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(Citation)
Journal of Obstetrics and Gynaecology Research,47(9):3370-3373

(Issue Date)
2021-09

(Resource Type)
journal article

(Version)
Accepted Manuscript

Abdominal compartment syndrome in a monochorionic-triamniotic triplet pregnancy complicated by feto-fetal transfusion syndrome

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Running title: Abdominal compartment syndrome

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Abstract

A 40-year-old primigravida woman with a monochorionic-triamniotic triplet pregnancy was hospitalized due to threatened abortion at 16 gestational weeks. Polyhydramnios in two fetuses and oligohydramnios in the third supported a diagnosis of feto-fetal transfusion syndrome at 23 weeks and 3 days of gestation. Severe dyspnea and liver dysfunction required intensive care unit admission and mechanical ventilation support, and abdominal compartment syndrome caused by polyhydramnios was clinically diagnosed. When her general condition was not improved regardless of intensive care, the patient delivered the three fetuses by caesarean section at 23 weeks and 5 days gestation. Abdominal decompression was achieved with delivery, and the patient was discharged 13 days after operation without morbidity. This is the first case report of abdominal compartment syndrome caused by feto-fetal transfusion syndrome in a monochorionic-triamniotic triplet pregnancy resulting in extremely preterm birth.

Key words: Abdominal compartment syndrome, amnioreduction, feto-fetal transfusion syndrome, polyhydramnios, triplet pregnancy

Introduction

Abdominal compartment syndrome (ACS) is a life-threatening condition where increasing intra-abdominal pressure (IAP) leads to multiple organ failure and respiratory disorder. ACS can be caused by severe peritonitis,[1] ileus,[2] or significant hemorrhage due to abdominal trauma or surgery.[3] Several ACS cases have been associated with pregnancy. One study reported of ACS cases due to massive intra-abdominal bleeding after caesarean section,[4] while other cases were due to massive ascites caused by ovarian hyperstimulation syndrome.[5] However, no reports of pregnant women with ACS following polyhydramnios have been published. We hereby report the case of a pregnant woman who developed ACS caused by polyhydramnios resulting from feto-fetal transfusion syndrome (FFTS) in a monochorionic-triamniotic (MT) triplet pregnancy.

Case report

This case report followed the principles of the Declaration of Helsinki, and written informed consent was obtained from the patient. A 40-year-old primigravida woman, who conceived by intracytoplasmic sperm injection, was referred to the Kobe University Hospital from another hospital at 16 weeks and 1 day of gestation for a MT triplet pregnancy and idiopathic chronic thrombocytopenia (platelet count 69,000 /μL). A transvaginal ultrasound at the first visit revealed shortening of the cervical length (12 mm), and the patient (height; 163 cm, body weight; 69.5 kg, body mass index; 26.2 kg/m^2) was admitted to the university hospital for threatened abortion caused by cervical incompetency. Transvaginal ultrasound examination at 17 weeks and 5 days of gestation showed a shortening of cervical length (11 mm)

and a placenta overlapping the internal os of the cervix (Figure 1). We attempted cervical cerclage at 17 weeks and 5 days of gestation, but it was unsuccessful due to severe bleeding of the uterine cervix. After operation, continuous intravenous infusion of ritodrine hydrochloride was administered, and an exacerbation of threatened abortion was not observed until 21 gestational weeks (GW). However, a discordance in amniotic fluid volume among the three fetuses was observed (the heights of the maximal vertical pockets [MVP] were 7.4 cm, 8.5 cm, and 3.2 cm) at 23 weeks and 1 day of gestation, and the patient was suspected to be in pre-onset FFTS (Figure 2).

Threatened premature delivery was exacerbated by polyhydramnios in two fetuses. Therefore, continuous intravenous infusion of magnesium sulfate was added, and betamethasone was administered for fetal lung maturity at 23 weeks and 1 day of gestation. At 23 weeks and 3 days of gestation, fetal ultrasound examination revealed polyhydramnios in two fetuses (the heights of the MVP were 11.8 cm and 14.7 cm) and oligohydramnios in the third fetus (the height of MVP was 0.3 cm), and the patient was diagnosed with FFTS. She did not receive fetoscopic laser photocoagulation for FFTS because she had severe threatened premature labor with 2 cm cervical dilation and thrombocytopenia (platelet count 61,000 /μL). At 23 weeks and 3 days of gestation, because she complained of dyspnea caused by polyhydramnios, we tried to remove amniotic fluid from each sac of the two fetuses with polyhydramnios as much as possible. However, she had painful and frequent uterine contractions just after needle puncture, we could only remove 700 mL of amniotic fluid from the sac of one fetus with the largest MVP.

