ANNALI ITALIANI DI CHIRURGIA

Digital Edition
e-publish on-line
ISSN 2239-253X
Direttore Nicola Picardi

Non traumatic right Bochdalek hernia with loss of Domain in adults.

Our experience and review of literature

Ann Ital Chir, 2023; 12 - Sept. 4
pii: S2239253X23039877
Online Epub

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Non traumatic right Bochdalek hernia with loss of Domain in adults. Our experience and review of literature.

INTRODUCTION: *Right-sided Bochdalek hernia (BH) is a rare developmental defect in the posterolateral diaphragm, allowing herniation of abdominal contents into the thorax. To date only 44 reported cases of right-sided BH have been surgically managed in adults in literature. We report one additional case of right-sided BH with loss of Domain. "Loss of domain" (LOD) is a term used commonly in the hernia literature to describe the distribution of abdominal content between the hernia and residual abdominopelvic cavity. After repairing hernias with significant LOD, serious physiological complications can arise.*
METHODS: *PubMed and Cochrane bibliographical databases were searched (last search: February 2022) for studies concerning BH.*
CASE PRESENTATION: *We report the case of a 50-year-old woman whose right-sided diaphragmatic hernia strangulated loops of small bowel and who was thus treated via urgent laparoscopy. After reduction of the intrathoracic contents we were unable to primarily close the midline fascia.We performed a staged abdominal wall reconstruction as the chronicity of the hernia led to loss of intra-abdominal domain.*
DISCUSSION: *Bochdalek hernia (BH) is the most common type of congenital diaphragmatic hernia and is usually left-sided. It typically presents in neonates and diagnosis in adults is a rarity. Various surgical repair options include open surgery, laparoscopic repair, thoracoscopic approach and robotic transthoracic approaches.*
CONCLUSION: *BH should be managed timely regardless of its symptoms to avoid future complications. The closure of the defect can be done by different methods. When, after diaphragmatic hernia repair, it is suspected that the herniated viscera have lost their domain, it is preferable to use a Temporary Abdominal Closure to prevent compartment syndrome.*

KEY WORDS: Bochdalek hernia with loss of Domain, Bochdalek hernia in adults, Non traumatic Bochdalek hernia, Right-sided diaphragmatic hernia

Introduction

Diaphragmatic hernia is a protrusion of abdominal contents into the chest through a diaphragm defect, which can be either congenital or acquired, usually after blunt trauma [1]. With prevalence of 1.7 and 5.7/ 10 000 births, congenital diaphragmatic hernia is a rare developmental anomaly of diaphragm [2-3]. It develops due to the failure of closure of the posterolateral aspect of pleuroperitoneal canal, which takes place between 8 and 10 weeks of gestation [4].
Congenital diaphragmatic hernias are classified according to their location. Usually, a diaphragmatic hernia involving the posterolateral portion of the diaphragm is called a Bochdalek's hernia (BHD) and is found in 85% of cases [3]. Morgagni's hernia, on the other hand, is a defect

