Congenital Morgagni's hernia in infants and children: a national review

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Background **Congenital Morgagni's hernia (CMH) is rare and has unique features in terms of clinical presentation, high incidence of bilaterality, and associated anomalies. This is a review of all CMH cases reported from Saudi Arabia, highlighting clinical features, associated anomalies, aspects of diagnosis, and management.**

Patients and methods **A Medline search was made for all manuscripts published from Saudi Arabia on CMH. The total number of patients, their age at diagnosis, sex, clinical features, associated anomalies, site of hernia, and methods of treatment were recorded.**

Results **From 1991 to 2014, 19 manuscripts were published from Saudi Arabia. The total number of patients reported was 106 (72 male and 31 female). In three patients, the sex was not reported. Their mean age was 23.8 months (1 month–9 years). Recurrent chest infection was the most common presentation [56 (54.4%)]. In seven (6.8%) patients, the hernia was discovered incidentally and 19 (18.4%) presented acutely. Associated anomalies were seen in 66 (64%) of 103 patients. Down's syndrome (30%) and congenital heart disease (23%) were the most common. Sixty-four (60.4%) underwent repair through an abdominal approach, 36 (34%) underwent laparoscopic-assisted repair, and in five (4.7%) patients the hernia was repaired laparoscopically. In 94 patients, the site of the hernia was specified [45 (47.9%) right, 22 (23.4%) left, and 27 (28.7%) bilateral]. Four (3.8%) developed recurrences and three had an incisional hernia. One developed midgut volvulus 3 years postoperatively.**

Conclusion **CMH is rare and the presentation is nonspecific and variable but the majority present with repeated attacks of chest infection. Physicians caring for these patients should be aware of this, and infants and children with repeated attacks of chest infection should be investigated. It is also important to evaluate these patients for possible associated anomalies, including an echocardiogram. Once the diagnosis of CMH is confirmed, they should be repaired surgically regardless of whether symptomatic or asymptomatic. The laparoscopic-assisted approach is a simple, safe, and sound technique, and leaving the hernia sac intact has no adverse effects.** ***Ann Pediatr Surg*** **13:182–187 © 2017 Annals of Pediatric Surgery.**

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Introduction

Congenital hernia of Morgagni was first described by Giovanni Battista Morgagni, an Italian anatomist and pathologist in 1761, while performing a postmortem examination on an Italian stonecutter who died of a head injury [1–3]. CMH, which is also called congenital Morgagni–Larrey's hernia, is a congenital herniation of abdominal contents into the thoracic cavity through a retrosternal diaphragmatic defect, the foramen of Morgagni. In the pediatric age group, CMH is rare, accounting for about 1–5% of all types of congenital diaphragmatic hernia [2,3]. It can be asymptomatic and discovered incidentally or cause vague gastrointestinal symptoms, but more commonly it causes recurrent attacks of chest infection and rarely does it cause severe respiratory symptoms resembling the more common congenital posterolateral diaphragmatic hernia, especially during infancy [1–6]. CMH has unique features in terms of clinical presentation, high incidence of bilaterality, and associated anomalies [3,5,7,8]. This as well as its occurrence in identical twins raised the possibility that it may result from an inherited defect [9,10]. In Saudi Arabia, CMH made up about 10.5% of all types of congenital diaphragmatic hernia [11]. This is relatively high when compared with that reported internationally. One contributing factor for this is the high rate of consanguinity in this part of the world.

Patients and methods

A Medline search was made for all manuscripts published from Saudi Arabia on Morgagni's hernia. This also included locally published manuscripts. A total of 19 manuscripts were published concerning Morgagni's hernia from Saudi Arabia [7,8,11–27]. Some of these manuscripts were published as case reports or small series initially and were included in larger series subsequently. This was taken in consideration during the analysis. The total number of patients reported, their age at diagnosis, sex, clinical features, associated anomalies, site of the hernia, and methods of treatment were recorded.

