#### CASE REPORT

1757

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# Maternal Strangulated Diaphragmatic Hernia with Gangrene of the Entire Stomach During Pregnancy: A Case Report and Review of the Recent Literature

Ah Yeong Chae<sup>1</sup>, So Yeon Park<sup>1</sup>, Jung Hyun Bae<sup>1</sup>, So Yeon Jeong<sup>1</sup>, Ji Su Kim<sup>1</sup>, Jeong Soo Lee<sup>2</sup>, Soo Jin Kim<sup>2</sup>, Soo Jeong Lee <sup>3</sup>, Sang Hun Lee <sup>3</sup>

Department of Obstetrics and Gynecology, University of Ulsan College of Medicine, Asan Medical Center, Seoul, Korea; <sup>2</sup>Department of Obstetrics and Gynecology, Eulji University, Nowon Eulji Medical Center, Seoul, Korea; <sup>3</sup>Department of Obstetrics and Gynecology, University of Ulsan College of Medicine, Ulsan University Hospital, Ulsan, Korea

Correspondence: Sang Hun Lee, Department of Obstetrics and Gynecology, Ulsan University Hospital, University of Ulsan College of Medicine, 290-3 Joenha-Dong, Dong-Gu, Ulsan, 682-714, Korea, Tel +82-052-250-8786, Fax +82-052-250-7163, Email shlee73@uuh.ulsan.kr

Background: Bochdalek hernia (BH) of congenital diaphragm hernia is infrequently seen in adults. Strangulation of the diaphragm hernia has been recognized as a severe complication. Among several factors, pregnancy is an important cause of diaphragm hernia's deterioration. However, nausea, vomiting, and upper abdominal pain are often considered non-specific pregnancy-related symptoms. Case Presentation: We report a case of a 39-year-old (gravida II, para I) multigravida woman with a delayed diagnosis of strangulated herniated viscera complicating total gastric gangrene at 26+1 weeks' gestation. The preoperative diagnosis was confirmed by an X-ray examination and magnetic resonance imaging (MRI). After identifying the size and severity of the herniated contents through video-assisted thoracoscopy (VAT), we immediately converted to abdominal laparotomy. Antenatal corticosteroids were administered simultaneously with diagnosis to promote fetal maturity. The fetal condition was maintained well in the maternal uterus during the operation. Careful monitoring of the fetus and the mother's clinical conditions should be performed during expectant management to achieve delayed delivery after maternal surgical correction. Delivery was completed through cesarean delivery at 27+1 weeks of gestation.

Conclusion: Despite the rarity of maternal Bochdalek hernias during pregnancy, early diagnosis and appropriate treatment via multidisciplinary care are essential for maternal and fetal outcomes.

Keywords: strangulation, Bochdalek hernia, pregnancy

### Introduction

Diaphragmatic hernia, which was first described in 1610 through Ambroise Paré's necropsy findings, is categorized into three types: congenital, acquired, and traumatic. Congenital types include posterolateral (Bochdalek) or substernal (Morgagni) portions, while acquired types include the esophageal hiatus (hiatal) or the aortic or caval openings. Bochdalek hernia (BH) of congenital diaphragm hernia (CDH) is considered a life-threatening disease in neonatal infants with a prevalence of 0.8 to 5/ 1000.<sup>1</sup> However, Mullins et al reported in a previous paper that the incidence of asymptomatic adult Bochdalek hernia was 0.17% based on 13,138.<sup>2</sup> Despite the rarity of maternal Bochdalek hernias during pregnancy, pregnancy is associated with one factor that suddenly exacerbates symptoms of asymptomatic adult Bochdalek hernia.<sup>3</sup> Therefore, if diagnosis and treatment are delayed, the results may be poor outcomes due to obstruction, perforation, ischemia, gangrene, or necrosis of internal organs.<sup>4</sup> This is the rare case of delayed delivery of a pregnant patient who survived after antenatal surgery for a congenital diaphragmatic hernia with gangrene of the entire stomach.

### **Case Report**

A 39-year-old woman (gravida 2, para 1) at 26+1 weeks' gestation visited our emergency room with severe epigastric pain accompanied by projectile vomiting. The patient had been suffering from epigastric pain for the last two months. A few days ago, before she came to the emergency room, she could not even drink water due to vomiting and nausea. On arrival at the emergency department, she had blood pressure of 95/72 mmHg, pulse rate of 160/ min, body temperature of 37 0.2° C, and respiratory rate of 20/ min. An abdominal examination showed left-side tenderness and a gravid uterus. Laboratory parameters had a white cell count of  $21,960/\mu$ L, hemoglobin of 11.1 g/dL, platelet count of  $253,000/\mu$ L, D-dimer of 4.61 µg/mL, and C-reactive protein of 14.70 mg/dL. Chest X-ray revealed the left hemidiaphragmatic eventration accompanied by multiple passive collapses in the left lung. Magnetic resonance imaging (MRI) of the abdomen was requested for a definitive diagnosis. A Levin tube was inserted as the patient waited for the MRI examination. Through the MRI reporting, the stomach and distal transverse colon had herniated in the left intrathorax (Figure 1). The gastroesophageal junction and the duodenal bulb were squeezing and stretching, and the spleen's anterior displacement was shown (Figure 1). As a result of evaluating the fetal condition in the uterus through an emergency obstetric ultrasound such as echocardiography and Doppler, the fetal condition was good. The estimated fetal weight was 988g, the presentation was breech, the fetal heartbeat rate was 150/min, and the fetal movement was normal. The patient underwent surgery under emergency conditions in the cardiothoracic and general surgery departments. Laparoscopic surgery was attempted first, but the surgical team switched to open surgery because of a large amount of bloody fluid and severe stomach and spleen infarction. The left posterior diaphragm had a defect of more than 10cm (Figure 2). The entire stomach and the splenic flexure of the transverse colon were found inside the left hemithorax. When the contents of the diaphragmatic hernia were moved manually into the abdomen, the stomach showed a totally ischaemic change with gangrene (Figure 2). Therefore, the total gastrectomy and Roux-en-Y esophagojejunostomy were performed. Additionally, severe splenomegaly and necrotic changes in the spleen were found. A splenectomy was performed. Later on, primary closure of the diaphragmatic defect and chest tube insertion at the anterior axillary line of the 9th intercostal space was performed. Meanwhile, fetal monitoring by portable Doppler was performed during surgery. After surgery, she was treated in the intensive care unit for three days and then transferred to a general ward. The patient received a tocolytic agent by continuous intravenous infusion of ritodrine to inhibit preterm labor. However, on the 7th day after surgery, the patient had to have a cesarean delivery due to labor pain. The baby was born at 1110g at 27+1 gestational weeks. (The Apgar score was 6 at 1 min and 7 at 5 min after birth.) Two months later, the patient was discharged with a pigtail inserted in the left subphrenic space. Abdominal computerized tomography (CT) was performed and revealed a 6-cm width of complicated fluid collection on the day after the cesarean delivery (Figure 3). One month after discharge from the hospital, the pigtail was removed. One year later, an outpatient abdominal CT showed no diaphragmatic hernia recurrence (Figure 3).