Pervenuto in Redazione Maggio 2023. Accettato per la pubblicazione Luglio 2023
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involving the anterior portion of the diaphragm and accounts for about 2% of cases [5]. Postero- lateral diaphragmatic hernia first described in 1848 by the anatomist Victor Alexander Bochdalek is a congenital anomaly normally diagnosed in the neonatal and postnatal period, while in adults it is extremely rare. Most hernias (80 to 90 per cent) are found on the left side. Right-sided hernias are rarer because the right pleuroperitoneal canal closes earlier and the liver protects the right diaphragm like a 'shield' [6]. There are less than 100 cases of Bochdalek's hernia reported in adults in the literature and less than 44 of these cases involve the right side [7]. Diaphragmatic hernias in adults are relatively asymptomatic, but in some cases they may become symptomatic due to: strangulation of the intestine, intra-abdominal organ dysfunction or severe pulmonary disease. The diagnosis of congenital diaphragmatic hernia is based on clinical investigation and is confirmed by barium X-ray and computed tomography. Surgery is necessary to reposition the intestine in the abdomen and to correct the diaphragmatic defect. We submit a brief review of the literature and a case report of a 50-year-old woman whose right-sided diaphragmatic hernia strangulated loops of small bowel and who was thus treated via urgent laparoscopy. The repositioning of the migrated organs in the abdominal cavity can be particularly difficult because of the ''loss of domain (LOD)'' of the organs, which makes it impossible to close the abdominal wall. "Loss of domain" (LOD) is a term used commonly in the hernia literature to describe the distribution of abdominal content between the hernia and residual abdominopelvic cavity. After repairing hernias with significant LOD, serious complications may arise because increased intra-abdominal pressure pushes up the diaphragm and may be responsible for respiratory insufficiency and pneumonia, and also causes increased tension along the laparotomic incision, which may cause wound complications and hernia recurrence [8].

Embryology

The diaphragm is formed by a musculotendinous wall that divides the coelomic cavity into the abdominal and thoracic cavities, which allows the passage of digestive, vascular, lymphatic and nervous elements. It derives from four embryonic structures (transverse septum, mesoesophagus, pleuroperitoneal membranes and lateral body walls). The pleuroperitoneal membranes may be partially or totally missing. Bochdalek's hernia results from improper fusion of the transverse septum and pleuroperitoneal folds at approximately 8 weeks gestation. As a result of the failure of the diaphragm to develop, intra-abdominal organs invade the thoracic cavity around the 10th week of intra uterine life. Bochdalek's orifice, which tends to be more frequent on the left, is located in the posterolateral part of the diaphragm, outside the pillars, in front of the 10th and 11th ribs. Bochdalek's orifice can vary from an oval defect 2-3 cm in diameter to a loss of substance affecting most of a haemidiaphragm. The anterior border of the orifice, concave posteriorly, is well defined, while the posterior border may be reduced to a simple connective or muscular pad. Its medial border is muscular, while its lateral border may contact the chest wall and be marked only by a slight projection of the serosa. This embryological knowledge plays an important role from a surgical point of view when repairing the defect using a mesh. Since the posterior portion of the diaphragm may sometimes be missing, it may be necessary to anchor the mesh to the rib arch. In about one third of newborns, congenital diaphragmatic hernia (DH) may be associated with pulmonary hypoplasia. The latter is supported by experimental studies of surgically induced congenital diaphragmatic hernia in lamb fetuses, the lungs of which showed abnormalities similar to those found in infants with (DH). However, recent studies in rats have suggested that lung hypoplasia develops independently of the diaphragmatic defect and is already present before normal diaphragm closure. There is a theory, known as the 'two-hit' theory, according to which after an initial event that causes the defect in the lungs and diaphragm, the presence of herniated viscera in the thorax interferes with the foetus' respiratory movements and the further development of the lungs, both at the respiratory and vascular component level. Supporting the hypothesis of a complex embryopathogenesis is the high incidence of associated malformations, present in about one third of congenital (DH) patients. These include cardiac, genitourinary, gastrointestinal, central nervous system, and chromosomal malformations [4-9].

Case Presentation

We describe the case of a 50-year-old woman who presented to the emergency room reporting abdominal pain in the right upper quadrants for about three days. She denied vomiting and previous trauma. Regular bowel movements but tended to be constipated in the last period. On clinical examination the patient reported pain on deep palpation in the right hypochondrium. On fast ultrasonography it was not possible to see the liver in its normal location, but rather completely shifted to the left hypochondrium. Computed tomography of the chest and abdomen showed the presence of a right diaphragmatic hernia, with part of the abdominal viscera having migrated into the thoracic cavity (Figs. 1, 2). The patient was admitted and surgery was scheduled. Due to the deterioration of her clinical condition with increased pain in the right upper abdomen and right hemithorax, the patient underwent urgent laparoscopy 12 hours after her admission. The laparoscopy showed that part of the small intestine, omentum and ascending and transverse colon