Results

Nineteen manuscripts were published from Saudi Arabia [7,8,11–27]. The first publication of CMH from Saudi Arabia was published in 1991 [26]. This manuscript reported three children with CMH. The second manuscript was published in March 1998 and reported four

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children among 38 infants and children treated for different types of congenital diaphragmatic hernia [11]. According to this manuscript, CMH accounted for 10.5% of all types of congenital diaphragmatic hernia. One month later, Al-Arfaj [17] reported five infants and children with CMH. From 1991 to 2014, the total number of patients with CMH reported was 106 infants and children. There were 72 male and 31 female patients (male : female was 2.3 : 1). In three patients, the sex was not reported. Their mean age at presentation was 23.8 months (1 month–9 years). In three patients, the age at presentation was not reported. Their clinical presentations are shown in Table 1. These excluded the three patients in whom clinical features were not available. Recurrent chest infection was the most common presentation in 56 (54.4%) patients. Eight (7.8%) patients presented with vomiting and feeding difficulties. In seven (6.8%) patients, the hernia was discovered incidentally during evaluation of other unrelated conditions. Nineteen (18.4%) patients presented acutely with respiratory distress resembling the more common congenital posterolateral diaphragmatic hernia.

Associated anomalies were seen in 66 (64%) of 103 patients as shown in Table 2. Down's syndrome was the most common associated anomaly seen in 31 (30%) patients. This was followed by congenital heart disease in 24 (23%) and inguinal hernia in 13 (12.6%) patients. Thirteen (12.6%) patients had associated malrotation. In four patients, CMH was precipitated by an increase in intra-abdominal pressure. In one patient, this was the result of road traffic accident with blunt abdominal trauma, which resulted in herniation of intestines into an already existing bilateral CMH [19]. This was confirmed intraoperatively where bilateral CMH with hernia sacs were found containing intestines. Two patients had hydrocephalus with a ventriculoperitoneal shunt, which contributed to the appearance of the hernia by increasing the intra-abdominal pressure. The fourth patient was a case of β-thalassemia major and underwent splenectomy because of frequent blood transfusion. Following this and as a result of progressive liver enlargement, the left lobe of the liver herniated into an already existing left CMH, which enlarged gradually [24].

The diagnosis of CMH was suspected on chest radiography including a lateral film to show anterior herniation of bowel loops (Fig. 1a and b). This was confirmed by barium meal and follow through in 16 patients and barium enema in 20 patients (Fig. 2a and b). In seven patients, chest and abdominal computed tomography (CT)-scan confirmed the diagnosis of CMH. This is especially in those with a soft tissue density seen in the cardiohrenic angle on chest radiography suggesting herniation of a part of the left lobe of the liver or the omentum (Fig. 3). CT-scan was also valuable in the diagnosis of those with bilateral Morgagni's hernia (Fig. 4). In one patient, the diagnosis was made intraoperatively. He had a right-sided soft tissue density mass in the right cardiophrenic angle, and through a right thoracotomy he was found to have a right CMH with herniation of a part of the left lobe of the liver [18].

All patients were operated on. In the early years of the study, 64 (60.4%) underwent repair using an abdominal approach and in all except nine the hernia sac was excised [16]. In one patient, the repair was performed using a transthoracic approach, because in this patient there was herniation of a part of the left lobe of the liver, which was mistaken to be a thoracic mass [18]. Thirty-six (34%) patients underwent laparoscopic-assisted repair of the hernia and in five (4.7%) the hernia was repaired laparoscopically. In all those operated using a laparoscopic or laparoscopic-assisted approach, the hernia sac was not excised. In 94 patients, the site of the hernia was specified. In 45 (47.9%) patients, the hernia was on the right side, 22 (23.4%) had left-sided hernia, and 27 (28.7%) had bilateral CMH. On follow-up, four (3.8%) developed recurrence, three of them underwent repair using the open approach and one following a laparoscopic-assisted approach. All four had Down's syndrome and three of them also had an incisional hernia. One of the patients developed midgut volvulus 3 years postoperatively [19]. This patient had malrotation that was discovered intraoperatively but was not corrected.

