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A review of Morgagni and Bochdalek hernias in adults

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The incidence of Bochdalek and Morgagni hernias among adults is very rare. The purpose of this study was to determine retrospectively the prevalence and characteristics of adult Bochdalek and Morgagni hernias in a decade. Consequently, we demonstrated 12 patients with Bochdalek and 8 patients with Morgagni hernias. We presented plain radiography, operation images, and computed tomography findings of an adult patient with symptoms due to Bochdalek and Morgagni hernias. In surgical repair, the Morgagni hernia is best approached via laparotomy, and the Bochdalek hernia can be treated through thoracotomy or laparotomy. (Folia Morphol 2011; 70, 1: 5–12)

Key words: congenital diaphragmatic hernias, Bochdalek hernia, Morgagni hernia, adult

INTRODUCTION

The topic of congenital diaphragmatic hernia (CDH) has frequently appeared in medical literature since its first description in the early 18th century. CHD is a term applied to a variety of congenital birth defects that involve abnormal development of the diaphragm. Congenital diaphragmatic hernia occurs in 1 out of every 2000–3000 live births and accounts for 8% of all major congenital anomalies. The risk of recurrence of isolated congenital diaphragmatic hernia in future siblings is approximately 2%. Familial congenital diaphragmatic hernia is rare (< 2% of all cases), and both autosomal recessive and autosomal dominant patterns of inheritance have been reported. Congenital diaphragmatic hernia is a recognized finding in Cornelia de Lange syndrome and also occurs as a prominent feature of Fryns syndrome, an autosomal recessive disorder with variable features including diaphragmatic hernia, cleft lip or palate, and distal digital hypoplasia [22].

The three basic types of congenital diaphragmatic hernia include Bochdalek hernia (BH), anterior Morgagni hernia (MH), and hiatus hernia. Congenital hernias resulting from a developmental failure of posterolateral diaphragmatic foramina to fuse properly were first described by Czech anatomist Vincent Alexander Bochdalek in 1848 [60], although the origins of descriptions of diaphragmatic hernia can be dated to writings from as early as 1690 [21]. Diaphragmatic hernias through the posterolateral foramen of Bochdalek represent the commonest type of congenital diaphragmatic hernia [1]. The majority are present during neonatal life and have a poor prognosis, being associated with congenital pulmonary abnormalities [53, 66]. The left-sided BH occurs in approximately 85% of cases. Left-sided hernias allow herniation of both the small and large bowel and intra-abdominal solid organs into the thoracic cavity. In right-sided hernias (13% of cases), only the liver and a portion of the large bowel tend to herniate. Bilateral

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hernias are uncommon and are usually fatal [31]. Presentation of a BH in an adult is exceptionally rare. In 1959 Kirkland published the first review of 34 cases of adult BH, and as of 1992 only 100 cases of symptomatic adult BH have been reported in world literature [40]; however, with the growing use of abdominal CT this abnormality is being increasingly detected in asymptomatic individuals.

Morgagni hernia, rarely seen anterior defect of the diaphragm, is variably referred to as a Morgagni, retrosternal, or parasternal hernia. It was first described by the Italian anatomist and pathologist Giovanni Morgagni in 1769 [10]. Accounting for approximately 2% of all CDH cases, it is characterised by herniation through the foramina of Morgagni, which is located immediately adjacent to the xiphoid process of the sternum [65]. The majority of hernias occur on the right side of the body and are generally asymptomatic; however, new-borns may present with respiratory distress at birth similar to that found in Bochdalek hernias. Additionally, recurrent chest infections and gastrointestinal symptoms have been reported in those with previously undiagnosed Morgagni hernias [19, 64].

We reported 12 cases of Bochdalek hernias and 8 cases of Morgagni hernias. The aim of this report was to present rare cases of adult presentation of Bochdalek and Morgagni hernias, and to discuss the clinical presentation and management of these rare cases. Because there are limited reviews of these rare hernias in the literature, the majority of these are single patient case reports, but we consider that this presentation may provide useful data for surgeons.

MATERIAL AND METHODS

A retrospective analysis of all patients with Morgagni and Bochdalek hernias in adults diagnosed and treated at the Departments of General and Thoracic Surgery from 2000 to 2010 was carried out. All patients had an absence of trauma history. In total, 12 patients with Bochdalek hernias (age range 16–58 years) and eight patients with Morgagni hernias (age range 42–71 years) were surgically treated. To obtain a clinical history of the patients in whom Bochdalek and Morgagni hernias were identified individually, we performed a chart review for each patient including Bochdalek (Table 1) and Morgagni hernias (Table 2), noting the patients' age, sex, admission to hospital, site of hernia, symptoms, contents of hernial sac, width of hernial sac, operative treatment, and hospital stay, respectively.