were herniated through the diaphragmatic defect (Fig. 3). After releasing the adhesions and removing the hernia sac, the viscera were repositioned in the abdominal cavity. The migration of the intestine and colon into the thorax had over time resulted in the displacement to the left of the liver, which appeared macroscopically normally developed. However, the absence of a posterior anchorage point of the diaphragm made the laparoscopic technique difficult to approximate the diaphragmatic borders and implant the mesh (Fig. 4). Therefore, the approach was converted to laparotomy, where the diaphragm was repaired using interrupted 2-0 polydioxanone sutures and reinforced using a 15x15 cm composite mesh (Figs. 5, 6). The expansion of the right lung was normal after the procedure. A chest drain was inserted on the right. The liver was repositioned in its normal location and the falciform ligament, which appeared slack, was plicated and secured to the peritoneum to avoid possible hepatic rotation in the postoperative period. After returning the intrathoracic contents to its normal location, we were unable to close the midline fascia. We performed a gradual reconstruction of the abdominal wall using a Temporary Abdominal Closure, as the chronicity of the hernia had resulted in a loss of the intra-abdominal viscera domain. The patient was transferred to the intensive care unit and kept intubated. Every three days we performed dressing, with gradual reapproximation of the fascia, until complete closure of the laparotomy. After being extubated and the chest drain removed, the patient was discharged in good clinical condition on 20 post-operative days.

The CT (Temporary Abdominal Closure) we used incorporates some concepts from the Vacum Pack/Bogotá Bag technique, Mesh closure and Wittmann Patch (Fig. 7) [10].

To protect the abdominal viscera, a porous biologically inert plastic sheet (V.A.C. GranuFoam Dressing, Fig. 7a) is placed on top of them, which prevents the formation of adhesions with them and facilitates drainage; at this point, before using the sponge directly (typical step of the Vacuum Pack technique), we apply a mesh (Fig. 7b). There are different types of mesh material, either resorbable (such as Vicryl or Dexon), or non-resorbable (such as polypropylene or Dacron). Normally we use a Vicryl mesh and anchor it to the fascia (Fig. 7c) in order to prevent its retraction.

The choice of this material is related to its lower rate of enterocutaneous fistula formation. This method allows easy access to the abdomen for surgical re-exploration (by simply opening the mesh medially without detaching it from the fascia) and facilitates the closure of the abdomen with accurate serial mesh reductions.

At this point we apply the mouldable open mesh sponge (Fig. 7d), which has a triple function, to evenly distribute the sub-atmospheric pressure, to allow efficient drainage of fluids and to ensure direct and complete contact with the wound bed. Finally, the abdominal wall is sealed with an adhesive cloth (Fig. 7e), on which a small breach is made to apply a valve to allow the passage of the drainage tube (Fig. 7f) and guarantee the hermetic seal of the dressing.

At the level of the sponge is a baroreceptor, which measures the pressure in the open abdomen by sending feedback to the third element of the system, a computerised pump. This pump is connected to the drainage tube to which it applies negative pressure that causes the sponge

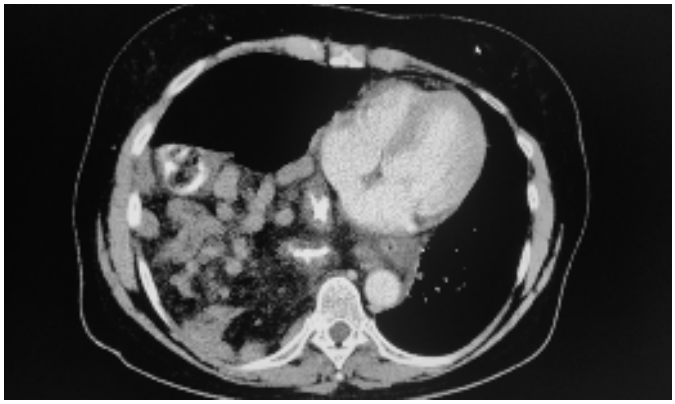

Fig. 1-2: CT thorax-abdomen view demonstrates a right Bochdalek hernia. There is herniation of part of the small intestine, omentum and ascending and transverse colon through a diaphragmatic defect, Bochdalek foramen, in the posterolateral right hemidiaphragm.