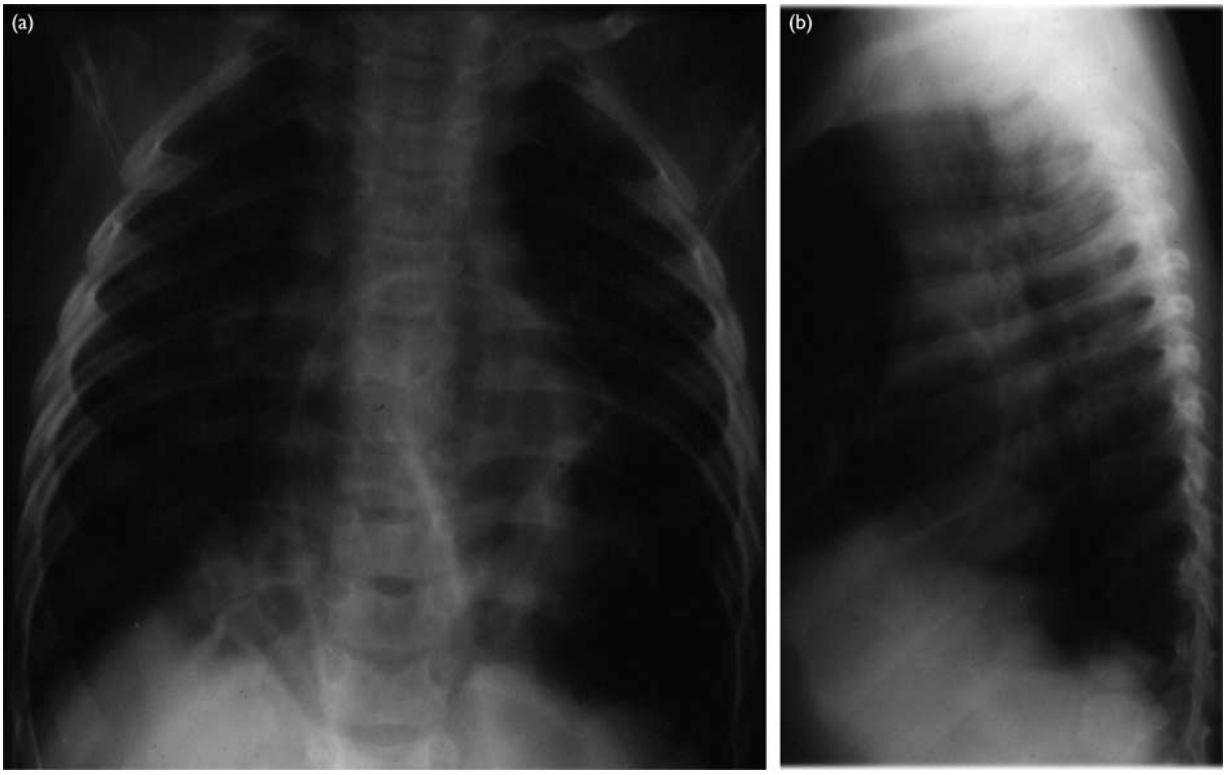
Table 1 Clinical features of 103 patients with congenital Morgagni's hernia

Clinical features	*n* (%)
Recurrent chest infection	56 (54.4)
Nonspecific upper respiratory tract symptoms	5 (4.9)
Gastroesophageal reflux disease	2 (1.9)
Discovered incidentally	7 (6.8)
Following road traffic accident with blunt abdominal trauma	1 (0.97)
Following splenectomy	1 (0.97)
Feeding difficulties and vomiting	8 (7.8)
Respiratory distress	19 (18.4)
Regurgitation from the mouth and nose	1 (0.97)
Intestinal obstruction	1 (0.97)
Failure to thrive	1 (0.97)
Asthma-like symptoms	1 (0.97)

Table 2 Associated anomalies in 103 patients with congenital Morgagni's hernia

Associated anomalies	*n* (%)
Congenital heart disease	24 (23.3)
Down's syndrome	31 (30)
Inguinal hernia	13 (12.6)
Malrotation	13 (12.6)
Umbilical hernia	5 (4.9)
Hypospadias	4 (3.5)
Hydrocephalus	2 (1.9)
Undescended testes	2 (1.9)
Hemangioma	2 (1.9)
Scoliosis	1 (0.97)
Anorectal malformation	2 (1.9)
Desmorphic, congenital dysplasia of the hip, arthrogryphosis multiplex congenital, Werdnig–Hoffman disease	1 (0.97)
Short stature, plagiocephaly, atrophied left frontal lobe, hyperteleorism, low-set ears	1 (0.97)
Congenital dysplastic hip, congenital talipes equinovarus	1 (0.97)
Mongolian spots	1 (0.97)
Duodenal web	1 (0.97)
Pectus carnitum	1 (0.97)
Patent urachus	1 (0.97)
Pyloric atresia	1 (0.97)
Sickle cell anemia	1 (0.97)
Hypertrophic pyloric stenosis	1 (0.97)

