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Congenital retrosternal hernias of Morgagni: Manifestation and treatment in children

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ABSTRACT

Background: Due to scarcity of congenital diaphragmatic hearnias of Morgagni (CDHM), non-specific clinical presentation in the pediatric age group, we aimed to investigate the incidence, clinical manifestations, anatomical characteristics, and develop diagnostic algorithm and treatment of CDHM in children. **Materials and Methods:** The patients' records of children with CDHM treated in our hospital during past 20 years were retrospectively reviewed for the age at diagnosis, gender, clinical findings, anatomical features, operative details and outcome. **Results:** Since 1995 to 2014 we observed 6 (3 boys, 3 girls) patients with CDHM, that comprise 3.2% of all congenital diaphragmatic hernia cases (*n* = 185). Age at diagnosis varied from 3 mo. to 10y.o. Failure to thrive was main symptom in 4 patients, followed by recurrent respiratory infections (*n* = 3), dyspnea (*n* = 3), and gastrointestinal manifestations: constipation (*n* = 2), abdominal pain (*n* = 1). Work-up consisted of plain X-ray for all (*n* = 6), upper GI (*n* = 3), barium enema (*n* = 2), sonography (*n* = 6) and CT (*n* = 2). Abdominal approach used in 5 patients, and thoracotomy in one. Herniated contents were: liver lobes (*n* = 4), transverse colon (*n* = 3) and greater omentum (*n* = 1). 5 had right-sided lesion, 1- left-sided. Defect repaired using local tissues. Post-operative course was uneventful; all patients appeared well during follow-up. **Conclusion:** CDHM is very uncommon anomaly, very occasionally diagnosed at the early age. Failure to thrive and recurrent respiratory infections are most frequent clinical manifestations. In suspected CDHM we advocate the following work-up: plain chest and abdominal X-ray, contrast study (upper GI series or barium enema), ultrasonographic screen and CT scan. Surgical repair via abdominal approach, using local tissues and hernia sac removal is preferred.

Key words: Children, diaphragmatic hernia of Morgagni, surgical repair

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INTRODUCTION

Despite the advances in surgical and intensive care treatment congenital diaphragmatic hernia (CDH) remains vital anomaly with rather low survival rate among newborn infants and older children all over the world.[1-4] The most common incidence of CDH reported in literature is one in 3500-5000 live births, but the reported numbers are highly dependent upon selectivity of patients by various paediatric surgery centres. The true incidence rate of CDH may constitute 1 in 2000 pregnancies[1-6] taking into account aborted foetuses after prenatal diagnosis, or stillbirth infants due to severe associative malformations and those who died en-route to tertiary medical facilities.[5,7]

The overwhelming majority of CDH appear through posterolateral opening (pleuroperitoneal hiatus) in foetal diaphragm, so-called Bogdalek hernia, which usually manifests early in the neonatal period.[8] Congenital retrosternal diaphragmatic hernias that occur through foramen of Morgagni is a very uncommon congenital anomaly, constituting around 2-3% of all CDH.[9]

MATERIALS AND METHODS

Over the period from 1995 to 2014, there were 185 children who underwent treatment for various anatomical variants of CDH in frames of our Pediatric Surgery Department, six of them had CDH of Morgagni

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(CDHM), three boys and three girls. We retrospectively reviewed patients' records for the age at diagnosis, gender, clinical presentation, diagnostic modalities, hernia site, surgical treatment and outcomes of CDHM.

RESULTS

The relevant demographic, clinical and operative findings are summarised in Table 1. The series of 6 CDHM cases represent the 3.2% of all CDH cases (n = 185), treated in our medical facility. Both genders were equally distributed: 3 girls and 3 boys. The lesion was diagnosed during the first year of life in 4 children, 1 male patient diagnosed at 1 year 2 months, 1 female patient at the age of 10 years old. Our patients were diagnosed after 1 month of age; therefore, our series represent late-presenting cases of CDH. All of our patients were symptomatic. The clinical manifestations ranged from failure to thrive (FTT) (n = 4) to recurrent respiratory infections (n = 3), dyspnoea (n = 3) and nonspecific gastrointestinal (GI) symptoms (constipation — 2, abdominal pain — 1).