FIG. 3

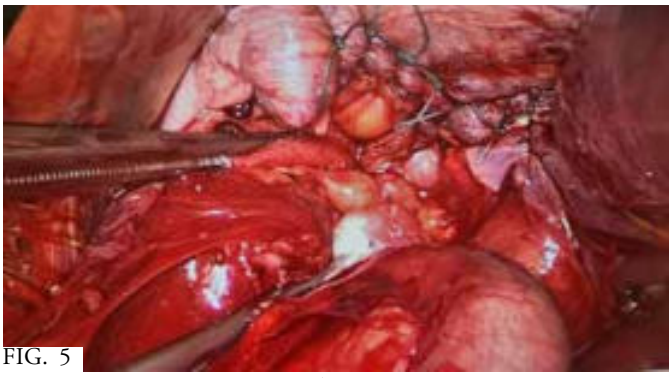
FIG. 5

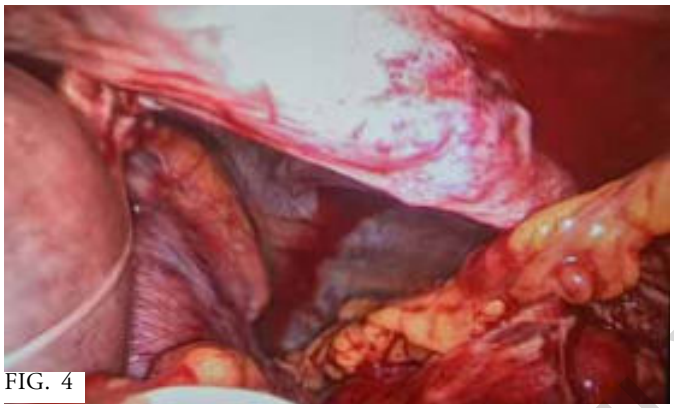
FIG. 4


FIG. 6

Figs. 3-4-5-6: Intraoperative findings before and after hernia repair.