Radiological evaluation

For this analysis, the patients' thoracic and abdominal CT scans were included as a routine protocol. We only demonstrated CT scans and operation images of selected patients with reports indicating both Bochdalek (Figs. 1A, B) and Morgagni hernias (Figs. 2A, B, C) individually. As a result, 10 male and 2 female patients indicating Bochdalek hernias were presented. On the other hand, 4 men and 4 female patients indicating Morgagni hernias were presented.

RESULTS

Bochdalek hernias

Clinical symptoms included dyspnoea, cough, wheezing, thoracic and abdominal pain, ileus, and recurrent chest infections. We determined left-sided hernia in 10 patients and right-sided hernia in 2 patients. The mean age of the patients was 36.5 years. In right-sided Bochdalek hernias, the contents were hepar, ren, colon transversum, and omentum majus. In the left-sided Bochdalek hernias, the contents were gaster, caecum, appendix vermiformis, colon ascendens and transversum, intestinum tenue, omentum majus, and ren. The mean area of the hernial sac was 34.8 square centimeter. Our surgical approach was determined according to the individual criteria of each patient. A female patient in our series was presented as a case report (Patient no. 3, Table 1) [24]. Her chest radiograph and CT revealed a large left-sided BH. Intestinal organs, containing bowel, small intestine, caecum, and appendix, were seen in the left hemithorax. We operated on her via thoracotomy and laparotomy. She had a left-sided BH with concomitant partial situs inversus. Furthermore, the right side of the abdominal cavity was empty. Bochdalek hernia with concomitant partial situs inversus has not been reported before in medical literature [24]. A 16-year-old male was admitted to the outpatient clinic with dyspnoea and recurrent chest infection (Patient no. 11, Table 1). Computed chest tomography was performed and revealed left-sided BH. Gaster was in the contents of the hernial sac (Fig. 1A). The width of the hernial sac was 6 × 4 cm (Fig. 1B). Finally, we operated on him via thoracotomy (Fig. 1B). The rest of the patients were operated on via laparotomy. In

Table 1. Individual presentations for Bochdalek hernia

The chart of Bochdalek hernias									
Patient no.	**Age**	**Sex**	**Admission to hospital**	**Site**	**Symptoms**	**Content of hernial sac**	**Width of hernial sac [cm]**	**Operative treatment**	**Hospital stay**
1	50	M	Emergency	Right	Dyspnoea Cough Wheezing	Hepar Ren Colon transversum Omentum majus	7 × 5	Laparotomy Prolene mesh	9
2	35	M	Emergency	Left	Ileus	Hepar Colon transversum Intestinum tenue	8 × 5	Laparotomy Primary closure	18
3	44	F	Elective	Left	Dyspnoea Chest pain Wheezing	Omentum majus Colon ascendens Colon transversum Intestinum tenue Caecum Appendix vermiformis	12 × 7	Thoracotomy Laparotomy Primary closure Prolene mesh	27
4	58	M	Elective	Left	Dyspnoea Chest pain	Colon transversum Omentum majus	5 × 5	Laparotomy Primary closure	13
5	38	F	Elective	Left	Chest pain	Omentum Ren	8 × 4	Laparotomy Primary closure	8
6	41	M	Elective	Left	Chest pain Dyspnoea Cough	Ren Omentum majus Splen	8 × 6	Laparotomy Primary closure	10
7	29	M	Elective	Left	Dyspnoea Recurrent chest infection	Omentum majus Colon transversum	5 × 6	Laparotomy Prolene mesh	12
8	26	M	Emergency	Left	Abdominal pain Ileus	Omentum majus Colon transversum	4 × 3	Laparotomy Primary closure	9
9	31	M	Elective	Right	Dyspnoea Chest pain Wheezing	Hepar Omentum majus Colon transversum	4 × 7	Laparotomy Primary closure	14
10	47	M	Elective	Left	Dyspnoea Chest pain Abdominal pain	Omentum majus Hepar	9 × 5	Laparotomy Prolene mesh	21
11	16	M	Elective	Left	Dyspnoea Recurrent chest infection	Stomach	6 × 4	Thoracotomy Primary closure	12
12	23	M	Emergency	Left	Dyspnoea Abdominal pain	Omentum majus	3 × 5	Laparotomy Primary closure	13

addition to these operative treatments, diaphragmatic defects were strengthened via primary closure or prolene mesh. All of our patients were discharged in good health.

