

## Silver sulfadiazine as the topical treatment for giant omphalocele: a case report



Ni Luh Putu Herli Mastuti<sup>1</sup>, Risa Etika<sup>1</sup>, Martono Tri Utomo<sup>1\*</sup>,  
Dina Angelika<sup>1</sup>, Kartika Darma Handayani<sup>1</sup>

### ABSTRACT

**Background:** Major omphalocele is defined as an abdominal defect greater than 5 cm with the presence of the liver and most parts of the bowel in the sac. The management remains challenging for pediatricians and surgeons with remarkably high mortality. Reports about topical agents/dressing used as escharotic therapy, such as silver sulfadiazine, povidone-iodine, topical antibiotics, or honey, have been published with different results. Continued application of a thick layer of silver sulfadiazine on the omphalocele surface is needed to promote eschar formation and neo-epithelialization.

**Case description:** A term, 2700-gram newborn male infant presented with abdominal defect and herniated abdominal contents covered by a membranous sac containing liver that is widely known as omphalocele. A thick layer of silver sulfadiazine was applied repeatedly onto the omphalocele sac to promote epithelialization and successfully reduce the sac diameter. The patient showed a positive outcome as it reduced the omphalocele size after nine days. The patient family has been educated about the continued application of silver sulfadiazine. Although non-operative delayed treatment has lower mortality rates and better clinical outcomes, the patients should wait for the reconstruction surgery for a long time.

**Conclusion:** Conservative treatment with silver sulfadiazine is safe and shows satisfying results to the giant omphalocele.

**Keywords:** Conservative Treatment, Omphalocele, Sulphadiazine.

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<sup>1</sup>Department of Child Health, Faculty of Medicine, Universitas Airlangga, Dr. Soetomo General Academic Hospital, Surabaya, Indonesia;

\*Corresponding author:

Martono Tri Utomo;  
Department of Child Health, Faculty of Medicine, Universitas Airlangga, Dr. Soetomo General Academic Hospital, Surabaya, Indonesia;  
mrmartono73@gmail.com

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### INTRODUCTION

Omphalocele is one of the major congenital abdominal wall defects leaving abdominal content eviscerated into the umbilical cord through the umbilical ring and exposed to the environment. The disease's incidence is 1/4000-7000 live births and affects 10-30% of cases of chromosomal anomalies with a high mortality rate. Only about 60% of children with such malformations survive until the end of their first years of age.<sup>1</sup> The well-established risk factors contributing to the prognosis include the defect's size, antenatal rupture of the sac, low birth weight, gestational age, associated anomalies, and prenatal respiratory distress. Omphalocele occurs due to failure of the four embryonic folds to meet in the midline and form an umbilical ring before the 10<sup>th</sup> week of gestation, resulting in ventral abdominal wall defect of varying degrees.<sup>2</sup>

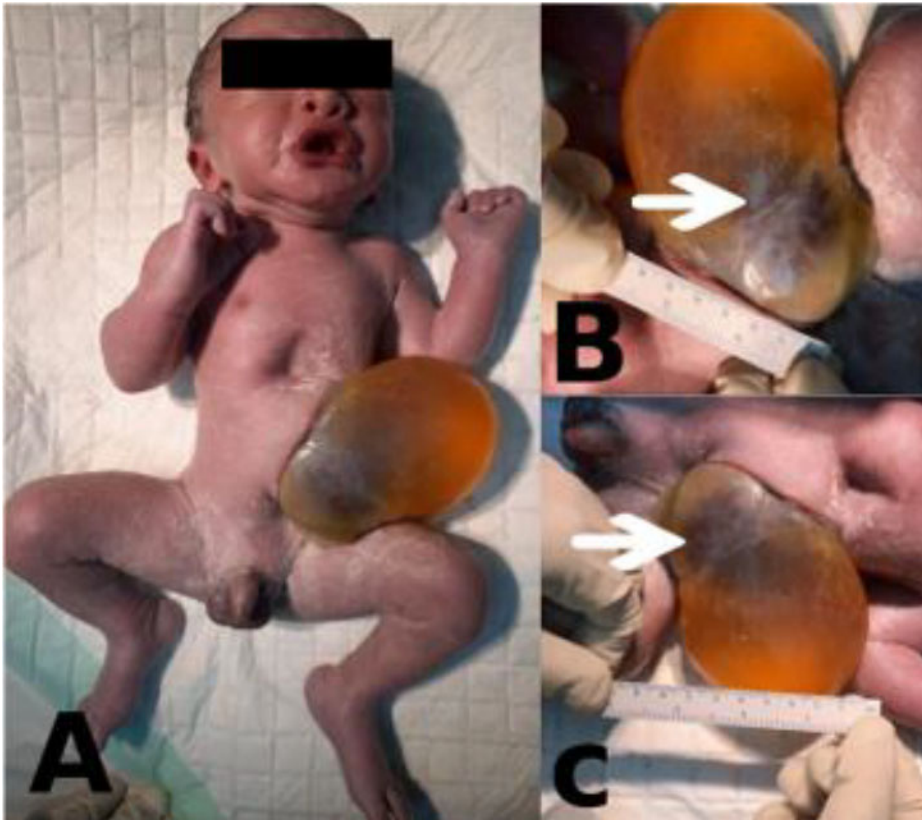
The optimal route of delivery is still controversially discussed. The clinicians should consider the defect size, herniated organs in the sac, the integrity of the sac