Figure I (A and B) Coronal T2-weighted fast spin-echo MR images showing herniation of the stomach (arrowhead) and splenic flexure of the distal transverse colon(arrows) in the left intrathorax. (C) Transaxial magnetic resonance (MR) imaging showing severe anterior diaphragmatic hernia (arrow) of the spleen.



Figure 2 (A) Perioperative photograph showing a large defect in the left posterior dome of the diaphragm. (B) Postoperative photograph showing necrosis of the entire stomach.

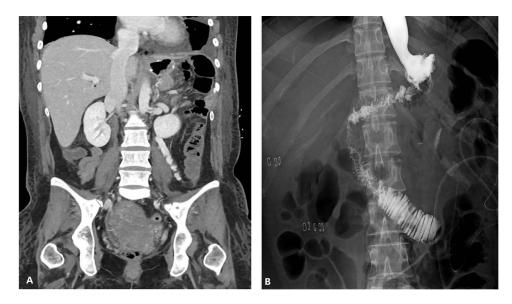


Figure 3 (A) Early postoperative coronal computed tomography (CT) scan after total gastrectomy and splenectomy due to incarcerated left diaphragmatic hernia. (B) After postoperative and delayed delivery, upper gastrointestinal (UGI) series with gastrografin swallow show no evidence of contrast leakage or passage disturbance at the anastomosis site.

### Discussion

We reported a case of a multigravida woman who had strangulated herniated viscera, complicating total gastric gangrene in 2nd trimester. Brown et al announced that pregnancy causes symptomatic Bochdalek hernia in an adult.<sup>3</sup> Choi et al<sup>5</sup> systemically reviewed case reports and case series reporting maternal Bochdalek hernia in pregnant women, in which mean age at diagnosis accompanying symptoms are 28.5 years, the parity was primigravida (43%), multiparous (36%), and unknown (21%), and the gestational age was antenatal period (65%) and postpartum including intrapartum (35%). These hernia defects can easily occur during delivery but can occur without delivery in the second or third trimester of pregnancy.

Pregnancy suddenly exacerbates symptoms of asymptomatic adult Bochdalek hernia regardless of gestational age.<sup>3</sup> Pregnant women with symptoms should undergo emergency surgery regardless of their gestational age. However, it is debatable whether to choose conservative or surgical treatment for accidentally recognized asymptomatic Bochdalek hernia of pregnant women. On maternal Bochdalek hernia during pregnancy, Choi et al<sup>5</sup> found that obstruction, perforation, ischemia, gangrene, or necrosis of the herniated organ accounted for 44%. Obstruction, perforation, ischemia, gangrene, or necrosis of the herniated organ increases maternal (11% of collected cases)<sup>6,7</sup> and fetal-neonatal mortality (21% of collected cases).<sup>6–9</sup>

Therefore, many authors argue that immediate treatment under elective surgery is needed regardless of gestational age.<sup>10</sup> For pregnant women before gestational 36 weeks, maternal expectant management of pregnancy maintenance is important until delivery after emergent surgery, which delays delivery until full administration of antenatal corticosteroids, and in a previous study, maternal expectant management periods ranged from 3 days to 13 weeks. In this case, after the surgical approach of the total gastrectomy and Roux-en-Y esophagojejunostomy. We achieved an additional one-week extension of the gestation period until delivery by uncontrolled preterm labor after steroid loading and fetal maturation during expectant management, including close monitoring of the fetal and maternal clinical conditions.

Proper nutrition support is important because it can be related to the morbidity of pregnant women and fetuses. Therefore, nutrition support can be supplied through a nasogastric tube, jejunostomy, or parenteral feeding through peripheral or central intravenous vessels. Recommendations for dietary allowance for pregnant women are 2200–2900 kcal/day with 7.6 g/day protein, 175 g/day carbohydrates, and additional vitamin-mineral supplement requirements.<sup>11</sup> Our patient maintained NPO for five days until the bowel movements recovered. The alternative option for this NPO period was taken into account as total parenteral nutrition (TPN; smofkabiven 80 cc/hr; 2074 kcal/day) via a peripherally inserted central catheter (PICC) to achieve the objective target calories for pregnancy nutrition (2100 kcal/day) of the 2nd trimester through counseling of NST (nutrition support team). However, it should be considered that complications of parenteral nutrition occur frequently and may be severe if clinicians do not change to enteric feeding. As a result, she took earlier delivery at 27+1 weeks by cesarean delivery due to fetal breech presentation and uncontrolled preterm labor within 7 days after maternal emergent surgical repair by visceral incarceration.

