Vol. 6, No. 1, Jan-Jun 2022

eISSN : 2579-8324 pISSN : 2579-8323

ANDALAS OBSTETRICS AND GYNECOLOGY JOURNAL

Address for Correspondence:
Editorial Room Andalas Obstetrics and Gynecology Journal, 3rd floor of KSM of Obstetrics and Gynecology, RSUP DR. M. Djamil Padang, Jl. Perintis Kemerdekaan Padang, Sumatera Barat 25127

Website:
http://jurnalobgin.fk.unand.ac.id/index.php/JOE

CASE REPORT

Amnioinfusions to Treat Early Onset Anhydramnios Caused by Bilateral Renal Agenesis : Potter's Syndrome

Yusrawati[1], Rizka Fadhillah Yusra[2]

Affiliations: *Sub Division of Fetomaternal Medicine, Obstetrics and Gynecology Department, Faculty of Medicine, Andalas University, Dr. M. Djamil Central General Hospital Padang; 2. Obstetrics and Gynecology, Faculty of Medicine, Andalas University, Dr. M. Djamil Central General Hospital Padang*

Correspondence: *Rizka fadhillah Yusra,* email: *rizkayusra@yahoo.com Hp:081363478125*

Abstract

Introduction*: Anhydramnios is a very small amount of amniotic fluid where the MVP measurement is 2cm by ultrasound. The most common cause of anhydramnios that persists into the second trimester of pregnancy is bilateral renal agenesis. Bilateral renal agenesis is closely related to Potter's Syndrome. Potter's syndrome is a picture of reduced amniotic fluid regardless of the cause. The most common cause of newborn death in cases of anhydramnios is pulmonary hypoplasia. Amnioinfusion is an action of adding fluid into the amniotic cavity which is expected to reduce uterine pressure due to anhydramnios and maintain alveolar distension to increase fetal lung growth.*

Objective: *The aim of this case report is to share amnioinfusion on anhydramnios.*

Case Report: *A 26-year-old woman G3P1A1H1 gravid 27-28 weeks was referred to the fetomaternal polyclinic of RSUP M. Djamil Padang with suspicion of anhydramnios caused by bilateral renal agenesis. Physical and obstetric examinations were found to be within normal limits. On ultrasound examination, there was no amniotic fluid, so MVP was difficult to assess, no fetal kidney and bladder were seen, so it is suspected that this is a bilateral renal agenesis disorder and leads to Potter's Syndrome. The patient was subjected to amnioinfusion to prevent contractures and pulmonary hypoplasia in the fetus. From the first amnioinfusion, the MVP increased to 2.99 cm. Monitoring and amnioinfusion are carried out periodically until the fetus is viable to be born*

Conclusion: *Amnioinfusion in bilateral renal agenesis is useful for assisting diagnosis and as a preventive therapy for pulmonary contractures and hypoplasia in the fetus as well as increasing life expectancy when the fetus is born.*

Keywords**:** *Bilateral Renal Agenesis, Anhydramnios, Amnioinfusion, Potter's Syndrome*

INTRODUCTION

Anhydramnios is a very small amount of amniotic fluid where the MVP measurement is 2cm.[1,2] Anhydramnios that persists in the 2nd trimester before the stage of lung organ formation will cause pulmonary hypoplasia.[3] This situation will give a high mortality rate that is more than 80% and babies who survive will experience chronic lung disorders. In addition, anhydramnios will increase the risk of umbilical cord compression, joint contractures, skeletal system disorders, stunted growth and fetal death. The management of this case is still a big challenge for obstetricians and pediatricians.[1,3,4,12]

Received : January 10th, 2022
Accepted : January 22th, 2022
Correspondence: Rizka fadhillah Yusra, email: *rizkayusra@yahoo.com*

Vol. 6, No. 1, Jan-Jun 2022
ANDALAS OBSTETRICS AND GYNECOLOGY JOURNAL
Address for Correspondence:
Editorial Room Andalas Obstetrics and Gynecology Journal, 3rd floor of KSM of Obstetrics and Gynecology, RSUP DR. M. Djamil Padang, Jl. Perintis Kemerdekaan Padang, Sumatera Barat 25127
eISSN : 2579-8324 pISSN : 2579-8323
Website:
http://jurnalobgin.fk.unand.ac.id/index.php/JOE

The condition of anhydramnios and the inability to visualize the kidneys and bladder that are difficult to identify after several examinations are characteristic of bilateral renal agenesis. This condition occurs in 0.1 to 0.3 per 1000 births. The condition of prolonged anhydramnios will cause uterine compression on the fetus, causing deformity of the fetal limbs, contractures of the head, face and pulmonary hypoplasia. This condition is known as Potter's Syndrome or Potter Sequence.[5,13]

Amnioinfusion is a prenatal action in the form of adding fluid into the amniotic cavity which is expected to restore intra uterine physiological conditions. Amnioinfusion is both diagnostic and therapeutic. Amnioinfusion can be performed antepartum or intrapartum. Diagnostic amnioinfusion is performed antepartum with the aim of improving sonographic assessment in the interest of prenatal diagnosis. While therapeutic amnioinfusion aims to improve the quality of life and biophysics of the fetus.[6,14]

Based on the explanation above, this case report aims to evaluate the current management efforts for cases of anhydramnios with bilateral renal agenesis; in the case of a suspected Potter's Syndrome

CASE REPORT

A 26-year-old multiparous woman G3P1A1H1 gravid 27-28 weeks was referred to the fetomaternal polyclinic M.Djamil Hospital Padang with suspicion of anhydramnios caused by bilateral renal agenesis. The patient did not have certain complaints such as bleeding or discharge from the vagina, abdominal pain, shortness of breath, or other complaints. Fetal movements have been felt. The patient admitted that he did not take any drugs other than pregnancy vitamins, did not smoke or consume alcohol and herbs. The patient had no history of using any contraception. The patient had no history of previous illness, allergies or previous surgery. Regular control patients went to the midwife 5 times, from 2,3,4,5 and 6 months of gestation, 4x controls to Sp.OG and 3,4,5 and 6 gestational ages. At 4 months of gestation it was known that the amniotic fluid was slightly , at 6 months of gestation the patient was referred to a fetomaternal consultant. This is the 3rd pregnancy, the first child was born at term spontaneously at the midwife, and the second pregnancy miscarried at 2 months of gestation.