Morgagni hernias

Clinical symptoms included dyspnoea, cough, epigastic pain, ileus and subileus, chest and abdominal pain, and recurrent chest infections. All patients had an absence of trauma history. We determined left-sided hernia in 7 patients and right-sided hernia in 1 patient. The mean age of the patients was 59.5 years. In right-sided Morgagni hernias, the contents were colon transversum, intestinum tenue, and omentum majus. In the left-sided Bochdalek hernias, the content was only omentum majus. The mean area of the hernial sac was 19.12 m^2. A 42 year-old man was admitted to the outpatient clinic with lower chest discomfort, dyspnoea, and cough (Patient no. 2, Table 2). Computed chest tomography was performed revealing a right-sided MH, localization of omentum majus (Fig. 2A). When we performed laparotomy we ob-

Table 2. Individual presentations for Morgagni hernia

The chart of Morgagni hernias									
Patient no.	**Age**	**Sex**	**Admission to hospital**	**Site**	**Symptoms**	**Content of hernial sac**	**Width of hernial sac [cm]**	**Operative treatment**	**Hospital stay**
1	70	F	Elective	Right	Epigastric pain Subileus	Colon transversum Omentum majus	5 × 3	Laparotomy Primary closure Prolene mesh	9
2	42	M	Elective	Right	Dyspnoea Chest pain Cough	Omentum majus	4 × 3	Laparotomy Primary closure	6
3	58	M	Emergency	Right	Ileus Abdominal pain	Colon transversum Omentum majus	6 × 4	Laparotomy Primary closure	14
4	68	F	Elective	Right	Dyspnoea Cough	Omentum majus	6 × 3	Laparotomy Primary closure	7
5	71	M	Elective	Right	Dyspnoea	Omentum majus	7 × 4	Laparotomy Prolene mesh	9
6	55	F	Elective	Right	Dyspnoea Abdominal pain	Colon transversum Omentum majus	5 × 4	Laparotomy Primary closure	8
7	59	F	Elective	Right	Dyspnoea Recurrent chest infection	Omentum majus Intestinum tenue	7 × 3	Laparotomy Prolene mesh	10
8	53	M	Elective	Left	Abdominal pain	Omentum majus	5 × 3	Laparotomy Primary closure	7

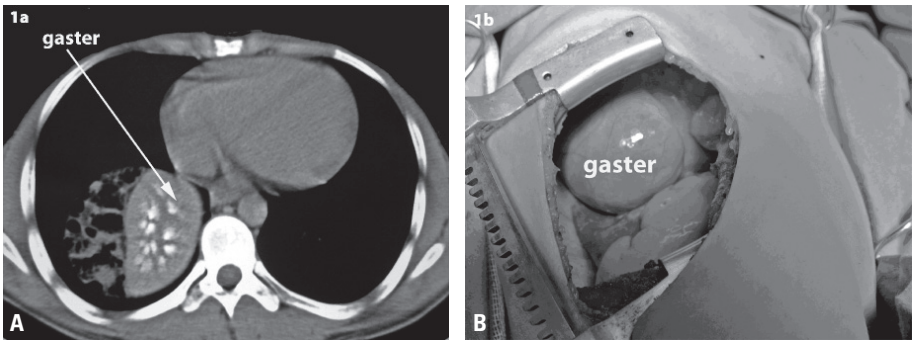

Figure 1 A. Computed tomography of the abdomen, showing a left-sided Bochdalek hernia; **B.** Intra-operative image of the abdominal cavity showing content of Bochdalek hernia sac.

served that omentum majus was in the contents of the hernial sac (Fig. 2B). The width of the hernial sac was 5 × 3 cm (Fig. 2C). All our patients were operated via laparotomy. In addition to these operative treatments, diaphragmatic defects were strengthened via primary closure or prolene mesh. All our patients were discharged in good health.

DISCUSSION

We evaluated retrospectively the patients with Bochdalek and Morgagni hernias for the last decade in the Department of General Surgery. Congenital diaphragmatic hernias clinically presenting in adulthood are exceedingly rare lesions [39, 53]. They can occur through an anterior parasternal foramen (Morgagni) or through a posterolateral, mainly left-sided defect (Bochdalek) representing persistence of the pleuroperitoneal canal. The location of the foramina of Bochdalek is defined by the location of the diaphragmatic coronary ligaments bilaterally. Bochdalek hernias occur when these soft-tissue anastomoses fail to close or when they reopen. If the herniation is present from the time of birth, it is termed "congenital". If the herniation forms later, perhaps because of extension of intra-abdominal or perirenal fat into the thorax, it is termed "acquired".