She gained temporary dyspnea improvement, but severe dyspnea relapsed

only 6 hours after the amnioreduction. Therefore, at 23 weeks and 4 days of gestation, the patient required intensive care unit admission and non-invasive positive pressure ventilation due to severe respiratory distress. Chest X-ray showed diaphragmatic eventration, reduction in bilateral lung permeability, and the absence of pleural effusion (Figure 3). In addition, rapid elevation in liver enzyme levels and macroscopic hematuria were observed (Figure 2), but she did not have hypertension, cardiac or renal dysfunction. Because she had severe abdominal distension and respiratory distress together with the absence of pleural effusion, she was clinically diagnosed with ACS caused by polyhydramnios resulting from FFTS. Because the patient's respiratory distress was not improved regardless of intensive care and also because there was a risk that repeated amnioreduction would cause premature delivery in this case, her pregnancy was terminated by caesarean section to treat ACS by removing the amniotic fluid and fetuses, thereby reducing IAP, at 23 weeks and 5 days of gestation. The caesarean section was performed under general anesthesia. The induction of anesthesia was preceded by amnioreduction, in which 800 mL amniotic fluid was removed, to facilitate respiration with mechanical ventilation. She delivered three female newborns weighing 446 g, 490 g, and 462 g with Apgar scores of 1, 1, and 2 at 1 min and 8, 6, and 6 at 5 min, respectively. The total amount of amniotic fluid of all three infants was estimated to be over 3,500 mL.

The patient's respiratory and liver functions dramatically improved after delivery. She was successfully extubated 2 hours after operation, and oxygen inhalation therapy was stopped 4 days after operation. She was discharged without complications at 13 days after operation. The infants were discharged at 173, 194,

and 225 days after birth, respectively, and have survived without morbidity at present (9 months after birth).

Discussion

ACS is a life-threatening condition where the resulting intra-abdominal hypertension (IAH) leads to multiple organ failure and respiratory disorder. In previous reports, ACS associated with pregnancy were mainly caused by massive intra-abdominal bleeding after caesarean section in patients with disseminated intravascular coagulation followed by preeclampsia or HELLP (hemolysis, elevated liver enzymes, and low platelets) syndrome.[4] To the best of our knowledge, this is the first case report of a pregnant woman who developed ACS caused by polyhydramnios resulting from FFTS in a MT triplet pregnancy.

According to the World Society of the Abdominal Compartment Syndrome (WSACS), ACS is defined as a condition where IAP is sustained at more than 20 mmHg and is further characterized by the presence of newly appearing organ dysfunction or failure.[6] It is thought that the physical compression of the liver and kidney caused by IAH leads to liver and renal dysfunction, whereas diaphragmatic eventration caused by IAH results in cardiac and pulmonary dysfunction.[7] The gold-standard treatment for ACS is abdominal decompression, including opening the anterior abdominal wall with subsequent temporary abdominal closure.[8] IAP is generally measured indirectly by measuring bladder pressure.[9] Bladder pressure was not measured in the present case. However, the rapid worsening of respiratory distress together with abdominal distension, liver dysfunction, and macroscopic hematuria observed following an abrupt increase in amniotic fluid were considered

significant indications of ACS. Respiratory and liver functions and macroscopic hematuria were dramatically improved after caesarean section, which achieved abdominal decompression by removing the three fetuses and a large amount of amniotic fluid. Thus, early abdominal decompression is important in improving the prognosis in patients with ACS even without measuring the bladder pressure. In addition, repeated amnioreduction from the sacs of fetuses with polyhydramnios might enable to prolong pregnancies, but in this case exacerbation of threatened premature delivery induced by needle puncture and early relapse of respiratory distress after the initial amnioreduction made it impossible to perform repeated amnioreduction.

Symptoms of dyspnea together with pulmonary edema and liver and renal dysfunction during pregnancy are often observed in patients with hypertensive disorders in pregnancy (HDP). In contrast, dyspnea in patients with ACS were caused by the restriction of lung expansion due to diaphragmatic eventration by IAH. Therefore, diaphragmatic eventration and the absence of pulmonary effusion in chest X-ray or computed tomography together with the absence of hypertension are useful indications for distinguishing dyspnea associated with ACS from that with HDP.

The earlier newborns are delivered, the poorer their outcomes become. Therefore, clinicians hesitate to terminate pregnancies at an extremely early gestational age. However, ACS is so severe that delayed treatments can lead to severe organ failure and even to maternal mortality. Hence, clinicians should consider early termination of pregnancies to avoid adverse outcomes associated with ACS caused by polyhydramnios if amnioreduction and intensive care,

including respiratory support, do not improve ACS.