In 100% of cases (n = 6), the diagnostic workup commenced with plain anterior-posterior and lateral chest X-ray. The radiographic findings included semi-oval or pear-shaped hernia shadow in the right (n = 5) or left (n = 1) retrosternal space [Figure 1a]. Hernia shadow overlapped cardiac silhouette and mediastinum in the lateral view [Figure 1b].

The contrast study was performed in five patients (upper GI series in 3, barium enema in 2 cases of CDHM). Herniated colon was clearly visualised after barium enema [Figure 2a and b]. Fluoroscopic examination during upper GI series turned out inconclusive in two patients.

Ultrasonographic investigation was part of the work-up in all cases. Neurosonography (n = 2), echocardiography (n = 6), abdominal and retroperitoneal sonography in six patients were performed. These imagine modalities were used to diagnose possible congenital or perinatal central nervous system pathology, associated heart, or kidney malformations and to visualise herniated organs. In particular, the liver was found in hernia sac in four children.

Intravenous contrast-enhanced computed tomography was used for definite diagnosis in two patients with CDHM, Notably, computed tomography unveiled herniated greater omentum and liver through the foramen of Morgagni in a 7-month patient with suspected tumour of anterior mediastinum [Figure 3].

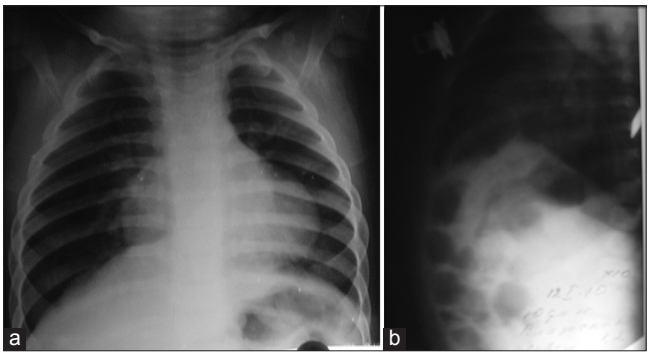

Figure 1: (a) Chest X-ray anterior-posterior (a) and lateral (b) view in 1 year 2 month patient with congenital diaphragmatic hernia of Morgagni. (b) Chest X-ray anterior-posterior (a) and lateral (b) view in 1 year 2 month patient with congenital diaphragmatic hernia of Morgagni

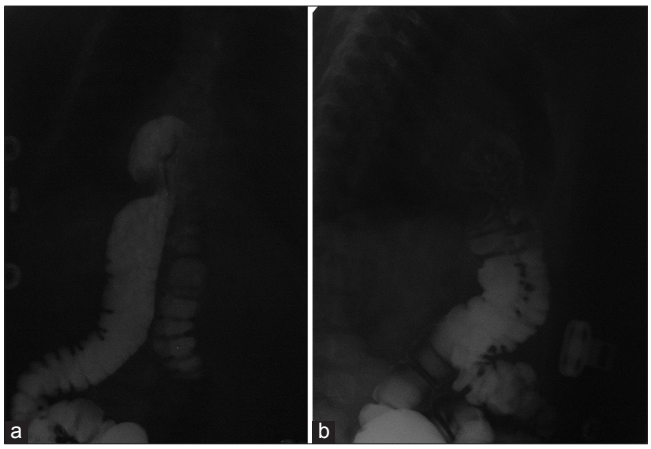

Figure 2: (a) Barium enema in anterior-posterior (a) and lateral (b) view in 4-month patient with congenital diaphragmatic hernia of Morgagni. (b) Barium enema in anterior-posterior (a) and lateral (b) view in 4 months patient with congenital diaphragmatic hernia of Morgagni

Table 1: Demographic, clinical and anatomical data of patients with congenital diaphragmatic hernia of Morgagni