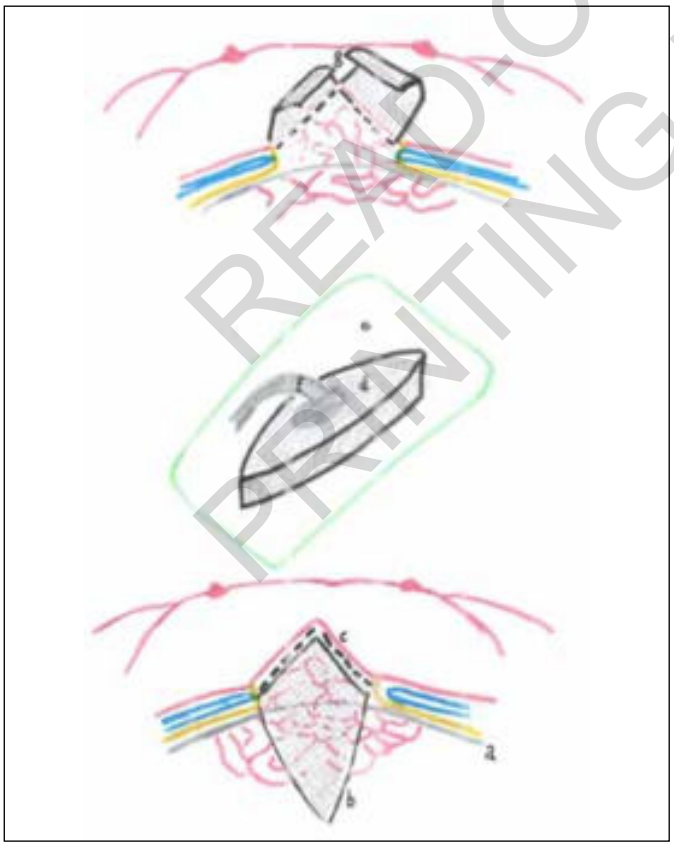

Fig. 7: (a-g) Temporary abdominal wall closure technique.

to collapse, exerting medial traction approximating the fascia and abdominal wall [11].

We start by applying a negative pressure of approximately 125 mmHg, which stimulates cell reproduction and minimises the negative effects of oedema. The dressing should be changed every 2-3 days [10].

At each dressing after disconnecting the pump and removing the sponge, access to the abdominal cavity is provided by opening the mesh medially (Fig. 7g). After washing the cavity and replacing the abdominal plastic sheet, the mesh is closed not by direct suturing but by overlapping the two flaps in such a way that at each change the bandage is brought about one cm closer together. In our experience on average after 3-4 dressings we are able to reapproximate fascia defects of about 5 cm.

The last step involves the dressing and mesh being completely removed and the fascia being sutured with interrupted sutures. In cases where it is not possible to approximate the fascia directly, we resorted to component separation techniques (Rives Stoppa or Posterior Componenet Separation) using mesh.

Discussion

We report a rare case of right diaphragmatic hernia in a 50-year-old woman with no previous history of trau-

ma. Clinical examination and imaging of the chest and abdomen revealed the presence of loops of small intestine and colon in the right hemithorax. After being admitted, there was a rapid worsening of the clinical condition which led to the need for urgent laparoscopy. Abdominal viscera can migrate into the thoracic cavity either as a result of diaphragmatic rupture after trauma or due to congenital defect of the diaphragm and as we have seen are more common on the left side [12]. It is extremely rare to detect a right-sided diaphragmatic hernia in an adult without trauma because the right pleuroperitoneal canal closes earlier and the liver protects the diaphragm from possible herniation [13].

The diagnosis of traumatic rupture of the diaphragm can sometimes remain elusive despite a variety of imaging options, due to the absence of symptoms at the time of injury [14].

In most patients with congenital diaphragmatic hernia (DH) this is diagnosed either in early childhood or immediately after birth. Neonatal Bochdalek's hernia frequently impairs cardiopulmonary function and carries a high mortality rate. In contrast to congenital DH, adult-onset hernias usually manifest as gastrointestinal symptoms rather than pulmonary symptoms. Abdominal pain was reported in 62% of cases and thus considered the most frequent complaint [15]. Due to the non-specific symptomatology of ADH and its rarity, diagnosis can be delayed, resulting in serious complications such as incarceration and strangulation.

Carter et al. described the four stages in the development of strangulating diaphragmatic hernia, and they believe that it is important for clinical doctors to recognize each of these four stages in the eventual development of a diaphragmatic hernia [16]: (1) asymptomatic, (2) minimal symptoms, (3) obstruction, and (4) strangulation. The stage of obstruction is characterized by severe upper abdominal or lower thoracic pain, nausea, and vomiting. In the fourth stage, the tension of the symptoms is increased, and a surgical intervention is necessary.

Most of the patients develop the last complication (strangulation) weeks, months, or even years after the first diagnosis of a congenital or posttraumatic diaphragmatic hernia, as reported by Carter et al. [16]. Due to the nonspecific symptomatology of ADH and its rarity, diagnosis may be delayed, resulting in serious complications such as incarceration and strangulation.

When symptomatic, they can lead to often positional postprandial pain, vomiting, dysphagia and sometimes a full-blown occlusive syndrome and should be differentiated from other forms of intestinal obstruction in children [17].