Fig. 1



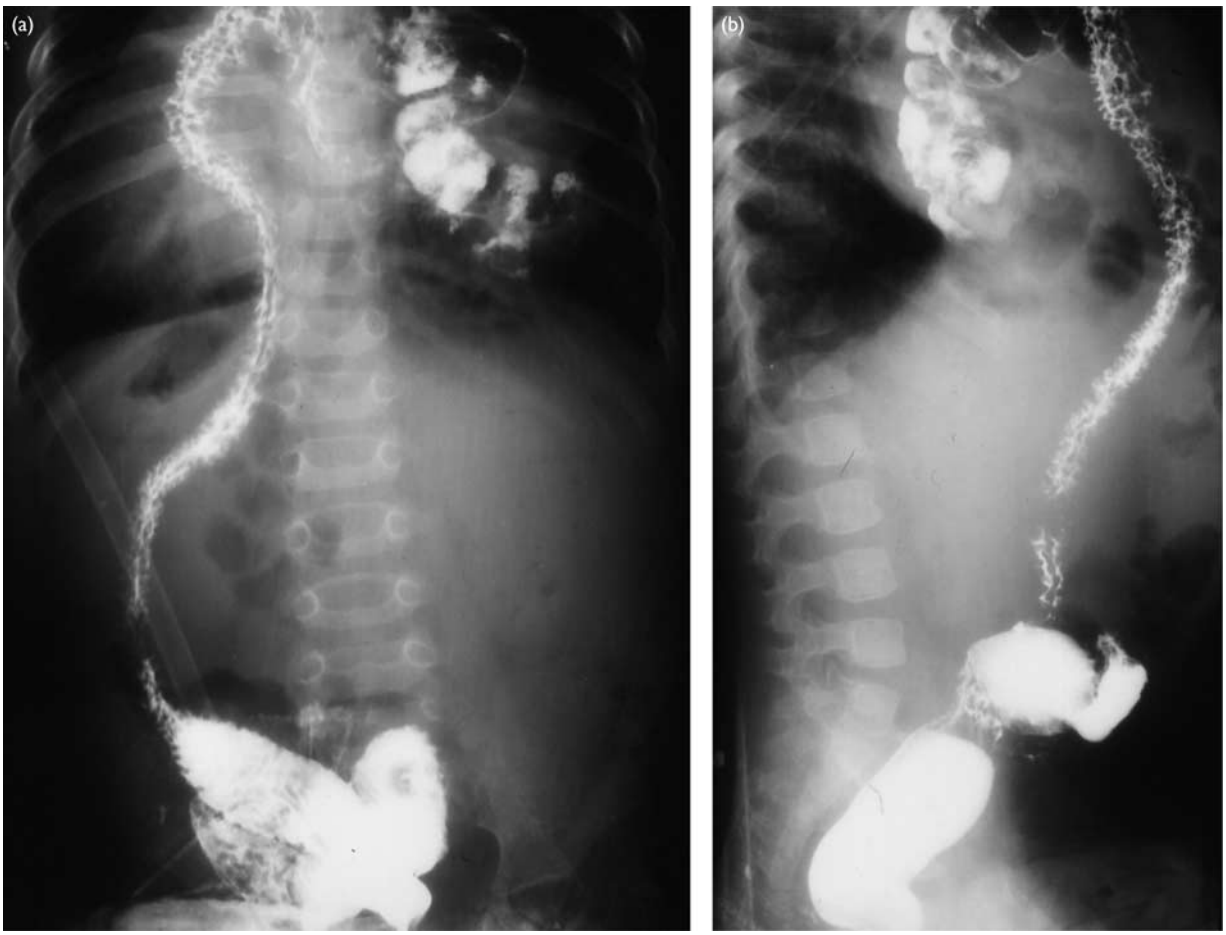
(a, b) Anteroposterior and lateral chest radiographs showing bowel loops herniating through the foramen of Morgagni.