Age/gender	Clinical signs	Work-up	Side/size of defect (cm)	Herniated contents
3 months/male	Pneumonia, dyspnoea, FTT	Plain X-ray, US, CT	Right/6×7	Right, left liver lobes
4 months/male	FTT	Plain X-ray, upper GI, US	Right/4×5	Left liver lobe
7 months/female	FTT, dyspnoea, barrel-chest	Plain X-ray, upper GI, barium enema, US, CT	Right/5×8	Transverse colon, greater omentum, left liver lobe
7 months/female	Chronic cough, constipation, pulmonary artery stenosis, down syndrome	Plain X-ray, barium enema, US	Right/4×4	Left liver lobe
1 year 2 months/male	Acute bronchitises, dyspnoea	Plain X-ray, US	Left/4×4	Splenic flexure of colon
10-year-old/female	Constipation, FTT	Plain X-ray, upper GI, US	Right/6×3	Transverse colon

FTT: Failure to thrive; GI: Gastrointestinal; CT: Computed tomography

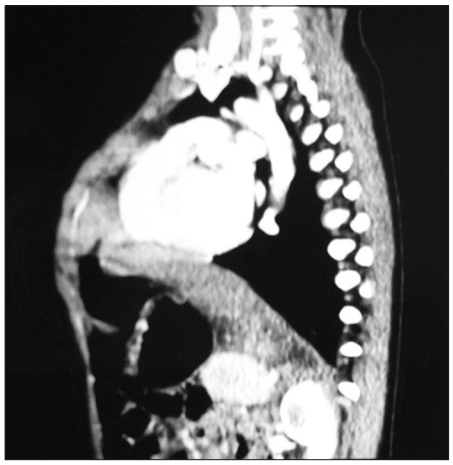

Figure 3: Contrast-enhanced computed tomography in 7 months patient with congenital diaphragmatic hernia of Morgagni

Surgical treatment is imperative in all cases of CDHM. Operative treatment was performed on an elective basis after final diagnosis in every patient. Upper midline laparotomy incision was the preferred surgical approach ($n = 5$). Only once we used lateral thoracotomy through VII intercostal space. Surgical repair consisted of complete ($n = 4$) or partial ($n = 1$) sac excision. We have left the hernia sac in one patient. During laparotomy, the herniated contents were descended back abdominal cavity together with the hernia sac. We used local tissues for the diaphragmatic defect repair with mattress and individual sutures, and anchoring stitches to sternum ($n = 4$), rib ($n = 1$), rib arch ($n = 1$) or intercostal muscles because of anterior diaphragmatic rim absence. There were no cases of patch closure. Only nonabsorbable suture materials (silk 2/0, ethibond 2/0) were utilised. In most cases, the surgical procedure was carried out without drainage ($n = 4$), but after thoracotomy approach the chest drain was placed ($n = 1$) and 1-time sub-diaphragmatic space was drained after laparotomy ($n = 1$). Mean operative time was 2.6 ± 0.2 h.

After review the operating room log book and analyses of the intraoperative details we found that CDHM appeared on the right side more frequently ($n = 5$) and occasionally — on the left ($n = 1$). All patients had hernia sac; it was represented by the leaves of parietal peritoneum ($n = 6$). The size of the defect varied from 16 to 42 cm, mean 25.5 ± 4.6 cm^2. Typically, muscular defect margins yielded normal, except the absence of anterior diaphragmatic rim. The latter is a particular anatomical feature of CDHM. Sternum serves as anterior hernia wall.

Herniated contents were following: Transverse colon ($n = 3$), liver ($n = 4$), (left lobe [$n = 3$], left and right lobes 1) and greater omentum ($n = 1$). We did not have any intra- and post-operative complications. Mean hospital stay was 15 ± 2.0 days (from 10 to 24 days). There were no observed recurrences during a follow-up period that ranges from 2 to 19 years.