In addition, we planned to perform amnioreduction before the induction of general anesthesia to facilitate respiration by mechanical ventilation and a safe caesarean section. Well-planned caesarean deliveries are essential in treating pregnant women with ACS caused by polyhydramnios.

This first report on FFTS in a MT triplet pregnancy leading to ACS provides information useful in future cases for clinical practitioners in perinatal medicine.

Acknowledgments

We acknowledge and thank all the members of the multidisciplinary teams at Kobe University Hospital and in particular: Dr. Kazumichi Fujioka (Department of Pediatrics), Dr. Kana Ozaki, Dr. Yuki Sasagawa, Dr. Akiko Uchida, Dr. Yutoku Shi, Dr. Tokuro Shirakawa, and Dr. Masashi Deguchi (Department Obstetrics and Gynecology).

Disclosure

The authors state that they have no conflict of interest.

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Figure legends

Figure 1. Transvaginal ultrasound image of the patient at 17 weeks and 5 days of gestation.

Figure 2. Clinical course of the patient. In the upper chart, (●), (▲), and (■) indicate the heights of the maximal vertical pockets (MVP) of the three fetuses (cm). In the lower chart, (○) and (△) indicate the serum levels of alanine aminotransferase (ALT, U/L) and aspartate aminotransferase (AST, U/L), respectively.

Abbreviations: GW, gestational week; POW, postoperative week.

Figure 3. Chest X-ray of the patient at 23 weeks and 4 days of gestation.