Finally, in Table 1, we summarized the clinical features, treatment approaches, and adverse outcomes for mothers and fetuses/neonates in all documented cases associated with maternal diaphragmatic hernia, including congenital (Bochdalek hernia), hiatal, and traumatic hernia during pregnancy. From a clinical perspective, through the reviewed 69 cases, this is the first case of delayed delivery of a pregnant patient who survived after antenatal surgery for a congenital diaphragmatic hernia with gangrene of the entire stomach. Etiological classification of diaphragmatic hernia was 32 (46%) congenital hernia,<sup>7,9,10,12–39</sup> 6 (9%) hiatal hernia,<sup>40–45</sup> 17 (25%) traumatic hernia,<sup>21,23,46–57</sup> 1 congenital hernia combined with hiatal hernia,<sup>7</sup> 13 (19%) unspecified type.<sup>11,58–69</sup>

From the reviewed 69 cases, diaphragmatic hernia demonstrated maternal death in 10% (7/69) of the cases (including 1 case that died within 1 hour on admission) and fetal deaths in 17% (12/69) of cases (FDIU, 2; stillborn, 8; death after birth, 2). Out of 29 cases with signs of bowel obstruction, ischemia, and perforation of herniated organs, <sup>7,9,16–18,22–24,26,28,30,35,37,41,44,64,84,9,53–57,60,64,66,67</sup> there were 6 maternal deaths and 7 fetal deaths (FDIU, 1; stillborn, 4; death after birth, 2). Our data showed that there were 68% (17/25) in 1st and the 2nd trimester, 32% (8/25) in the third trimesters for delayed delivery after surgery with hernia diagnosis, 20% (2/10) in 1st and 2nd trimesters, and 80% (8/10) in 3rd trimesters for simultaneous delivery with surgery. It is important to recognize that premature birth has been reported in 26 of the 69 cases of diaphragmatic hernia, <sup>7,9–11,21,23,24,26–29,32,38,39,41,49–51,55,56,60,64,65,70</sup> resulting in 2 maternal and 7 fetal deaths.

### Conclusion

Any delay in diagnosing and treating abdominal herniated viscera's strangulation will lead to a poor outcome by obstruction, perforation, ischemia, gangrene, or necrosis of visceral organ. Therefore, maternal diaphragmatic hernias during pregnancy should undergo surgical repair under elective surgery through multidisciplinary cooperation. Providing

Author and Date	Age (Years)	Туре	Side	Defect Size (cm)	Herniated Organs	Bowel Obstruction, Ischemia, Perforation of Herniated Organs	Surgical Procedure	Delivery Type	GA at Delivery (wks)	GA at Diagnosis of Hernia (wks)	GA at Surgical Repair (wks)	Maternal Survival	Fetal/ Neonatal Survival
Diddle et al, 1941 <sup>58</sup>	23	Traumatic	Lt	13	Stomach, duodenum, ileum, appendix, T-colon, cecum	Positive	Conservative treatment	NA	38.6	38.6	Non- operative	Death	Stillborn
DeLee et al, 1941 <sup>46</sup>	20	NA	Lt	NA	Intestine	Negative	Conservative treatment	VD	40.3	Postpartum	Non- operative	Alive	Alive
Thompson et al, 1945 <sup>12</sup>	31	Congenital (Bochdalek hernia)	Lt	10	Stomach, intestine, pancreas	Negative	Reduction and diaphragmatic hernia repair	VD	NA	Postpartum	Postpartum	Alive	Alive
Bourgeois et al, 1949 <sup>59</sup>	31	NA	Lt	NA	NA	Negative	Conservative treatment	Vaginal C/ S	38.7	38.6	Non- operative	Alive	Alive
PEARSON et al, 1950 <sup>13</sup>	31	Congenital (Bochdalek hernia)	Lt	7	Stomach	Negative	Reduction and diaphragmatic hernia repair	VD	NA	Postpartum	Postpartum	Alive	Alive
Hushlan et al, 1951 <sup>14</sup>	25	Congenital (Bochdalek hernia)	Lt	NA	NA	Negative	Conservative treatment	VD	NA	Postpartum	Non- operative	Alive	Alive
Osborne et al, 1953 <sup>7</sup>	25	Congenital (Bochdalek hernia) Hiatal	Lt	6	Stomach, T-colon	Positive	Conservative treatment	NA	27.4	17.6	Non- operative	Death	Stillborn
Hobbins et al, 1953, <sup>15</sup>	18	Congenital (Bochdalek hernia)	Lt	I	Colon (splenic flexure and the T-colon with mesentery and omentum)	Negative	Reduction and diaphragmatic hernia repair	VD	Term	lst tri.	lst tri.	Alive	Alive
Gorbach et al, 1956 <sup>40</sup>	45	Hiatal	NA	NA	NA	NA	NA	VD	36.5	36	Postpartum	Alive	Alive
Bernhardt et al, 1966 <sup>47</sup>	26	Traumatic	Lt	7–8	Stomach, SB, colon, spleen	Negative	Reduction and diaphragmatic hernia repair	VD	37	37	Postpartum	Alive	Alive
Craddock et al, 1968 <sup>41</sup>	17	Hiatal	Lt	I	Small gut	Positive	Resection of gangrenous SB and double-layered anastomosis	VD	31.4	31	31	Alive	Stillborn

 Table I The Current Literature on a Diaphragmatic Hernia During Pregnancy

(Continued)