DISCUSSION

The first case of diaphragmatic hernia was reported in 1575 by the famous French barber-surgeon Ambroise Paré, who discovered two traumatic diaphragmatic hernias on autopsy.[10] Holt described in 1701 the case of neonatal CDH in his article "Letter from Sir Holt to the Publisher, concerning a Child, Who Had Its Intestines, Mesentery, etc. in the Cavity of Thorax..." in the first ever scientific Journal "philosophical transactions" of the Royal Society.[11] More profound and detailed description of different anatomical variants of CDH in the neonates were discussed by Giovanni Battista Morgagni in his monograph "Seats and causes of disease investigated by anatomy" which laid foundation for pathology as a separate field in medical science. However, Morgagni described retrosternal CDH through the right sternocostal triangle, which currently bears his name, in the adult stonecutter, who died from strangulated herniated colon. Morgagni did not find any signs of traumatic hernia and concluded, that it had been congenital.[12-14] Baron Dominique Jean Larrey, who was surgeon-in-chief of the Napoleon's Grande Armée, became famous for establishing new triage system of flying ambulances amidst battle and staged amputations for wounded warriors. However, neither had he any relations to the treatment of diaphragmatic hernias nor was he a paediatric surgeon.[15] D.J. Larrey in 1829 depicted surgical approach to pericardium through the left sternocostal triangle.[16] Nevertheless, most authors in the literature name retrosternal diaphragmatic hernia Morgagni hernia. It is worth mentioning that in the different periods of time hernia of Morgagni has been termed as substernocostal[17] or retro-costo-xiphoid.[18] In various literature sources authors label right retrosternal hernia as Morgagni's and left Larrey's.[19]

Central part of diaphragm originates from septum transversum,[20] which in its turn derives from the merging of pleuroperitoneal folds with dorsal mesentery. The muscle fibres from the third, fourth and fifth cervical myotomes migrate in-between primitive pleural folds and peritoneum, thus completing diaphragm formation on the 9^{th} week of gestation. Fibres spreading from tendinous centre towards six lower rib arches form costal part of diaphragm. Sternal part arises from the fibres that stretch to the inner surface of the

xiphoid process, partially extending to the posterior aspect of the rectus abdominis sheath. The failure of fusion between the fibrotendinous elements of the sternal part and fibres originating from costochondral arches leaves muscle-free area, so-called sternocostal triangle, known as foramen of Morgagni on the right and Larrey's space on the left.[19]

CDHM always have a hernia sac,[17,21] which makes them entirely different entity from the central diaphragmatic defect present in Pentalogy of Cantrell.[17,22,23] 100% of patients in our series had hernia sac represented either by parietal peritoneum (n = 4) or hypoplastic diaphragm (n = 2). There is no clear evidence of gender association with CDHM. However, Horton *et al*. in his series reports female predominance,[24] while Al-Salem *et al*. discloses 80% of cases in boys.[8] Our data suggest no gender predisposition: We had 3 boys and 3 girls with CDHM. Herniation through the right sternocostal triangle happens in the overwhelming majority of patients (90%), left-sided and bilateral lesions are far less frequent, constituting 8% and 2%, respectively.[2-4,24-26] In our series, we observed 5 right-sided hernias and 1 left-sided CDHM.

Congenital Morgagni hernias in 75% of patients are associated with one congenital anomaly, yet in 66% patients, CDHM is a part of multiple malformations or chromosomal disorders in 37%.[27] Most common co-morbidities encountered are Down syndromes,[28-30] cardiovascular diseases (specifically ventricular septal defects),[31] or intestinal malrotations,[32,33] undescended testes, pyeloureteral stenoses and diaphragm relaxations.[34] According to our data, one of six patients suffered from Down syndrome and pulmonary artery stenosis.

Clinically, CDHM presents with respiratory disorders in 56.5%, GI symptoms in 19% or combination of nonspecific respiratory and gastrointestinal signs in 6.8%.[3,35] Our patients' clinical findings included FTT as most frequent manifestation (n = 4), followed by recurrent respiratory infections (n = 3), dyspnoea (n = 3) and nonspecific GI symptoms (constipation (n = 2), abdominal pain (n = 1). We did not observe any asymptomatic patients. However, up to one-third of cases are reported as clinically asymptomatic, when CDHM is accidental radiographic discovery. Rarely, these patients have intestinal obstruction, strangulation and hollow viscus necrosis as debut of the disease.[36,37] Usually, CDHM is suspected on plain X-ray and confirmed by radiological contrast studies or computed tomography.[38] Despite the congenital nature of illness, CDHM is rarely diagnosed in childhood. Most cases manifest in adulthood.[26,39]