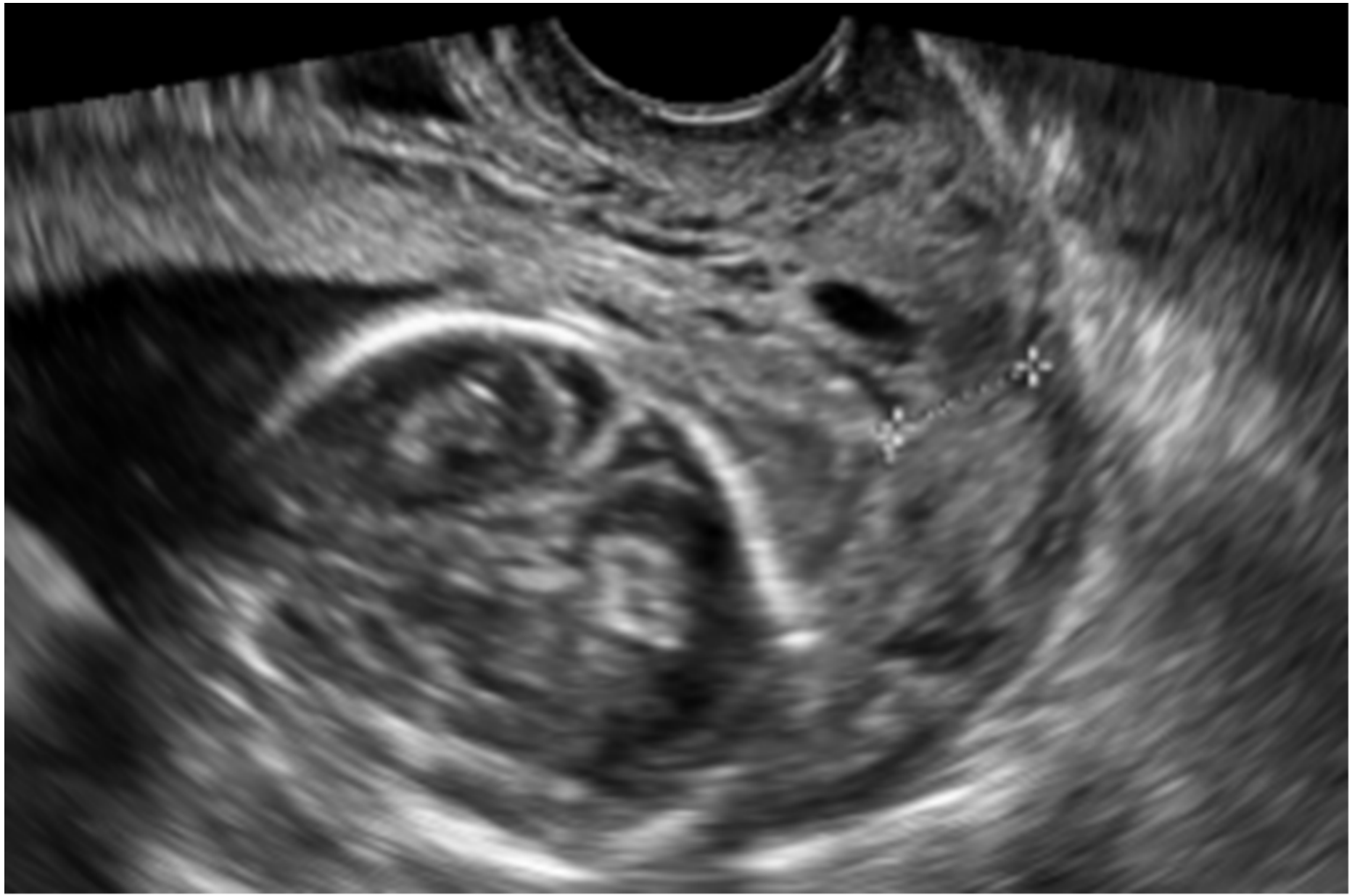

Figure 1

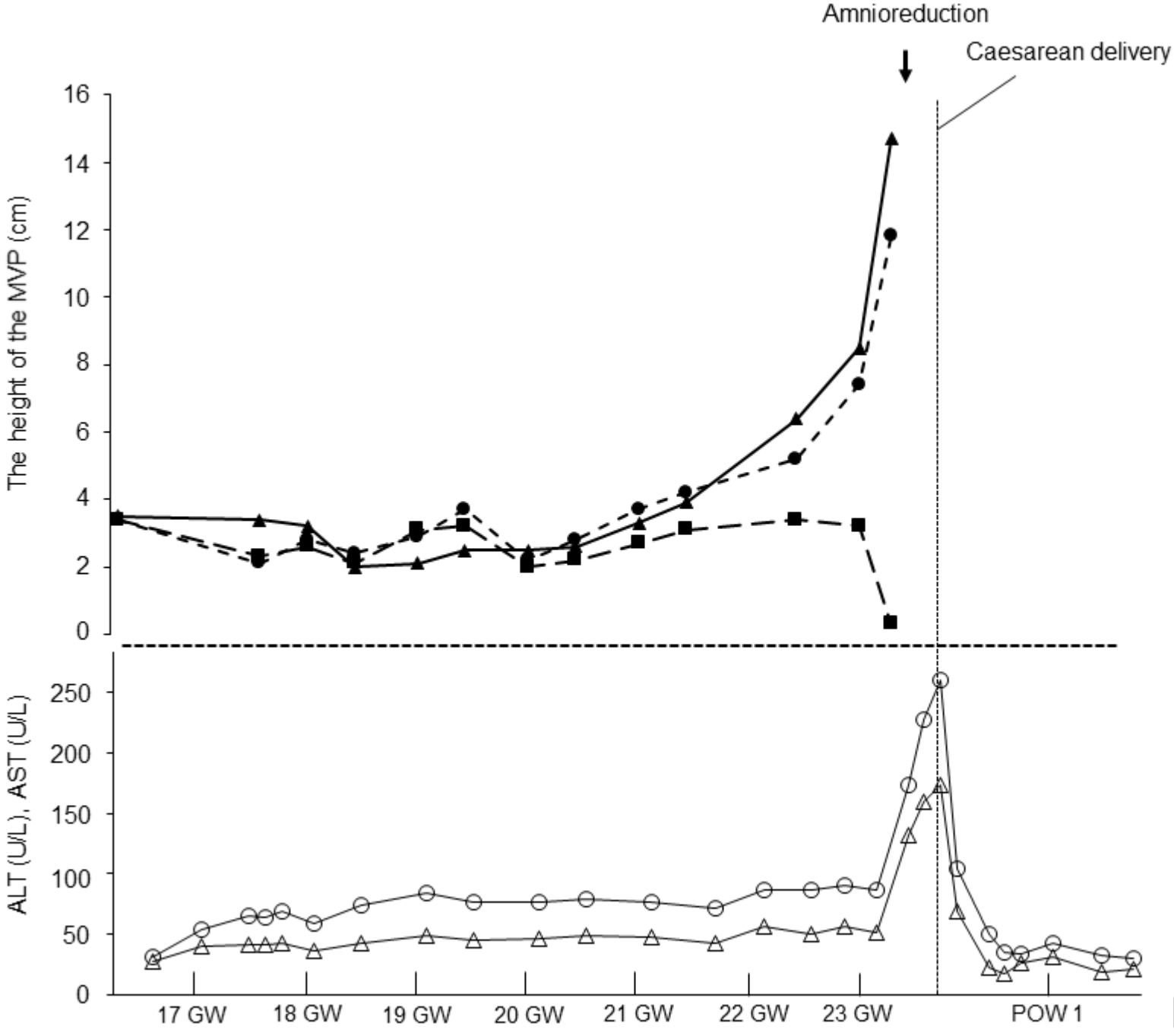
Amnioreduction
Caesarean delivery
The height of the MVP (cm)
16
14
12
10
8
6
4
2
0
ALT (U/L), AST (U/L)
250
200
150
100
50
0
17 GW
18 GW
19 GW
20 GW
21 GW
22 GW
23 GW
POW 1

