


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

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## Dear editor

I feel honored to publish our manuscript titled “Maternal strangulated diaphragmatic hernia with gangrene of the entire stomach during Pregnancy: A Case Report and Review of the Recent Literature” on behalf of my co-authors in the International Journal of Women's Health.

Thank you for your interest and insightful comments on this paper. I would like to note that this paper is a case report prepared to aid in effective treatment by reporting and reviewing rare cases, given the nature of the disease.

Regarding the reply to the comments on this paper

1. If there is “Review of the recent literature” in the title, it is presumed that the review is about the condition in the title. The authors made the review of MDH in general, not about strangulated gangrenous stomachs.
2. The authors state the exacerbation of symptoms of asymptomatic adult Bochdalek “hernia with” the reference by Brown et al. That article mentions pregnancy as a precipitating factor for acute MDH but without details or systematic analysis and correlation between MDH and pregnancy. After the description of MDH in pregnant patients, the authors state about poor outcomes “due to obstruction, perforation, ischemia, gangrene, or necrosis of internal organs” referencing the article on the general population, published in 1931. In the Discussion, this is explained using correct reference by Choi et al.
3. First, what is multiple passive collapse?
4. The gastroesophageal junction and the duodenal 33 bulb were squeezing and stretching”. These are dynamic terms that cannot be seen in static images.
5. The authors then stated, “The estimated fetal weight was 988g”. It is a very precise weight measurement by ultrasound and the technique for such precision measurement should be described.
6. With “blood pressure of 95/72 mmHg, pulse rate of 160/ min, body temperature of 37.2° C” I doubt that “respiratory rate of 20/ min” is correct. With hypotension and pulse of 160/min, I presume it was higher.
7. While MDH is not always an emergency, the description in this case indicates an emergency situation.
8. The presence of the stomach and transverse colon in the thorax without complete obstruction or indirect signs of ischemia is noted. The stomach and transverse colon are in the thorax without complete obstruction or indirect signs of ischemia. Clinically, the peritonism was not described.
9. A splenectomy was performed due to severe splenomegaly and necrotic changes. While splenic infarcts are often treated conservatively, this case necessitated surgical intervention.

10. The diaphragmatic defect of 10 cm was closed with sutures, a challenging procedure that was necessary in this case.
11. Further, the authors write, “a cesarean delivery due to labor pain”. Why “labor pain”. It sounds like something was wrong. Labor is accompanied by pain due to uterine contractions. The gestation was 27 weeks. Was the patient on cardiotocography and what about postoperative fetal status?
12. The use of prophylactic tocolysis with ritodrine to inhibit preterm labor, although not typically indicated in acute abdominal conditions in pregnancy, was deemed necessary.
13. In the Discussion, there is no formal review, neither narrative nor systematic. Only maternal and fetal mortality of 69 cases was calculated. The authors reviewed all types of MDH. In the same year (2023), we published a systematic review of MDH in pregnancy of all types, with 158 cases (Chae et al “only” 69). This significant difference resulted in more robust conclusions. In our article, we made a detailed analysis of risk factors, correlations between many factors, and outcomes. Finally, we constructed a treatment algorithm not found in the literature. Maybe the authors were unaware of our article. Reviewed and published at the same time.

First, in 2021, Choil and my co-worker reported a systematic review of congenital Bochdalek hernia during pregnancy.<sup>1</sup>

However, the strength of this new case report lies in our comprehensive review of all reported cases of pregnant women with strangulated diaphragmatic hernia with gangrene, including acquired and traumatic hernias, as well as congenital cases occurring during pregnancy. We also have updated a detailed list of these rare cases.

Therefore, I am afraid I must disagree with this comment: “The authors made the review of MDH in general, not about strangulated gangrenous stomachs”.

Second, Brown et al conducted an English language literature search using PubMed, EMBASE, and Google Scholar for case reports, series, and literature reviews relating to congenital Bochdalek Hernia in adults.<sup>2</sup> The literature review encompassed 141 articles, including 173 cases from 31 countries. Precipitating factors or triggering events were identified in 25% of the cases of congenital Bochdalek Hernia in adults. Among these, pregnancy was the most common precipitating factor, representing 34% of cases with at least one precipitating factor, or 8% of all cases.

Meanwhile, in 1925, Carl Hedblom<sup>3</sup> surveyed 375 diaphragmatic hernias of various types, including 44 congenital hernias. Later, in 1931, he studied 476 cases of patients who had undergone surgery.<sup>4</sup> His findings indicated that the likelihood of strangulation in diaphragmatic hernias varied with their etiological type. Among the 476 cases, 55 were congenital, with 36.3% experiencing obstruction; 64 were acquired, with 15.6% obstructed; 145 resulted from war injuries, with 47.5% obstructed; and 186 followed trauma in civilian life, mostly from knife stabbings or blunt trauma, with 20.2% obstructed. These findings suggested that hernias more prone to strangulation are a stronger indication for immediate repair.

Therefore, the references to Brown et al and Hedblom et al in this paper are correctly mentioned in the introduction and are not incorrect.

The co-worker professor of radiology provided insights regarding your queries about the third and fourth points. Third, radiological findings on the chest X-ray related to a diaphragmatic hernia showed multiple areas of passive collapse. Numerous regions of passive collapse are attributed to the inadequate expansion of the lungs due to the diaphragmatic hernia rising towards the diaphragm, resulting in a reduced chest cavity size. Fourth, There is no evidence to suggest that the terms “squeezing” and “stretching” about the gastroesophageal junction and duodenal bulbs are exclusively used in dynamic imaging.