and any other associated abnormalities.<sup>3</sup> Associated anomalies include congenital heart disease, chromosomal, renal genitourinary fascial, skeletal, and gastrointestinal anomalies.<sup>4</sup> The omphalocele size varied, ranging from 4 to 12 cm. Omphalocele major has a defect of more than 5 cm diameter, while the minor has a less than 5 cm diameter. The size of the omphalocele and abdominal cavity is related to surgical planning. Omphalocele treatment aims to close the abdominal wall defect after reducing the abdominal content and stabilization with supportive care. In general, treatment strategies can be classified as immediate (primary), staged repair with delayed primary closure, and delayed repair (paint and wait) with secondary closure of abdominal wall hernia. In recent years, the most used method is non-operative delayed closure, which involves the maintenance of the sac with topical medication and regular dressing, providing epithelialization and subsequent closure of the ventral hernia with delayed surgery. This treatment is often chosen for infants with giant

omphalocele and/or a high degree of abdominal-visceral disproportion.<sup>5</sup> Here, we present one omphalocele case that successful decrease the size defect with non-operative treatment using Silver sulfadiazine topical. Informed consent was obtained from parental consent.

### CASE DESCRIPTION

A 2700 grams male newborn was born by cesarean section from a 25-year-old mother at 38-39 weeks of gestation. General activities of the baby were normal with spontaneous crying and 7-8 of APGAR score and normal hemodynamic status. The patient has an initial heart rate of 126 beats per minute, a respiratory rate of 42 breaths per minute, and a temperature of 36.7° Celcius. A herniated-out bowel on an 8 cm sac protruding from his umbilical cord with liver apparent on the sac (**Figure 1A-C**). Complete blood count reported hemoglobin 11.8 mg/dl, leukocytes 16,820, hematocrit 36.2%, platelets 294,000. The abdominal x-ray revealed a cavity with tissue intensity on



**Figure 1.** Omphalocele size of  $\pm 5$  cm and  $\pm 8$  cm with a liver appearance on the sac (White arrow).



**Figure 2.** Babygram showed a cavity with tissue intensity within the anterior abdominal wall.

the anterior abdominal wall, as seen in [Figure 2](#).

On the 7<sup>th</sup> day of life, the echocardiography result was Patent Ductus Arteriosus (PDA), 0,27 mm diameter, right to left shunt. Ibuprofen drop was given for PDA management ([Figure 3](#)). The management aims to maintain the sac and avoid membrane rupture. A thick layer

of silver sulfadiazine was applied topically onto the omphalocele sac, reapplying as needed. The layer was applied repeatedly if the prior layer was completely absorbed. The surrounding omphalocele is wrapped with sterile gauze. After nine days, the patient was discharged from the hospital with a decrease in omphalocele diameter (4x6 cm), as shown in [Figure 4](#).

## DISCUSSION

Maldevelopment of the midline abdominal wall in early embryonic development (ED) is the etiology of omphalocele. The normal development of the primitive intestine allows, at six weeks of embryonic development, a normal physiologic herniation of the primitive mid-gut after undergoing the 90 degrees counterclockwise rotation back into the umbilical cord.<sup>6,7</sup> omphalocele occurs caused by two events: (i) an incomplete embryonic lateral plicature between 4 and 8 weeks of embryonic development. (ii) an incomplete migration and differentiation of mesodermal somites into myotomes originate from cutaneous tissue and