Discussion

CMH is relatively rare when compared with other types of congenital diaphragmatic hernia. Over a period of 40 years, Berman *et al*. [1] treated only 18 cases of CMH, and over a period of 25 years Pokorney *et al.* [3] treated only four cases of CMH. Cigdem *et al.* [5] over a period of 23 years treated only 16 cases of CMH. Al-Salem *et al.* [7] over a period of 18 years treated 23 cases of CMH. The majority of congenital diaphragmatic hernias occur through the foramen of Bochdalek, and the exact incidence of CMH is not known and difficult to estimate as many of these cases may remain asymptomatic or are discovered during adulthood [28]. The incidence of CMH in Saudi Arabia is not known but it was estimated to be relatively higher than that reported from other parts of the world. In a previous study from Saudi Arabia, CMH made up about 10.5% of all types of congenital diaphragmatic hernia [11]. Over a period of 23 years, we found 19 publications from Saudi Arabia reporting a total number of 106 infants and children with CMH. We feel this is an underestimation of the actual number as many patients with CMH are not reported or present late as adults. A national congenital malformation registry is important in this regard. One contributing factor for the increased number of CMH in this part of the world is the high rate of consanguinity. This as well as the high incidence of associated anomalies and its occurrence in identical twins raised the possibility that CMH may result from an inherited defect [9,10].

An interesting feature of CMH is the high incidence of associated anomalies [3,5,7,8]. In a review of the literature, Pokorny *et al.* [3] found 22 patients with CMH, 13 (81%) of 16 patients in whom detailed descriptions were available had associated anomalies. Al-Salem found associated anomalies in 18 (78.3%) of 23 patients with CMH [7]. Cigdem *et al.* [5] reported 16 patients with CMH; 12 (75%) of them had at least one associated congenital malformation and eight (66.6%) of them had multiple anomalies. In this review of 106 infants and children with CMH, 66 (64%) of 103 patients had associated anomalies (Table 2). Congenital heart disease and Down's syndrome are the most common associated anomalies. This was the case in our series as 24 (23.3%) of our patients had congenital heart disease and 31 (30%) had Down's syndrome. The severity of congenital heart disease was variable but atrial septal defect and ventricular septal defect were the most common associated heart defects [6–8]. It is also well known that CMH can be associated with Down's syndrome, but the reported incidence is variable. Pokorney *et al.* [3] in a review of 22 infants with CMH found that three (14%) of them had Down's syndrome. In a collective series of 46 children with CMH, 16 (34.8%) of them had Down's syndrome [29]. Cigden *et al.* [5] reported a 31.25% incidence of Down's syndrome in a series of 16 patients with CMH seen over a period of 23 years. In 2001, Parmar *et al.* [30] reviewed the literature and revealed only 18 cases of Morgagni hernia associated with Down's syndrome. Jetley *et al.* [15] reported 22 patients with CMH; 11 (50%) of them had Down's syndrome and this association was responsible for delayed diagnosis and recurrence of CMH. In a series of 53 patients, 15 (28.3%) had Down's syndrome [8]. A causal relationship may exist between Down's syndrome and other congenital malformations including CMH [9]. The hypotonic muscle

Fig. 2



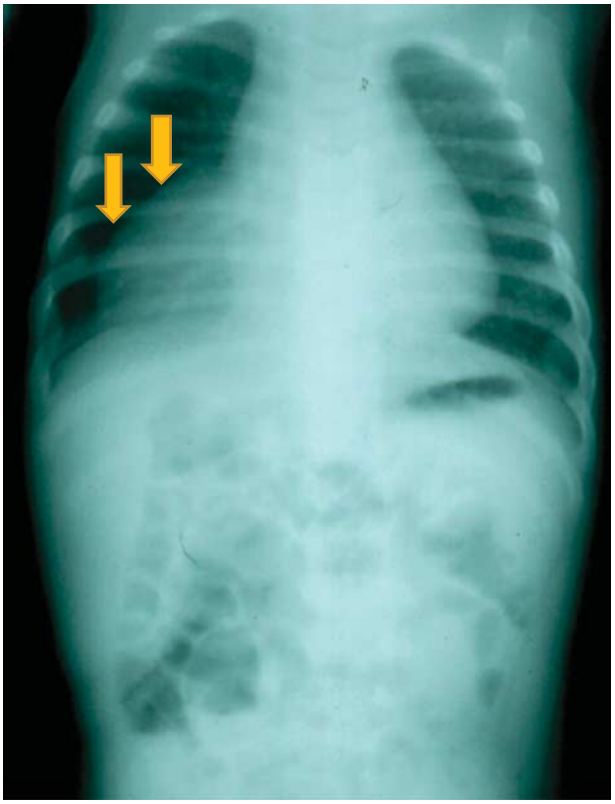
(a, b) Barium enema showing herniation of the colon through the foramen of Morgagni.