Chae et al

### Table I (Continued).

Author and Date	Age (Years)	Туре	Side	Defect Size (cm)	Herniated Organs	Bowel Obstruction, Ischemia, Perforation of Herniated Organs	Surgical Procedure	Delivery Type	GA at Delivery (wks)	GA at Diagnosis of Hernia (wks)	GA at Surgical Repair (wks)	Maternal Survival	Fetal/ Neonatal Survival
Dudley et al, 1979 <sup>48</sup>	16	Traumatic	Lt	NA	Stomach	Positive	Partial gastrostomy, gastrogastrectomy, and hernia repair	Non- delivery	Non- delivery	18	18	Death	FDIU
Dudley et al, 1979 <sup>48</sup>	41	Traumatic	Lt	NA	Omentum.colon (splenic flexure)	Positive	Partial omentectomy, reduction, and closure of the hernia	VD	NA	Postpartum	Postpartum	Alive	Alive
Gimovsky et al 1983 <sup>9</sup>	20	Congenital (Bochdalek hernia)	Lt	NA	T-Colon	Positive	Colon resection, colostomy	C/S	28	Postpartum	Postpartum	Alive	Death after birth
Mitchell et al, 1983 <sup>49</sup>	18	Traumatic	Lt	NA	T-Colon	Positive	Double-barrel colostomy	C/S	32	24	32	Alive	Alive
FARDY et al, 1984 <sup>42</sup>	25	Hiatal	Lt	NA	Stomach	Negative	Reduction and diaphragmatic hernia repair	VD	38	30	30	Alive	Alive
Rabinovici et al, 1986 <sup>50</sup>	21	Traumatic	Lt	10	Stomach, T-colon, SB	Negative	Reduction/bladder tear - repair, cystostomy	C/S	28	28	28	Alive	Stillborn
REED et al, 1987 <sup>16</sup>	18	Congenital (Bochdalek hernia)	Lt	12	Stomach	Positive	Repair of stomach tear by chest drain, reduction, and diaphragm repair	C/S	36.4	Postpartum	Postpartum	Alive	Alive
Wolfe et al, 1988 <sup>60</sup>	40	NA	Lt	8	Stomach, SB, T-colon, omentum	Positive	SB resection, partially?	C/S	31	30	30.4	Alive	Death after birth
KURZEL et al, 1988 <sup>10</sup>	27	Congenital (Bochdalek hernia)*	Lt	14	Stomach, SB, T-colon, A-colon, pancreas, spleen, Left kidney	Negative	Reduction and marlex mesh patch repair	C/S	36	33	33	Alive	Alive
Henzler et al, 1988 <sup>51</sup>	25	Traumatic	Lt	7	Stomach, T-colon	Negative	NA	VD	33	33	Postpartum	Alive	Alive
Maddox et al, 1991 <sup>52</sup>	33	Traumatic	Lt	7	Stomach, T-colon, spleen	Negative	Reduction and diaphragmatic hernia repair	VD	39	Postpartum	Postpartum	Alive	Alive
Toorians et al, 1993 <sup>17</sup>	29	Congenital (Bochdalek hernia)	Rt	7	T-colon, greater omentum	Positive	Reduction, partial omentectomy	VD	Term	15	15	Alive	Alive
Lacayo et al, 1993 <sup>53</sup>	20	Traumatic	Lt	NA	Mild transverse colon with perforation	Positive	Colon resectomy, omentectomy, reduction and repair	C/S	41	Postpartum	Postpartum	Alive	Alive

1762

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Hill et al,1996 <sup>18</sup>	30	Congenital (Bochdalek hernia)	Lt	5	SB, large bowel	Positive	Reduction and detorsion and resection and repair of ischemic herniated bowel, 2nd look laparotomy	VD	NA	Postpartum	Postpartum	Alive	Alive
Fleyfel et al, 1998 <sup>43</sup>	29	Hiatal	Lt	8	Stomach	Negative	Reduction and diaphragmatic hernia repair	C/S	39	28.?	28.?+3	Alive	Alive
Ortega et al, 1998 <sup>19</sup>	23	Congenital (Bochdalek hernia)	Lt	6	SB, T-colon	Negative	Reduction and diaphragmatic hernia repair	VD	NA	Postpartum	Postpartum	Alive	Alive
Dietrich et al, 2001 <sup>54</sup>	19	Traumatic	Lt	NA	T-colon	Positive	T-colectomy, gastrostomy tube placement	C/S	36.1	36.1	Postpartum	Alive	Alive
llaloo et al, 2001 <sup>61</sup>	32	NA	Lt	NA	T-Colon	Negative	Reduction and diaphragmatic hernia repair	VD	NA	Postpartum	Postpartum	Alive	Alive
Indar et al, 2001 <sup>55</sup>	24	Traumatic	Lt	8	Colon	Positive	Double-barreled colostomy	VD	34	34	34	Death	Stillborn
Indar et al, 2001 <sup>55</sup>	25	Traumatic	Lt	7	Stomach, SB, colon	NA	Died in I hours after admission	Non- delivery	Non- delivery	36	Non- operative	Death	FDIU
Indar et al, 2001 <sup>55</sup>	21	Traumatic	Lt	3	Stomach, SB	Negative	Reduction and diaphragmatic hernia repair	C/S	35	35	35	Alive	Alive
Indar et al, 2001 <sup>55</sup>	29	Traumatic	Lt	4	Colon	Negative	Reduction and diaphragmatic hernia repair	VD	38	20	20	Alive	Alive
Williams et al, 2003 <sup>21</sup>	36	Traumatic	Lt	6	Stomach, small bowel, T-colon	Negative	Reduction and diaphragmatic hernia repair	VD	33	28	28	Alive	Alive
Williams et al, 2003 <sup>21</sup>	32	Congenital (Bochdalek hernia) *	Lt	2	Stomach, omentum	Negative	Reduction, partial omentectomy	VD	NA	Postpartum	Postpartum	Alive	Alive
Genc et al, 2003 <sup>20</sup>	30	Congenital (Bochdalek hernia)	Lt	8	Stomach., T-colon	Negative	Reduction and diaphragmatic hernia repair	VD	39	29	31	Alive	Alive
Hamoudi et al, 2004 <sup>62</sup>	41	NA	Lt	7	Stomach, T-colon, spleen	Negative	Reduction and diaphragmatic hernia repair	VD	NA	Postpartum	Postpartum	Alive	Alive
Luu et al, 2006 <sup>24</sup>	34	Congenital (Bochdalek hernia)	Lt	2	Stomach with perforation	Positive	Resection of a necrotic portion of the stomach	VD	34	Postpartum	Postpartum	Alive	Alive
Eglinton et al, 2006 <sup>23</sup>	30	Congenital (Bochdalek hernia) *	Lt	NA	Stomach	Positive	Reduction, suture of perforation site of stomach due to chest tube	C/S	28	28	28	Alive	Alive

(Continued)