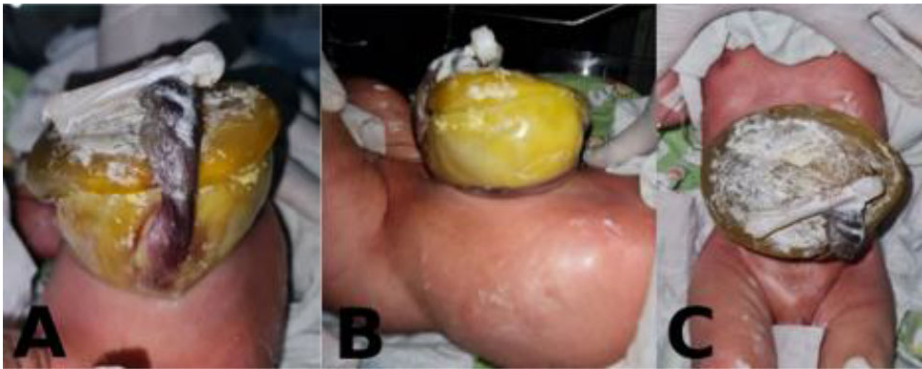
abdominal wall muscle.<sup>7,8</sup> The incomplete mechanism causes midline defects of the abdomen where the abdominal organs, especially the bowel segments, are herniated out.

The optimal route of delivery is still controversially discussed. Factors to consider when determining the delivery route are the defect size, involved organs, the integrity of the sac and other associated abnormalities.<sup>3</sup> Prenatal screening is essential for the early detection of omphalocele and related anomalies and increases the frequency of elective pregnancy termination.<sup>9</sup> In this case, the patient was delivered by cesarean section due to omphalocele. Children's Hospital of Orange County (CHOC Children's) states vaginal delivery may be possible for small omphaloceles. Cesarean section was recommended for giant omphalocele to prevent omphalocele membrane rupture and enclosed organ trauma (specifically liver). Full-term delivery was encouraged, but the latter depends on fetal and/or maternal indications.<sup>10,11</sup>

Omphaloceles are classified as 'minor' or 'major' depending on the sac's contents and the defect's diameter. Most 'minor' cases have diameters less than 5 cm and contain small portion loops of the small bowel. There are usually no other major congenital anomalies. The description of omphalocele major considers a defect  $>5$  cm, the presence of a liver and a large volume of bowel in the sac.<sup>12,13</sup> Primary surgical closure may be carried out in small omphaloceles cases; on the contrary, the surgery should not be attempted on giant omphaloceles.<sup>11,14</sup> Several life-threatening complications include sudden elevation of intraabdominal pressure, which leads to respiratory failure, hemodynamic instability, inferior vena cava compression or distortion, acute renal failure, bowel obstruction, and bowel ischemia.<sup>14</sup> Fifty-eighty percent may have other congenital anomalies, which may increase newborn mortality, and the defect seems more prevalent in the male gender. These additional anomalies were varied, and all organs can be affected depending on the etiology, including neural tube defect, cleft palate, single umbilical artery, and amniotic fluid anomaly (oligoamnios or polyhydramnios). Cardiovascular defects



**Figure 3.** Echocardiography at seven days old showed Patent Ductus Arteriosus.



**Figure 4A-C.** Omphalocele care using sulphadiazine cream.

were the most commonly associated disorder in 40-60% of cases.<sup>7,10,15</sup> Our patient is a male with a major omphalocele with an 8 cm sac diameter and liver involvement. He also suffers from 0,27 mm Patent Ductus Arteriosus (PDA) right to left shunt.

The omphalocele should be stabilized during resuscitation and transport to avoid bleeding from the liver or congestion of the liver veins. During primary stabilization, be aware of the hypothermia, establish vascular access to achieve and maintain the euvolemia but avoid the umbilical vessels, and avoid mask ventilation in case of respiratory distress; early intubation may be indicated. Children's Hospital of Orange country recommends maintaining sac integrity by using 1) utilized sterile gloves when handling, 2) placing the neonate in a bowel bag lined with a small amount of warm sterile saline solution, and 3) positioning the neonate sideline

while supporting the omphalocele with blanket rolls to optimized perfusion and prevent compression of blood vessels.<sup>3,11</sup>

Omphalocele treatment aims to reduce the abdominal content followed by the closure of the abdominal wall defect.<sup>16</sup> The current primary treatment is divided into two basic categories: 1) non-operative delayed closure (involves the maintenance of the sac with topical medications and regular dressings, providing epithelization, also known as Paint and Wait for methods), 2) removal of the graft and primary closure after ensuring epithelization with the graft in the early period. The topical medications used in non-operative delayed closure are antimicrobial or escharotic agents. Conservative management of omphaloceles allows wound contraction and epithelization by eschar formation, leaving a ventral hernia that may be repaired at the following time at adequate age to avoid the risk of major

neonatal surgery. This approach has been reported to have better outcomes than early surgery in terms of shorter hospital stay, early enteral feeding and reduced mortality due to fatal complications (e.g., abdominal compartment syndrome, wound dehiscence, intestinal obstruction, and perforation.<sup>13,17</sup> The choice of an escharotic agent depends on the local availability, cost, ease of application and low risk of adverse effects.<sup>17</sup>