Sometimes symptoms may occur due to the onset of complications in the herniated viscera (ulcer, perforation or gastric volvulus, colopleural fistula, haemorrhage), with a high mortality often associated with delayed diagnosis. Bochdalek's hernia may accompany several types of congenital anomalies, such as common mesentery, situs inversus, intestinal malrotation (IM), gastric volvulus and liver malformation [18,19].

Hypoplasia or atrophy of the right lobe of the liver can facilitate herniation of abdominal organs through the discovered defect and is frequently observed in Bochdalek's hernia of the right side. The malformation of the right lobe of the liver may be the consequence of impaired portal perfusion due to the long-standing compression effect of the dislocated organs.

The absence of breath sounds and the presence of bowel sounds in the chest are typical findings associated with diaphragmatic hernia [20].

In most cases, the disorder is detected unexpectedly on chest X-ray [1]. Due to the low sensitivity of chest X-ray, diaphragmatic hernias can be confused with other thoracic pathologies, including hypertensive pneumothorax, pericardial fat pad, mediastinal lipoma or anterior mediastinal mass [1,21]. The gold standard technique for diagnosis is computed tomography, which allows clinical physicians to assess the size, location and type of diaphragmatic hernia [22].

Two studies revealed that computed tomography has a sensitivity of 78% for left-sided hernias and 50% for right-sided hernias [1,21].

Chest X-ray and especially CT scans, possibly with digestive tract opacification, allow the diagnosis to be made by showing the presence of loops of the small intestine and colon in the chest. The posterolateral location of the breach allows the congenital nature of the hernia to be confirmed. According to the literature, in adults, less than 100 cases of posterolateral hernia are documented, and less than 20 cases are right-sided [23].

The most common contents include the colon (63%), stomach (40%), omentum (39%), and small bowel (28%). The spleen, tail of the pancreas, and kidney are the unusual contents [24].

Surgical repair options include the open technique, laparoscopy, thoracoscopy and the robotic approach. The best approach has yet to be determined with further large-scale studies. Campos and Sipes [25] performed the first laparoscopic repair of diaphragmatic hernia in 1991. Kuster et al. followed in 1992 [26]. So far, only small case series and case reports are available in the literature. In the past, an operative approach was typically used based on the chronicity of the condition, as the longer the hernia was, the more tenacious adhesions were believed to be present that were difficult to free via transabdominal incision and therefore a transthoracic approach was preferred. However, recently published experience in the use of the abdominal approach has shown its feasibility and safety, as demonstrated in a retrospective study by Payne et al. who reported a higher mortality rate with the thoracic approach than with the abdominal approach with a lower incidence of pneumonia compared to patients undergoing the classic transthoracic approach [27].

Kishore et al. reported a complication-free DH repair via laparotomy in 88% of their series [28]. With advances in minimally invasive techniques, laparoscopic repair has been tried and proven to be a viable option for DH repair. Although shorter hospital stay and quicker recovery are reported advantages of the laparoscopic technique, concerns regarding visceral injury and recurrence rates were initially present.

Liao et al. reviewed a total of 36 DH cases that were repaired laparoscopically, with no reported recurrence (mean follow-up was 15.1 months) [29]. Technical difficulties in the laparoscopic approach requiring conversion to open surgery have been reported in the literature, such as in our case, where the absence of the posterior flap of the diaphragm required a mesh suture at the costal arch, which is easier and safer to perform with the classical technique .