### Table I (Continued).

Author and Date	Age (Years)	Туре	Side	Defect Size (cm)	Herniated Organs	Bowel Obstruction, Ischemia, Perforation of Herniated Organs	Surgical Procedure	Delivery Type	GA at Delivery (wks)	GA at Diagnosis of Hernia (wks)	GA at Surgical Repair (wks)	Maternal Survival	Fetal/ Neonatal Survival
Eglinton et al, 2006 <sup>23</sup>	27	Congenital (Bochdalek hernia) *	Lt	NA	Stomach, T-colon, spleen	Positive	Resection of stomach	C/S	37	27	27	Alive	Alive
Eglinton et al, 2006 <sup>23</sup>	32	Traumatic	Rt	NA	Liver, colon	Negative	Reduction and diaphragmatic hernia repair	C/S	34	34	34	Alive	Alive
Barbetakis et al, 2006 <sup>22</sup>	31	Congenital (Bochdalek hernia)	Lt	7	Stomach, large segment of T-colon, Rt-colon, omentum	Positive	Segmental resection of large intestine and end-to-end anastomosis	C/S	39	23	23	Alive	Alive
Hanekamp et al, 2006 <sup>63</sup>	26	NA	Lt	NA	Stomach with perforation, colon, spleen	Negative	Reduction. Repair of perforated stomach	NA	NA	5 m	5 m	Alive	?
Osman et al, 2007 <sup>65</sup>	32	NA	Lt	NA	Stomach	Negative	Reduction and diaphragmatic hernia repair	VD	35	27	27	Alive	Alive
Rajasingam et al, 2007 <sup>26</sup>	24	Congenital (Bochdalek hernia) *	Lt	NA	Stomach	Positive	Partial gastrostomy (Polya type)	C/S	33.6	33.6	33.6	Alive	Alive
Jai et al, 2007 <sup>56</sup>	27	Traumatic	Lt	NA	Stomach, T-colon, omentum	Positive	Resection of stomach, excision of omentum, esojejunal anastomosis	VD	32	Postpartum	Postpartum	Death	Alive
Agarwal et al, 2007 <sup>64</sup>	28	NA*	Lt	NA	Stomach, SB, colon, spleen	Positive	Partial gastrectomy and gastrojejunostomy diaphragmatic hernia repair	VD	27.4	27.4	Postpartum	Alive	Alive
Pai et al, 2007 <sup>25</sup>	26	Congenital (Bochdalek hernia)	Rt	NA	Liver. A-colon, hepatic flexure, T-colon	Negative	Reduction and diaphragmatic hernia repair	C/S	Term	Postpartum	Postpartum	Alive	Alive
Palanivelu et al, 2008 <sup>27</sup>	23	Congenital (Bochdalek hernia)	Lt	5	Stomach, colon, omentum	Negative	Reduction/ to fix (stomach- peritoneum, colopexy)	NA	9 m	6 m	6 m	Alive	Alive
Sano et al, 2008 <sup>28</sup>	25	Congenital (Bochdalek hernia)	Lt	2	SB. Omentum	Positive	Resection of ileum and cecum	C/S	28	28	28	Alive	Alive
Ting et al, 2008 <sup>66</sup>	42	NA	Lt	NA	Stomach, SB	Positive	Partial gastrectomy	NA	NA	19	19	Alive	Alive

Chae et al

Hunter et al,	31	Congenital	Lt	NA	Stomach, SB,	Negative	Reduction and diaphragmatic	C/S	27	27	27	Alive	Stillborn
2009 <sup>29</sup>		(Bochdalek hernia)			T-colon, spleen	Ŭ	hernia repair						
Riad et al, 2009 <sup>57</sup>	27	Traumatic	Lt	NA	SB, large bowel, spleen	Positive	Large and small bowel resection, ileostomy, splenectomy,	C/S	37	Postpartum	Postpartum	Death	Alive
Rubin et al, 2009 <sup>44</sup>	27	Hiatal	Lt	NA	Stomach. Duodenum, colon	Positive	Reduction, omental wrapping section, diaphragm repair	VD	NA	Postpartum	Postpartum	Alive	Alive
lslah et al, 2010 <sup>30</sup>	30	Congenital (Bochdalek hernia)	Lt	6	Stomach, SB, colon, appendix	Positive	Sleeve gastrectomy	C/S	NA	30	30	Alive	Alive
Morcillo et al, 2010 <sup>31</sup>	35	Congenital (Bochdalek hernia)	Lt	15	Stomach, greater omentum, colon	Negative	Reduction and diaphragmatic hernia repair	C/S	38	15	15	Alive	Alive
Schwentner et al, 2011 <sup>67</sup>	30	NA	Lt	NA	Stomach	Positive	Reduction and diaphragmatic hernia repair	C/S	38.5	12	12	Alive	Alive
Chen et al, 2011 <sup>11</sup>	26	NA	Lt	8	Stomach, SB, T-colon, spleen	Negative	Reduction and diaphragmatic hernia repair	VD	24.3	23.3	23.3	Alive	Artificial abortion
Ngai et al, 2012 <sup>32</sup>	31	Congenital (Bochdalek hernia)	Lt	NA	MRI findings: stomach, SB, colon, spleen, pancreatic body	Negative	Reduction and diaphragmatic hernia repair	C/S	31	29	Postpartum	Alive	Alive
Chen et al, 2012 <sup>45</sup>	18	Hiatal	Lt	20	Stomach, SB, omentum	Negative	Reduction and diaphragmatic hernia repair	VD	15	Postpartum	Postpartum	Alive	Stillborn
Flamée et al, 2012 <sup>69</sup>	33	NA	Lt	NA	NA	Negative	NA	NA	NA	21	NA	?	Stillborn
Benson et al, 2012 <sup>68</sup>	23	NA	Lt	NA	Colon	Negative	Reduction and diaphragmatic hernia repair	C/S	38	20	30	Alive	Alive
Hamaji et al, 2013 <sup>33</sup>	39	Congenital (Bochdalek hernia)	Lt	5	T-colon, omentum	Negative	Reduction and diaphragmatic hernia repair	VD	NA	Postpartum	Postpartum	Alive	Alive
Kawashima et al, 2013 <sup>34</sup>	26	Congenital (Bochdalek hernia)	Lt	10	Stomach, T-colon, spleen	Negative	Reduction and diaphragmatic hernia repair	C/S	NA	26	26	Alive	Alive
Wieman et al, 2013 <sup>35</sup>	42	Congenital (Bochdalek hernia)	Rt	7	Colon, cecum (several serosal tears in colon, cecum)	Positive	Reduction/serosal tear- suture	NA	NA	27	27	Alive	Alive

(Continued)