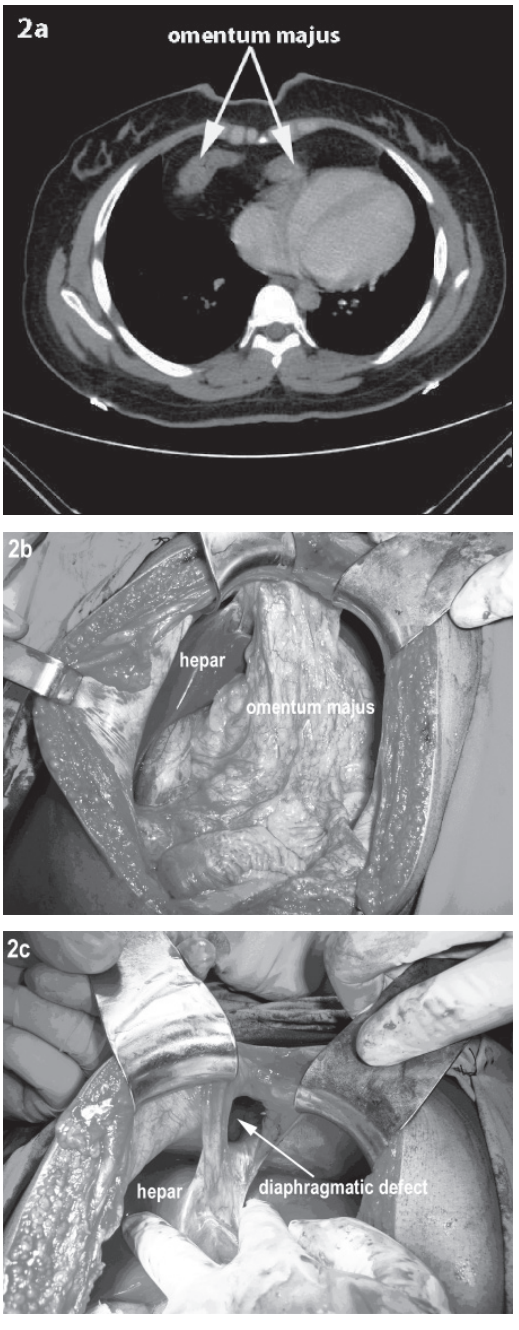

Figure 2. A. Computed tomography of the abdomen, showing a right-sided Morgagni hernia; **B.** Intra-operative image of the abdominal cavity showing content of Morgagni hernial sac; **C.** Intra-operative image of the abdominal cavity showing Morgagni hernia above the liver.

Acquired hernias are also called "incidental" or "subacute" hernias.

Bochdalek hernias most commonly manifest during the patient's first few weeks of life. Diagnosis beyond the first 8 weeks of life is estimated to represent 5–25% of all Bochdalek hernias [50]. In the neonate, Bochdalek hernias are one of the leading causes of respiratory distress and remain one of the most common congenital anomalies of the thorax [59]. Most neonatal Bochdalek hernias are left-sided [37].

Presentation of a BH in an adult is exceptionally rare. The overall prevalence of asymptomatic BH in adults is 6% [67]. From all patients with a congenital BH, only 5% will be diagnosed in childhood or adulthood [56]. Similarly, in a review of 940 consecutive chest and abdominal computed tomographic scans obtained at a university medical centre in 1984, a 6% prevalence of BH was reported [23].

The clinical symptoms of diaphragmatic herniation are frequently vague and nonspecific, including chest pains, dyspnoea, and gastrointestinal complaints [35], abdominal pain, nausea and vomiting, constipation or respiratory distress [56, 58], chest pain, dyspnoea, and wheezing symptoms, followed by severe attacks and episodes of incarceration with serious consequences. In our study, we observed similar clinical symptoms of Bochdalek herniation, as seen in Table 1. Characteristically, these symptoms can be intermittent as herniated viscera can spontaneously reduce, causing symptom regression. In such cases, radiological investigations demonstrate reduction of the hernia with symptom resolution [53]. Others will present with serious complications associated with strangulation of herniated viscera, especially when the diagnosis has been missed or treatment delayed [51]. There have been reports of BH presenting with sudden death from intrathoracic complications [60]. Gastric volvulus is one of the rare but recognized complications of BH [12, 58]. Presentation with severe symptoms has been reported in 46% of cases and the mortality in these has been high (32%) because of visceral strangulation [20]. The incidence of hernia with peritoneal sac varies from 10 to 38% [60]. In right-sided Bochdalek hernias, the contents are predominantly the liver, the kidney, and fat. A left-sided hernia may contain enteric tract, the spleen, the liver, the pancreas, the kidney, or fat. Colon-containing hernias are rare and usually occur through left-sided defects [8]. In our series, we determined left-sided hernia in 9 patients and right-sided hernia in 3 patients. Although colon-containing hernias are rare, usually occuring through left-sided defects [8], we confirmed that seven of twelve patients (58%) had colon-containing hernias. And also, five of these seven patients (71%) had left-sided defects, and the rest (29%) had right sided-defects. Regardless of hernial sac, 9 patients (75%) with