The treatment of congenital retrosternal hernias is always surgical. Operative repair can be performed either through transthoracic or laparotomy approach, via open or minimally-invasive technique.[40] Most surgeons prefer abdominal approach, because of easier reduction of herniated contents back to abdominal cavity, whereas thoracic approach cannot guarantee sufficient revision of both sternocostal triangles and bilateral hernia can be missed. Abdominal approach grants possibility for simultaneous repair of bilateral lesion, undiagnosed before surgery. In 26% of cases, intestinal malrotation co-occurs along with CDHM, which could be corrected via abdominal approach as well.[8] We used laparotomy incision for correction of CDHM in 5 patients and thoracotomy approach in one, because of suspected tumour in the chest cavity. Minimally-invasive technique is advocated due to shorter operative time, earlier post-operative recovery, shorter hospital stay and better cosmetic results.[41] Unfortunately, we did not possess sufficient experience of laparoscopic procedures to manage patients with CDHM.

In the vast majority of English publications, hernias of Morgagni are presented as single or anecdotal reports of accidental findings in the adult patients. Thus, Ahmad *et al*. describe giant retrosternal hernia in a 42-year-old male, diagnosed during an acute heart attack.[42] Rather interesting clinical case of acute appendicitis strangulated in the foramen of Morgagni in 76-year-old patient reported by Bettini *et al*.[43] Occasionally, asymptomatic CDHM presents with acute surgical condition as strangulated small bowel diverticulum,[44] organo-axial gastric volvulus inside hernia sac in 78-year-old female[45] or acute bowel loop strangulation in a 27-year-old pregnant woman on the 21st gestation week.[46]

Regardless of scarcity, there are recent retrospective studies of CDHM series, although mostly representing adult data. Aghajanzadeh *et al*. conducted a review of 36 patients with CDHM aged from 8 to 83 years old, 50% of whom had asymptomatic course, chronic cough occurred in 17% and 14% of patients suffered from constipation. Diagnostic algorithm included plain chest X-ray, upper GI study and computed tomography.[38] Considerable selection of 20 CDHM cases aged 17-50 years old. gathered and analysed by Abraham *et al*. 40% reported to be asymptomatic; abdominal pain and postprandial discomfort were dominating signs, along with stomach or colon, being only organs herniated through sternocostal triangle.[47] Foremost and thorough,

especially interesting for paediatric surgeons, selection of children with CDHM, retrospectively reviewed and analysed in the Kingdom of Saudi Arabia by Al-Salem *et al*. Series consisted of 53 children (38 males and 15 females), age ranging from 1 month to 9 years old from all over the country in the past 20 years. Recurrent respiratory infections observed in 81% of patients, 44.5% had right-sided hernia, 28% left-sided while bilateral lesion existed in 30% of children. Associated anomalies noted in the majority of cases, particularly cardiovascular diseases, intestinal malrotation and Down syndrome. Herniated contents included colon and liver. The small bowel, greater omentum and stomach were less commonly encountered in the hernia sac.[40] Comparing to the recently reported series, ours is smaller, but the outcomes of our retrospective study are consistent and corresponding with the data reported in wider selections.

CONCLUSIONS

CDHM is very uncommon anomaly that according to our data accounts for 3.2% of all patients with CDH. Congenital retrosternal hernias are very occasionally diagnosed at the early age. FTT and recurrent respiratory infections are most frequent clinical manifestation of Morgagni hernias in children.

Our elaborated diagnostic algorithm for suspected CDHM includes:

1. Plain chest and abdominal X-ray in anterior posteral and lateral view.
2. Radiographic contrast study (upper GI series or barium enema.
3. Sonographic evaluation: Neurosonography in infants under 12 months of age, echocardiography, abdominal and retroperitoneal ultrasonography.
4. Contrast-enhanced computed tomography.

Surgical correction is imperative in all cases. Abdominal approach is preferred. Operative treatment consists of hernia sac excision and diaphragm defect repair using local tissues.

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Conflicts of interest
There are no conflicts of interest.

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