Chae et al

#### Table I (Continued).

Author and Date	Age (Years)	Туре	Side	Defect Size (cm)	Herniated Organs	Bowel Obstruction, Ischemia, Perforation of Herniated Organs	Surgical Procedure	Delivery Type	GA at Delivery (wks)	GA at Diagnosis of Hernia (wks)	GA at Surgical Repair (wks)	Maternal Survival	Fetal/ Neonatal Survival
Ali et al, 2014 <sup>36</sup>	25	Congenital (Bochdalek hernia)	Rt	NA	Small bowel	Negative	Reduction and mesh diaphragmatic repair	NA	NA	2nd tri.	2nd tri.	Alive	Alive
Debergh et al, 2014 <sup>37</sup>	28	Congenital (Bochdalek hernia)	Rt	5	Small bowel	Positive	Reduction and mesh diaphragmatic repair	NA	Term	16	16	Alive	Alive
Hernández et al, 2015 <sup>38</sup>	32	Congenital (Bochdalek hernia)	Lt	6	A-colon, T-colon, SB	Negative	Reduction and diaphragmatic hernia repair	C/S	32	29	32	Alive	Alive
Reddy et al, 2018 <sup>39</sup>	30	Congenital (Bochdalek hernia)	Lt	NA	Stomach, SB, colon, appendix, spleen, omentum	Negative	Reduction and diaphragmatic hernia repair	C/S	32.3	31.3	32.3	Alive	Alive

Notes: The first trimester is defined as 0–13 6/7 weeks of gestation from the last menstrual period; the second trimester is defined as 14 0/7–27 6/7 weeks of gestation from the last menstrual period; the third trimester is defined as 28 0/7–42 0/7 weeks of gestation from last menstrual period. \*Six patients were diagnosed with a diaphragmatic hernia before their pregnancies.

Abbreviations: Rt, right; Lt, left; NA, not available; T-colon, Transverse colon; A-colon, ascending colon; SB, small bowel; C/S, cesarean section; tri., trimester, Wks, Weeks BH, Bochdalek hernia; NA, not available; VD, vaginal delivery; C/S, cesarean section.

adequate nutritional support for mothers and fetuses during postoperative pregnancy management care is crucial to reducing morbidity and mortality.

### **Ethics Approval and Informed Consent**

The patient reported in this manuscript has agreed in written informed consent to participate in this study and to publish details of her case and accompanying photographs. Institutional approval is not required to publish the case details in accordance with local legislation and institutional requirements.

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# **Author Contributions**

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis, and interpretation, or all these areas; took part in drafting, revising, or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

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### References

- 1. McGivern MR, Best KE, Rankin J, et al. Epidemiology of congenital diaphragmatic hernia in Europe: a register-based study. Arch Dis Child Fetal Neonatal Ed. 2015;100(2):F137-44. doi:10.1136/archdischild-2014-306174
- 2. Mullins ME, Stein J, Saini SS, Mueller PR. Prevalence of incidental Bochdalek's hernia in a large adult population. *AJR Am J Roentgenol*. 2001;177(2):363-366. doi:10.2214/ajr.177.2.1770363
- 3. Brown SR, Horton JD, Trivette E, Hofmann LJ, Johnson JM. Bochdalek hernia in the adult: demographics, presentation, and surgical management. *Hernia*. 2011;15(1):23–30. doi:10.1007/s10029-010-0699-3
- 4. Hedblom CA. THE SELECTIVE SURGICAL TREATMENT OF DIAPHRAGMATIC HERNIA. Ann Surg. 1931;94(4):776–785. doi:10.1097/00000658-193110000-00031
- 5. Choi JY, Yang SS, Lee JH, et al. Maternal bochdalek hernia during pregnancy: a systematic review of case reports. *Diagnostics*. 2021;11(7). doi:10.3390/diagnostics11071261
- 6. Hodge K. Diaphragmatic hernia complicated by gastric ulcer and pregnancy. Br J Radiol. 1950;23(273):573. doi:10.1259/0007-1285-23-273-573-b
- 7. Osborne W, Foster C. Diaphragmatic hernia complicating pregnancy. Am J Obstet Gynecol. 1953;66(3):682-684. doi:10.1016/0002-9378(53) 90087-8
- 8. Flood JL. Foramen of Bochdalek hernia in pregnancy. J Indiana State Med Assoc. 1963;56:32-34.
- 9. Gimovsky ML, Schifrin BS. Incarcerated foramen of Bochdalek hernia during pregnancy. A case report. J Reprod Med. 1983;28(2):156–158.
- 10. Kurzel RB, Naunheim KS, Schwartz RA. Repair of symptomatic diaphragmatic hernia during pregnancy. Obstet Gynecol. 1988;71(6):869-871.
- 11. Chen Y, Hou Q, Zhang Z, Zhang J, Xi M. Diaphragmatic hernia during pregnancy: a case report with a review of the literature from the past 50 years. J Obstet Gynaecol Res. 2011;37(7):709–714. doi:10.1111/j.1447-0756.2010.01451.x
- 12. Thompson J, Le Blanc LJ. Congenital diaphragmatic hernia: visceral strangulation complicating delivery. Am J Surg. 1945;67(1):123-130. doi:10.1016/0002-9610(45)90335-7
- 13. Pearson SC, Pillsbury SG, Mccallum M. Strangulated diaphragmatic hernia complicating delivery. J Am Med Assoc. 1950;144(1):22-24. doi:10.1001/jama.1950.62920010004006b
- 14. Hushlan S. Diaphragmatic hernia in pregnancy; significance and danger. Conn State Med J. 1951;15(11):969.
- 15. Hobbins W, Hurwitz C. Incarcerated diaphragmatic hernia of the colon occurring during pregnancy. N Engl J Med. 1953;249(19):773–774. doi:10.1056/nejm195311052491905
- 16. Reed MW, De Silva PH, Mostafa SM, Collins FJ. Diaphragmatic hernia in pregnancy. Br J Surg. 1987;74(5). doi:10.1002/bjs.1800740540
- 17. Toorians AW, Drost-Driessen MA, Snellen JP, Smeets RW. Acute hernia of Bochdalek during pregnancy. Hyperemesis for the first time in a third pregnancy? *Acta Obstet Gynecol Scand.* 1992;71(7):547–549. doi:10.3109/00016349209041449
- 18. Hill R, Heller MB. Diaphragmatic rupture complicating labor. Ann Emerg Med. 1996;27(4):522-524. doi:10.1016/S0196-0644(96)70248-2
- 19. Ortega-Carnicer J, Ambros A, Alcazar R. Obstructive shock due to labor-related diaphragmatic hernia. Crit Care Med. 1998;26(3):616-618. doi:10.1097/00003246-199803000-00042