left-sided hernia and 3 patients (25%) with right--sided hernia were determined.

Computed tomography is known to be the most accurate method of diagnosing and evaluating the contents of Bochdalek hernias, especially the smaller ones [62, 67]. The current treatment of choice of a BH is surgical repair, even in asymptomatic cases, because of the risk of visceral herniation and strangulation [48]. The surgical approach may be via a thoracotomy, laparotomy, or a combination of the two. We only operated one patient via thoracotomy and laparotomy [24]. Ten of twelve patients were operated via laparotomy, and one of twelve patients was operated via thoracotomy. In addition to these operative treatments, diaphragmatic defects were strengthened via primary closure or prolene mesh.

Of all the types of CDH, Morgagni hernias are relatively rare. They arise from a septum transversarium defect due to the failure of closure of the pars sternalis with the seventh costochondral arch [16, 25, 54]. Comer and Clagett [17] reported 54 patients with MH in a series of 1750 patients with diaphragmatic hernias. Similarly, Berman et al. [6] reported on 18 cases with MH over a period of 20 years. Recently, Kilic et al. [34] collected their data of 16 patients during a 16-year period. This defect also is referred to as the space of Larrey, after Napoleon's surgeon, who described the retrosternal space as an avenue through which pericardial tamponade could be treated [17]. Some authors refer to the potential retrosternal space on the right as "Morgagni's gap" and the space on the left as "Larrey's gap" [63]. In medical literature, this hernia was presented titled as Morgagni-Larrey hernia [4, 13, 14, 27, 38, 41, 61], Larrey hernia [42], or congenital anterior diaphragmatic hernia [52]. In Morgagni hernias, intra-abdominal organs are herniated into the thoracic space through a right retrosternal fissure in the diaphragm [26]. In Japanese people, the transverse colon and the great omentum are likely to herniate into the thoracic space [30, 43], because the herniated organs are usually covered with the hernial sac [44]. A hernial sac was present in all of our patients. Comer et al. [18] most often found the hernial sac containing transverse colon, omentum, liver, and, less frequently, small bowel or stomach. In our series, the hernial sac contained transverse colon and omentum in three patients, omentum in four, and omentum with small intestine in one patient. Morgagni hernias are far more common on the right despite protection from the liver. Except for one patient, we determined that all patients had right-sided hernia (Table 2). Morgagni hernia can be associated with the following syndromes and congenital defects: Down's syndrome, Turner's syndrome, Noonan syndrome, Prader Willi syndrome, tetralogy of Fallot, ventricular septal defects, scoliosis, Morquio syndrome, connective tissue disorders, dextrocardia, chest wall deformities, genitourinary abnormalities, and omphalocele [3, 5, 11, 15, 32, 33, 46, 47, 54, 55].

The surgical approach is still controversial regarding the operative technique in Morgagni hernias [53]. Some authors advocate the transthoracic [18, 34] or transabdominal approach [6, 45, 57], others the video-assisted endoscopic technique [2, 3, 7, 9, 28, 49]. Preoperative imaging is crucial for delineation of the hernia's nature and the extent of diaphragm defect. Although small hernias can be closed by direct suturing, mesh repair is usually used in cases of large defects or muscle weakness [29]. Recent reports have described successful treatment of these hernias by laparoscopic repair [25, 36]. In all the cases in our series, laparotomy was preferred on the right-sided hernias. As performed in Bochdalek hernias, diaphragmatic defects were strengthened via primary closure or prolene mesh.

In the present report, we tried to evaluate separately all characteristic features of Bochdalek and Morgagni hernias in a decade. Surgical repair of these two hernias may be performed via laparotomy or thoracotomy according to the individual criteria of the patient. A remarkable point in each hernia case was that all patients had an absence of trauma history. Because a limited number of reviews with respect to Bochdalek and Morgagni hernias have presented so far, our review can help guide surgeons in order to better assess their patients.

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