Ethical approval: Not required.

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