- 20. Genc MR, Clancy TE, Ferzoco SJ, Norwitz E. Maternal congenital diaphragmatic hernia complicating pregnancy. *Obstet Gynecol.* 2003;102 (5):1194–1196.
- 21. Williams M, Appelboam R, McQuillan P. Presentation of diaphragmatic herniae during pregnancy and labour. *Int J Obstet Anesth.* 2003;12 (2):130–134. doi:10.1016/S0959-289X(02)00189-9
- 22. Barbetakis N, Efstathiou A, Vassiliadis M, Xenikakis T, Fessatidis I. Bochdaleck's hernia complicating pregnancy: case report. *World J Gastroenterol*. 2006;12(15):2469–2471. doi:10.3748/wjg.v12.i15.2469
- 23. Eglinton TW, Coulter GN, Bagshaw PF, Cross LA. Diaphragmatic hernias complicating pregnancy. *ANZ J Surg.* 2006;76(7):553–557. doi:10.1111/j.1445-2197.2006.03776.x
- 24. Luu TD, Reddy VS, Miller DL, Force SD. Gastric rupture associated with diaphragmatic hernia during pregnancy. *Ann Thorac Surg.* 2006;82 (5):1908–1910. doi:10.1016/j.athoracsur.2006.02.083
- 25. Pai S, Balu K, Madhusudhanan J. Congenital diaphragmatic hernia complicating pregnancy: a case report. Internet J Gynecol Obstet. 2007;9:1.
- 26. Rajasingam D, Kakarla A, Jones A, Ash A. Strangulated congenital diaphragmatic hernia with partial gastric necrosis: a rare cause of abdominal pain in pregnancy. *Int J Clin Pract.* 2007;61(9):1587–1589. doi:10.1111/j.1742-1241.2005.00763.x
- 27. Palanivelu C, Rangarajan M, Maheshkumaar G, Parthasarathi R. Laparoscopic mesh repair of a Bochdalek diaphragmatic hernia with acute gastric volvulus in a pregnant patient. *Singapore Med J.* 2008;49(1):e26–e28.
- 28. Sano A, Kato H, Hamatani H, et al. Diaphragmatic hernia with ischemic bowel obstruction in pregnancy: report of a case. *Surg Today*. 2008;38 (9):836. doi:10.1007/s00595-007-3718-y
- 29. Hunter JD, Nimmagadda J, Quayle A. Maternal congenital diaphragmatic hernia causing cardiovascular collapse during pregnancy. *Br J Hosp Med.* 2009;70(3):166–167. doi:10.12968/hmed.2009.70.3.40573
- 30. Islah M, Jiffre D. A rare case of incarcerated bochdalek diaphragmatic hernia in a pregnant lady. Med J Malay. 2010;65(1):75-76.
- Morcillo-López I, Hidalgo-Mora JJ, Baamonde A, Díaz-García C. Gastric and diaphragmatic rupture in early pregnancy. *Interact Cardiovasc Thorac Surg.* 2010;11(5):713–715. doi:10.1510/icvts.2010.246140
- 32. Ngai I, Sheen -J-J, Govindappagari S, Garry DJ. Bochdalek hernia in pregnancy. Case Rep. 2012;2012:bcr2012006859.
- Hamaji M, Burt BM, Ali SO, Cohen DM. Spontaneous diaphragm rupture associated with vaginal delivery. *Gen Thorac Cardiovasc Surg.* 2013;61 (8):473–475. doi:10.1007/s11748-012-0146-8
- 34. Kawashima K, Nakahata K, Negoro T, Nishikawa K. "Spontaneous" Rupture of the Maternal Diaphragm. *Anesthesiology*. 2013;118(2):445. doi:10.1097/ALN.0b013e31825d95ed
- 35. Wieman E, Pollock G, Moore BT, Serrone R. Symptomatic right-sided diaphragmatic hernia in the third trimester of pregnancy. *J Soc Laparoend Surg.* 2013;17(2):358. doi:10.4293/108680812X13517013318157
- 36. Ali SA, Haseen MA, Beg MH. Agenesis of right diaphragm in the adults: a diagnostic dilemma. Indian J Chest Dis Allied Sci. 2014;56(2):121–123. doi:10.5005/ijcdas-56-2-121
- 37. Debergh I, Fierens K. Laparoscopic repair of a Bochdalek hernia with incarcerated bowel during pregnancy: report of a case. *Surg Today*. 2014;44 (4):753–756. doi:10.1007/s00595-012-0441-0
- Hernández-Aragon M, Rodriguez-Lazaro L, Crespo-Esteras R, Ruiz-Campo L, Adiego-Calvo I, Campillos-Maza J. Bochdalek diaphragmatic hernia complicating pregnancy in the third trimester: case report. Obstet Gynaecol Cases. 2015;2. doi:10.23937/2377-9004/1410057
- 39. Reddy M, Kroushev A, Palmer K. Undiagnosed maternal diaphragmatic hernia-a management dilemma. *BMC Pregnancy Childbirth*. 2018;18 (1):1-5. doi:10.1186/s12884-018-1864-4
- 40. Gorbach AC, Reid DE. Hiatus hernia in pregnancy. New Engl J Med. 1956;255(11):517-519. doi:10.1056/NEJM195609132551107
- 41. Craddock D, Hall J. Strangulated diaphragmatic hernia complicating pregnancy. Br J Surg. 1968;55(7):559-560. doi:10.1002/bjs.1800550717
- 42. Fardy HJ. Vomiting in late pregnancy due to diaphragmatic hernia. Case report. Br J Obstet Gynaecol. 1984;91(4):390–392. doi:10.1111/j.1471-0528.1984.tb05930.x
- 43. Fleyfel M, Provost N, Ferreira JF, Porte H, Bourzoufi K. Management of diaphragmatic hernia during pregnancy. Anesth Analg. 1998;86 (3):501-503.
- 44. Rubin S, Sandu S, Durand E, Baehrel B. Diaphragmatic rupture during labour, two years after an intra-oesophageal rupture of a bronchogenic cyst treated by an omental wrapping. *Interact Cardiovasc Thorac Surg.* 2009;9(2):374–376. doi:10.1510/icvts.2009.203075
- 45. Chen X, Yang X, Cheng W. Diaphragmatic tear in pregnancy induced by intractable vomiting: a case report and review of the literature. J Mat Fet Neonat Med. 2012;25(9):1822–1824. doi:10.3109/14767058.2011.640371
- 46. Diddle A, Tidrick R. Diaphragmatic hernia associated with pregnancy. Am J Obstet Gynecol. 1941;41(2):317–321. doi:10.1016/S0002-9378(41) 91184-5
- 47. Bernhardt LC, Lawton BR. Pregnancy complicated by traumatic rupture of the diaphragm. Am J Surg. 1966;112(6):918–922. doi:10.1016/0002-9610(66)90151-6
- 48. Dudley AG, Teaford H, Gatewood TS. Delayed traumatic rupture of the diaphragm in pregnancy. Obstet Gynecol. 1979;53(3 Suppl):25S-27S.
- 49. Mitchell R, Teare A. Traumatic diaphragmatic hernia in pregnancy. South Af Med J. 1983;63(13):474.
- 50. Rabinovici J, Czerniak A, Rabau MY, Avigad I, Wolfstein I. Diaphragmatic rupture in late pregnancy due to blunt injury. 1986;17 (6):416-417. doi:10.1016/0020-1383(86)90087-2
- 51. Henzler M, Martin ML, Young J. Delayed diagnosis of traumatic diaphragmatic hernia during pregnancy. Ann Emerg Med. 1988;17(4):350–353. doi:10.1016/S0196-0644(88)80780-7
- 52. Maddox P, Mansel R, Butchart E. Traumatic rupture of the diaphragm: a difficult diagnosis. *Injury*. 1991;22(4):299-302. doi:10.1016/0020-1383(91)90010-C
- 53. Lacayo L, Taveras JM, Sosa N, Ratzan KR. Tension fecal pneumothorax in a postpartum patient. Chest. 1993;103(3):950-951. doi:10.1378/ chest.103.3.950
- 54. Dietrich CL, Smith CE. Anesthesia for cesarean delivery in a patient with an undiagnosed traumatic diaphragmatic hernia. *Anesthesiology*. 2001;95 (4):1028–1031. doi:10.1097/00000542-200110000-00038
- 55. Indar A, Bornman P, Beckingham I. Late presentation of traumatic diaphragmatic hernia in pregnancy. Ann R Coll Surg Engl. 2001;83(6):392.
- 56. Jai SR, Bensardi F, Hizaz A, Chehab F, Khaiz D, Bouzidi A. A late post-traumatic diaphragmatic hernia revealed during pregnancy by post-partum respiratory distress. *Arch Gynecol Obstet*. 2007;276(3):295–298. doi:10.1007/s00404-007-0347-z