Figure 2

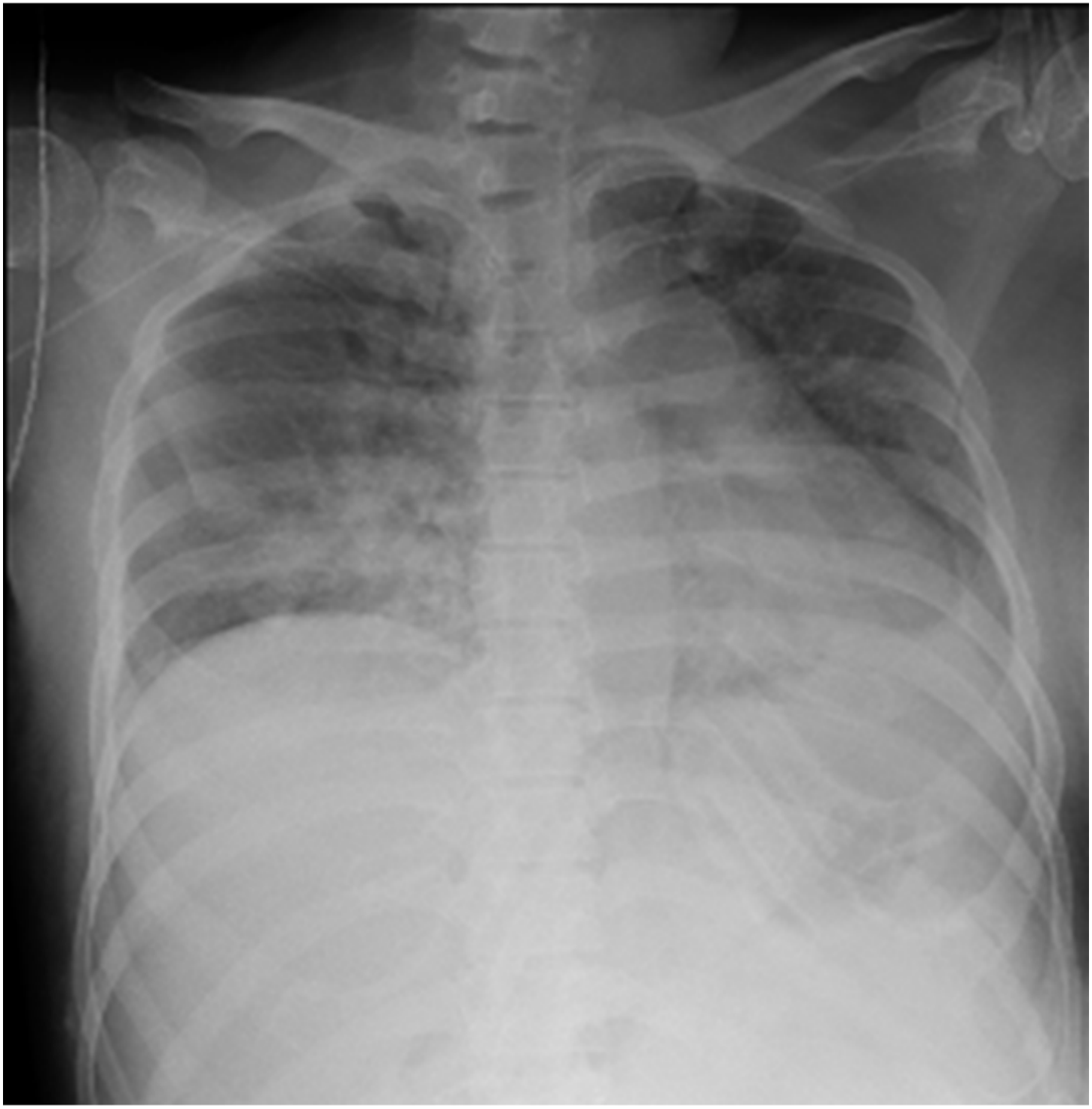

Figure 3