There are insufficient data on which to base a recommendation on repair methods; while the majority of reported cases used the primary suture technique, 40% used mesh alone or in combination with diaphragmatic flap suturing [29]. Primary repair is considered the method of choice in traumatic/acute settings, as reported by Kishore et al., with only 11% of cases requiring the use of mesh [28]. Defect closure can be performed by several methods. Defects smaller than 5 cm are repaired by interrupted sutures with 2-0 polydi-oxanone (Reider technique). Defects larger than 5 cm are repaired using tension-free mesh hernioplasty . When the defect is small it can simply be sutured, but when it is large (> 10 cm square) it will need a reinforcing prosthesis . There is a lack of sufficient evidence in favour of a particular type of mesh [30]. ACS (abdominal compartment syndrome) was first described more than a century ago, mainly found in the repair of gastroschisis and omphalocele. Conditions such as BDH and long-standing ventral hernias are associated with increased intra-abdominal pressure >25-30 mmHg due to insufficient space in the abdominal cavity to accommodate the volume of all migrated abdominal organs. As pressures exceed 25 mmHg, with noted organ dysfunction, surgical decompression is indicated urgently. ACS causes pressure-related organ failure. Presenting signs of ACS include a firm tense abdomen, increased peak inspiratory pressures, and oliguria, all of which improve after abdominal decompression. The authors recommended that when ACS is suspected, the abdomen could be repaired with a constructed silo of prosthetic material, Wittmann patch, or vacuum-assisted closure system, and following fluid shifts and abdominal wall stretching for several days, the abdomen could be closed.

Conclusion

Bochdalek hernia (BH) is a developmental defect in the posterolateral diaphragm, allowing herniation of abdominal contents into the thorax .Due to the non-specific symptomatology of ADH and its rarity, diagnosis may be delayed, resulting in serious complications such as incarceration and strangulation.

Treatment of a Bochdalek hernia is surgical, achieved by utilizing either open repair or laparoscopic techniques, to close the defect.The closure of the defect can be done by different methods.

When the defect is small it can be simply sutured closed, but when it is large (>10 cm square) it will need a prosthetic reinforcement . Sufficient evidence favoring any particular type of mesh is lacking . When, after diaphragmatic hernia repair, it is suspected that the herniated viscera have lost their domain, it is preferable to use a Temporary Abdominal Closure to prevent compartment syndrome.

Riassunto

INTRODUZIONE: L'ernia di Bochdalek del lato destro (BH) è un raro difetto dello sviluppo della porzione posterolaterale del diaframma, che consente l'erniazione di parte del contenuto addominale nel torace. Ad oggi solo 44 casi di BH destra nell'adulto sono stati descritti in letteratura, tutti gestiti chirurgicamente. Segnaliamo un ulteriore caso di BH destra ma con una particolarità, infatti i visceri erniati avevano perso il diritto di domicilio.

METODI: sono stati cercati i database bibliografici in PubMed e Cochrane (ultima ricerca: febbraio 2022) per studi riguardanti BH.

PRESENTAZIONE DEL CASO: Riportiamo il caso di una donna di 50 anni con ernia diaframmatica destra strozzata che aveva richiesto una laparoscopia urgente.

Dopo la riduzione del contenuto intratoracico, a causa della perdita di domicilio dei visceri erniati, non siamo stati in grado di chiudere la fascia e quindi abbiamo eseguito una ricostruzione graduale della parete addominale.

DISCUSSIONE: L'ernia di Bochdalek (BH) è il tipo più comune di ernia diaframmatica congenita e di solito coinvolge il lato sinistro.

Si presenta tipicamente nei neonati e la diagnosi negli adulti è rara. Varie opzioni di riparazione sono a disposizione del chirurgo tra cui la tecnica open, laparoscopica, toracoscopica e robotica.

CONCLUSIONE: BH dovrebbe essere gestita tempestivamente indipendentemente dai suoi sintomi, per evitare l'insorgere di possibili complicanze. La chiusura del difetto può essere effettuata con diversi metodi.

Quando, dopo la riparazione dell'ernia diaframmatica, si sospetta che i visceri erniati abbiano perso il loro domicilio, è preferibile utilizzare una chiusura addominale temporanea per prevenire la sindrome compartimentale.

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