Fifth, contrary to what you suggested, the estimated fetal weight of 988g is accurately described in the paper by an emergency obstetric ultrasound, such as echocardiography and Doppler.

Sixth, A high pulse rate and low blood pressure indicate a compensatory response to stress or underlying conditions, which could also affect the respiratory rate. The patient’s respiratory rate was measured at 20 breaths per minute in the vital signs taken after they visited the emergency room.

Seventh, unlike your comment, Seventh, contrary to your comment, this case report focuses on diaphragm hernia in pregnant women who suddenly exacerbate the symptoms of asymptomatic adult Bochdalek hernia, not the mother’s diaphragm hernia in general.

Therefore, if initial symptoms such as pain, nausea, and vomiting in the upper abdomen are misdiagnosed, and diagnosis and treatment are delayed, the results may not be suitable due to obstruction, perforation, ischemia, gangrene, and visceral necrosis, so we emphasized early diagnosis is crucial for appropriate therapy. Until now, there have been limited attempts to review the maternal and fetal outcome data about maternal hernia during pregnancy systematically.

Therefore, if initial symptoms like pain, nausea, and vomiting in the upper abdomen are misdiagnosed, and diagnosis and treatment are delayed, the outcomes could be unfavorable due to complications such as obstruction, perforation, ischemia, gangrene, and visceral necrosis. To date, there have been limited efforts to systematically review maternal and fetal outcome data regarding maternal hernia during pregnancy. Hence, we emphasize that early diagnosis is crucial for effective treatment.

Eighth, we did not state that “the stomach and lateral colon are in the chest without any complete obstruction or indirect signs of ischemia”. Therefore, the above claim would be a distortion of the facts.

Ninth, the colleague’s professor of surgery provided insights into your queries regarding the decision to perform a splenectomy due to severe splenomegaly and necrotic changes. While splenic infarcts are often managed conservatively, this particular case required surgical intervention. Nonoperative management for splenic infarction may not be suitable for hemodynamically unstable patients, those presenting with generalized peritonitis, or those with other intra-abdominal injuries that necessitate surgical exploration. The decision between splenectomy and splenic salvage techniques, such as splenorrhaphy or partial splenectomy, depends on the injury’s severity, associated injuries, the patient’s overall health, and the surgeon’s experience. In these cases, the decision-making process is guided by the surgeon’s expertise and the resources available at the facility.<sup>5</sup>

We discussed following a consultation into your 10th, 11th, and 12th queries with a colleague’s professor of a cardiothoracic surgeon and a specialized obstetrician.

As for the treatment guidelines for hernia defects, primary repair for hernia is the best treatment, but closure with excessive tension must be avoided to prevent hernia recurrence. In addition, if a tension-free closure due to a large defect is not possible, surgical techniques of past flaps and recent prosthetic materials may be used.

In our literature review on diaphragmatic hernia during pregnancy, we found that Chen et al, in their 2011 study, successfully performed simple suturing surgery on a 20 cm-sized diaphragmatic hernia defect.<sup>6</sup> Although this seems like an excessively large defect, primary suture surgery was feasible because the tissue was dilated.

The other important point is expectant treatment until pregnancy termination after primary emergent or elective surgical intervention. It is important to prevent preterm labor or preterm birth during maternal expectant management until delivery after surgery. Therefore, the use of prophylactic tocolysis with ritodrine to inhibit preterm labor, although not typically indicated in acute abdominal conditions in pregnancy, was deemed necessary.

Expectant management until the termination of pregnancy, especially following a primary emergency or elective surgical intervention, is crucial in cases of pregnancy-related maternal diaphragmatic hernia. Consequently, prophylactic tocolysis, such as ritodrine, to inhibit premature labor or birth during this period of expectant management from post-surgery until delivery is considered essential. The patient received a tocolytic agent by continuous intravenous infusion of ritodrine to inhibit preterm labor. The patient 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. The decision for cesarean delivery due to labor pain at 27 weeks gestation was based on the patient’s clinical status and fetal monitoring.

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.

The limitations of research on rare tumors are as follows. i) sample size limitation, ii) lack of data and quality issues, iii) difficulty in clinical research, v) lack of pathophysiologic understanding, and vi) absence of standard treatment guidelines. Therefore, in this paper, we tried to overcome these limitations and provide patients with better treatment options.

Unfortunately, we could not concur with all of your critical viewpoints. You did not provide a concrete basis for your statements, and most of the points raised appear to be personal opinions.

Although your recent paper has been analyzed with a lot of research data, the lack of explicit mention of how data loss was handled and the absence of a bibliography significantly undermine the reliability of the results.

Regarding the treatment guidelines in your journal, I suggest that the guidelines include information on the time interval from hernia diagnosis to hernia surgery, such as immediate hernia surgery following diagnosis or delayed hernia surgery after expectant management. Additionally, it's important to consider the specifics of treating maternal diaphragmatic hernia during pregnancy, including options like delayed delivery post-hernia surgery, concurrent delivery and hernia surgery, or hernia surgery after delivery. Therefore, Such considerations are essential in making informed clinical decisions and ensuring consistent patient management. Your presentation of the treatment guidelines also has numerous limitations.

## Disclosure

The author reports no conflicts of interest in this communication.

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