development, which is a well-established developmental defect in those with Down's syndrome, may play a role in the development of CMH and also may play a role in its recurrence. Honore *et al.* [9] suggested a defective dorsoventral migration of rhabdomyoblasts from the paraxial myotomes, caused by increased cellular adhesiveness in those with Down's syndrome. This association definitely affected the final outcome, and the poor hypotonic muscle development in them has resulted in recurrence in four of our patients; all of them had Down's syndrome and three of them also had incisional hernias. Thirteen (12.6%) of our patients had associated inguinal hernias and 13 (12.6) had malrotation. The association of CMH and malrotation is well established and this is of paramount importance when considering the operative approach to repair CMH as well as intraoperatively to obviate the risk for postoperative volvulus [7]. One of our patients had malrotation that was discovered intraoperatively but was not corrected, and this patient developed midgut volvulus 3 years postoperatively [17]. An interesting finding was the remarkably higher incidence of associated anomalies in those with bilateral CMH when compared with those with unilateral CMH [13].

The presentation of CMH is variable and nonspecific and this is one of the causes of delayed diagnosis [12]. CMH can be discovered incidentally or cause vague gastrointestinal symptoms, but more commonly it causes respiratory symptoms with repeated chest infections [4,8]. In our study, the majority of our patients presented with repeated attacks of chest infections necessitating several courses of antibiotics and hospitalizations in some of them. To obviate this, physicians caring for these patients should be aware of this and a high index of suspicion is important to obviate delay in diagnosis with its associated morbidity, and infants and children presenting with recurrent chest infection should be investigated [25,29,31–35].

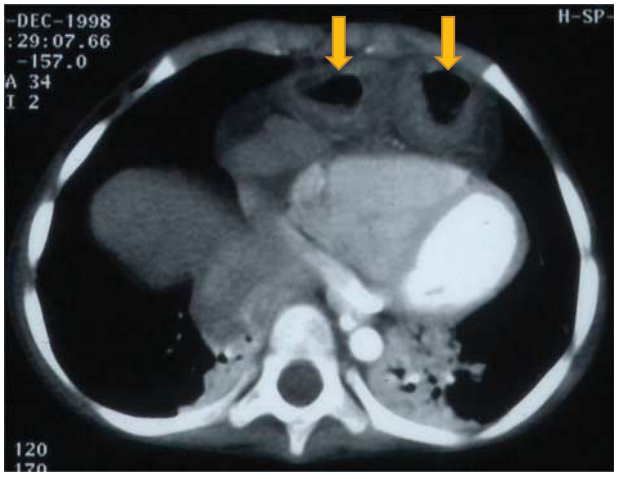
In the neonatal period, CMH can present acutely with respiratory distress indistinguishable from that of Bochdalek hernia [1,36]. Fifty-six (54.4%) of our patients presented with recurrent chest infection but 19 (18.4%) of them presented acutely with respiratory distress. Some of these patients were found to have bilateral CMH [8,13]. Berman *et al.* [1] concluded that late-presenting CMH is relatively benign; nevertheless, it causes significant morbidity [31–34]. This is specially so if the child is not adequately investigated, where the diagnosis can be missed or delayed. At times, CMH remains asymptomatic or discovered accidently during evaluation of other nonrelated conditions [1,12,17,29,35]. In seven (6.8%) of our patients, CMH was asymptomatic and was discovered during

Fig. 3



Chest radiography showing a soft tissue density in the left cadiophrenic angle, which should be included in the differential diagnosis of Morgagni hernia (arrows).

Fig. 4



Computed tomographic scan showing bilateral Morgagni's hernia (arrows).

evaluation of other unrelated conditions. Rapid weight gains as well as history of trauma and ventriculoperitoneal shunts have been reported as contributing factors for the delayed appearance of CMH as a result of an increase in intra-abdominal pressure [8,12,13,19].