- 57. Riad M, Shervington J, Woodward Z. Maternal death due to ruptured diaphragmatic hernia. J Obstet Gynaecol. 2009;29(7):669. doi:10.1080/01443610903078888
- 58. DeLee S, Gilson B. Diaphragmatic hernia complicating the puerperium. Am J Obstet Gynecol. 1941;41(5):904. doi:10.1016/S0002-9378(41)90889-X
- Bourgeois GA, Hood WT. Strangulated Diaphragmatic Hernia Complicating Pregnancy: report of a Case. New Engl J Med. 1949;241(4):150–151. doi:10.1056/NEJM194907282410406
- 60. Wolfe CA, Peterson MW. An unusual cause of massive pleural effusion in pregnancy. Thorax. 1988;43(6):484-485. doi:10.1136/thx.43.6.484
- Kaloo PD, Child A, Child A. Postpartum diagnosis of a maternal diaphragmatic hernia. Austra New Zeal J Obstet Gynaecol. 2001;41(4):461–463. doi:10.1111/j.1479-828X.2001.tb01333.x
- 62. Hamoudi D, Bouderka M, Benissa N, Harti A. Diaphragmatic rupture during labor. Int J Obstet Anesth. 2004;13(4):284-286. doi:10.1016/j. ijoa.2004.04.001
- 63. Hanekamp LA, Toben FM. A pregnant woman with shortness of breath. Neth J Med. 2006;64(3):84-95.
- 64. Agarwal P, Ash A. Gastric volvulus: a rare cause of abdominal pain in pregnancy. J Obstet Gynaecol. 2007;27(3):313-314. doi:10.1080/ 01443610701241183
- 65. Osman I, McKernan M, Rae D. Normal vaginal delivery following rupture of the maternal diaphragm in pregnancy. J Obstet Gynaecol. 2007;27 (6):625–627. doi:10.1080/01443610701554247
- 66. Ting JYS. Difficult diagnosis in the emergency department: hyperemesis in early trimester pregnancy because of incarcerated maternal diaphragmatic hernia. *Emerg Med Austral*. 2008;20(5):441–443. doi:10.1111/j.1742-6723.2008.01119.x
- 67. Schwentner L, Wulff C, Kreienberg R, Herr D. Exacerbation of a maternal hiatus hernia in early pregnancy presenting with symptoms of hyperemesis gravidarum: case report and review of the literature. *Arch Gynecol Obstet.* 2011;283(3):409–414. doi:10.1007/s00404-010-1719-3
- 68. Benson CC, Valente AM, Economy KE, et al. Discovery and management of diaphragmatic hernia related to abandoned epicardial pacemaker wires in a pregnant woman with {S, L, L} transposition of the great arteries. *Congenit Heart Dis.* 2012;7(2):183–188. doi:10.1111/j.1747-0803.2011.00547.x
- 69. Flamée P, Pregardien C. Tension gastrothorax causing cardiac arrest. CMAJ. 2012;184:1.
- Chen Y, Bai J, Guo Y, Zhang G. The simultaneous repair of an irreducible diaphragmatic hernia while carrying out a cesarean section. Int J Surg Case Rep. 2013;4(9):771–772. doi:10.1016/j.ijscr.2013.06.002

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