Some topical agents that have been well-established to have positive outcomes include silver sulfadiazine, povidone-iodine, *A. nilotica* paste, topical antibiotics, or honey.<sup>5,18</sup> A combination of povidone-iodine and antibiotic powder (polymyxin B sulfate, bacitracin zinc, and neomycin) shows faster escharization in infants with GO than povidone-iodine alone.<sup>16,19</sup> The study of giant omphalocele management using povidone-iodine shows complete epithelialization of the sac at  $10.0 \pm 2.5$  weeks, and the surgical intervention was done at 4-9 months of age.<sup>17</sup> The application of *A. nilotica* paste twice a day shows epithelialization in the mean period of  $7.83 \pm 4.82$  weeks. The mechanism of action in *A. nilotica* paste is inducing coagulation of the protein contents of the sac, changing the sac consistency into a rigid structure, which prevents fluid loss and acts as a barrier against microorganisms. The epithelialization by applying Manuka honey was achieved in 63 days (48-119).<sup>20,21</sup>

Silver sulfadiazine is low-cost and provides a moist wound healing environment, promoting early granulation with good broad-spectrum antibiotics properties. It also had antifungal coverage. It is widely used for burn treatment and has shown a good clinical outcome.<sup>10,22</sup> A thick layer (three to five layers) of silver sulfadiazine dressing provides a moist wound environment that increases angiogenesis and prevents wound degradation.<sup>23,24</sup> The application of silver sulfadiazine cream in our center is done by repeated application of silver sulfadiazine if the prior layer has been absorbed using sterile gloves and sterile gauze. The application may be continued at home with the same routine with a clean technique.<sup>10</sup> The NOC score assessment on severe burn patients shows

a NOC of 35.20 in the mean duration of wound healing of 25.4 days, indicating a significant process in wound healing (85.7% granulation tissue growth and 75-100% epithelialization).<sup>25</sup> The length of hospitalization and time to enteral feeding was also shorter (20 days and four days, respectively).<sup>14</sup> Ein and Langer have explained the omphalocele management technique using silver sulphadiazine. About 20 g of silver sulphadiazine cream was applied onto the omphalocele sac and then wrapped with sterile gauze using a clean method once a day. The granulation tissue usually appeared by the end of the third or fourth week, then slowly replaced by epithelialized scar over 4 to 12 months, leaving a large ventral hernia. If the neck of the sac was narrow (<5 cm) and blocked the viscera in the sac from reverting into the peritoneal cavity, the sac was removed, and the narrow neck was converted to a broad base (for later reduction and closure). A Silon pouch or Op-site was applied. The subsequent surgery is a repair of the ventral hernia, which started approximately one year old.<sup>10</sup>

It has been reported that silver sulfadiazine can disrupt the granulation tissue. Other reported risks related to silver toxicity include seizures, peripheral neuropathy, ocular pathology, nephrotic syndrome, elevated liver enzymes, argyria and leucopenia. However, there is no clear report regarding the duration of exposure, dosage, and silver serum level leading to this complication.<sup>26,27</sup>

Our patient showed a positive outcome as it reduced the omphalocele size after nine days. The patient family has been educated about the continued application of silver sulfadiazine. Although non-operative delayed treatment has lower mortality rates and better clinical outcomes, the patients should wait for the reconstruction surgery for a long time. Wound care during this waiting period is challenging for the family and the clinician.<sup>16</sup> The family should be trained to apply silver sulfadiazine and suggested for weekly monitoring to primary health care. The patient should be taken to the emergency room immediately if intestinal malrotation signs occur (e.g., recurrent vomiting or abdominal distention).<sup>17</sup> The limitation is that this case only presents

one case. In the future, it is necessary to present more cases with another treatment to increase the success of non-operative treatment in omphalocele cases.

## CONCLUSION

Conservative treatment with silver sulfadiazine is safe and showed satisfying results by reducing the omphalocele size.

## CONFLICT OF INTEREST

The authors declare that there is no competing interest regarding the manuscript.

## ETHICAL CONSIDERATION

The patient's birth mother gives consent for information about the patient (photograph and article) to appear in a journal article.

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## AUTHOR CONTRIBUTION

NLPHM was involved in writing the manuscript. RE, MTU, DA, and KDH supervised and revised the manuscript. All authors prepare the manuscript and agree for this final version of the manuscript to be submitted to this journal.

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