The diagnosis of CMH is based on chest radiography, which shows herniation of bowel loops into the chest and a lateral film will show their anterior herniation. At times, the hernia sac may be empty, and so it is not uncommon for some of these patients to have a previously normal chest radiography, which should not preclude a diagnosis of CMH [37,38]. In suspected cases, the diagnosis can be confirmed with a barium enema or barium meal and follow-through [12,13]. Barium enema is a more useful investigation as the colon is the most common organ to herniate in CMH [12]. In those with herniation of a part of the liver or omentum, they present as a mass in the anterior right or left cardiophrenic angle on chest radiography. CMH should be included in the differential diagnosis of children who present with a soft tissue density or mass on chest radiography located in the right or left cardiophrenic angle [18,24]. In such situations and where there is confusion as regards the diagnosis, ultrasound and/or CT-scan is useful in establishing the diagnosis. CT-scan is also useful in demonstrating bilateral CMH even when the hernia sacs are empty [13]. CT-scan is more useful in those with herniation of a part of the liver or omentum where CT-scan can demonstrate continuity between the herniating omentum or a part of the liver and the abdominal omentum and liver.

There is a general consensus that CMH whether symptomatic or asymptomatic should be treated surgically. This is to obviate the potential complication of irreducibility and strangulation despite its low frequency [1,31–34]. The transabdominal approach using either an upper midline or an upper transverse incision was the preferred approach in the past. This will make reduction and inspection of contents easy, allows access and repair of bilateral hernias simultaneously, and corrects an associated malrotation when present [12]. In the early years of this study, 64 (60.4%) underwent repair using an abdominal approach. The recent advances in minimal invasive surgery have made laparoscopic repair of CMH safe and effective both in children and adults. Georgacopulo *et al.* [38] reported the first successful laparoscopic repair of Morgagni's hernia in a child. A variety of techniques to repair CMH laparoscopically have been described [20,39–43]. The laparoscopic interrupted or continuous suture technique is not simple and time consuming. In addition, the anterior edge of the defect is not well developed and absent in a large number of these patients. This makes direct closure of the defect weak and liable to breakdown and recurrence. A better and simpler approach is to take the sutures through the posterior edge of the hernia defect and incorporate the entire anterior abdominal wall in the sutures and tie the knots extracorporealy in the subcutaneous tissues [14,20,44,45]. Thirty-six (34%) of our patients underwent laparoscopic-assisted repair of the hernia, and in five (4.7%) patients the hernia was repaired laparoscopically. This technique also obviates the need to use a prosthetic patch [41]. It is a simple technique and when compared with the open repair, it is definitely better [8,14]. It takes a shorter operative time and produces a sound repair. In addition, it has shorter hospital stay, earlier commencement of feeds, less requirement for postoperative analgesia, and better cosmetic appearance [8,14]. Robot-assisted laparoscopic Morgagni's hernia repair was also reported to be safe and feasible in children, but the setup time prolongs the procedure and the overall operative time is longer than the laparoscopic-assisted repair [46,47].

Another still controversial point at the time of repair is whether to excise or leave the hernia sac [20,23,43,44,48,49]. In all our cases treated using the open approach, the hernia sac was excised except in 9. This, however, was not the case for those treated using the laparoscopic-assisted approach. We, like others, advocate leaving the hernia sac as there was no difference between the two groups in this regard [20,23,48].

Conclusion

CMH is rare in infants and children. The presentation is nonspecific and variable but the majority present with repeated attacks of chest infection. Physicians caring for these patients should be aware of this and a high index of suspicion is important to avoid associated morbidity. An anteroposterior and lateral chest radiography are important to diagnose CMH, and, rarely, a barium enema is necessary to confirm the diagnosis. CT-scan is important to diagnose those with herniation of solid organs such as the omentum and a part of the liver. It is also important to evaluate these patients for possible associated anomalies and an echocardiogram should form a part of their investigations. Once the diagnosis is confirmed, these should be repaired surgically regardless of whether symptomatic or asymptomatic. The laparoscopic-assisted approach is a simple, safe, and sound technique to repair CMH, and leaving the hernia sac has no adverse effects.

Conflicts of interest

There are no conflicts of interest.

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