On physical examination, he was aware of compost mentis, with blood pressure 110/70 mmHg, pulse rate 84 times/minute, respiratory rate 20 breaths/minute, temperature 36.8o C, weight 53 kg and height 157 cm. General status within normal limits. In obstetrical status, the uterine fundus was found to be at the level of the navel, with a fetal heart rate of 154 beats/minute.

On ultrasound examination, it was found that a single live fetus was located right dorso-inferior latitude, fetal heartbeat (+), fetal movement was limited, with an estimated gestational age of 27 weeks, estimated fetal weight was 1021 grams, SDP was difficult to assess, renal and bladder images were not found. From the results of the ultrasound examination, it was

Received : January 10th, 2022
Accepted : January 22th, 2022
Correspondence: Rizka fadhillah Yusra, email: rizkayusra@yahoo.com

Vol. 6, No. 1, Jan-Jun 2022
ANDALAS OBSTETRICS AND GYNECOLOGY JOURNAL
Address for Correspondence:
Editorial Room Andalas Obstetrics and Gynecology Journal, 3rd floor of KSM of Obstetrics and Gynecology, RSUP DR. M. Djamil Padang, Jl. Perintis Kemerdekaan Padang, Sumatera Barat 25127
eISSN : 2579-8324 pISSN : 2579-8323
Website:
http://jurnalobgin.fk.unand.ac.id/index.php/JOE

suspected that there was bilateral renal agenesis in the fetus leading to Potter's syndrome (Figure 1).

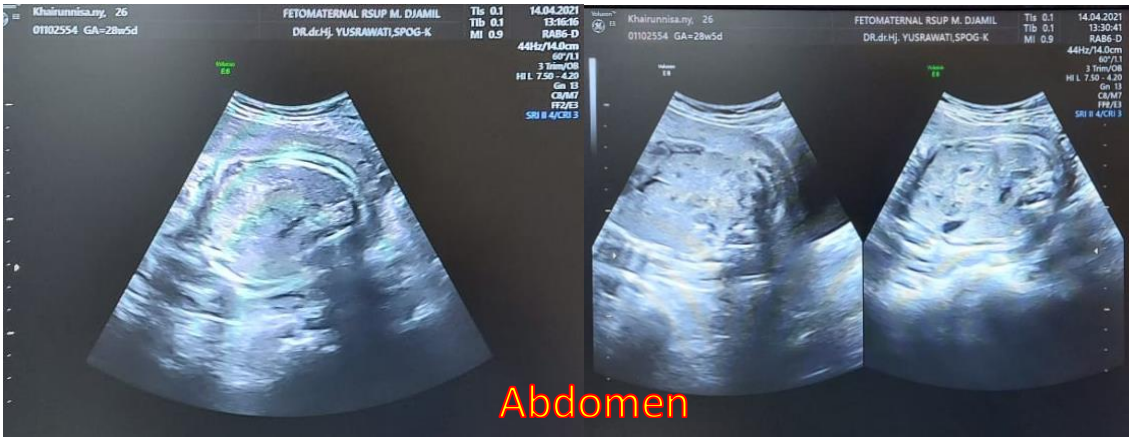

Figure 1: Ultrasound before amnioinfusion, SDP is difficult to assess, no kidney and bladder images are found (Black arrow: where the kidney should be, Red arrow: where the bladder should be)

Nine days later, a transabdominal amnioinfusion was performed on the patient. Prior to the amnioinfusion, the mother and husband were asked to give consent and were given an explanation regarding the procedure, risks to the mother, fetus and uncertainty about the success of the amnioinfusion. Medical risks associated with amnioinfusion include the risk of injury to the fetus, intrauterine fetal death, PROM, preterm labor, placental abruption, chorioamnionitis, and uterine rupture.

Amnioinfusion was carried out at the fetomaternal polyclinic of M. Djamil Hospital Padang, normal saline 0.9% NaCl was inserted using a spinocaine fr 23 needle. Amnioinfusion was performed using transabdominal ultrasound guidance, then the needle was inserted into the uterine part where there was no placenta and the fetus was inserted. Normal saline fluid as much as 150 ml (Figure 2) and re-evaluation after the procedure, from the results of ultrasound after the procedure, the SDP increased to 2.99 cm (Figure 2C).

Received : January 10th, 2022
Accepted : January 22th, 2022
Correspondence: Rizka fadhillah Yusra, email: *rizkayusra@yahoo.com*

Vol. 6, No. 1, Jan-Jun 2022
ANDALAS OBSTETRICS AND GYNECOLOGY JOURNAL
Address for Correspondence:
Editorial Room Andalas Obstetrics and Gynecology Journal, 3rd floor of KSM of Obstetrics and Gynecology, RSUP DR. M. Djamil Padang, Jl. Perintis Kemerdekaan Padang, Sumatera Barat 25127
eISSN : 2579-8324 pISSN : 2579-8323
Website:
http://jurnalobgin.fk.unand.ac.id/index.php/JOE

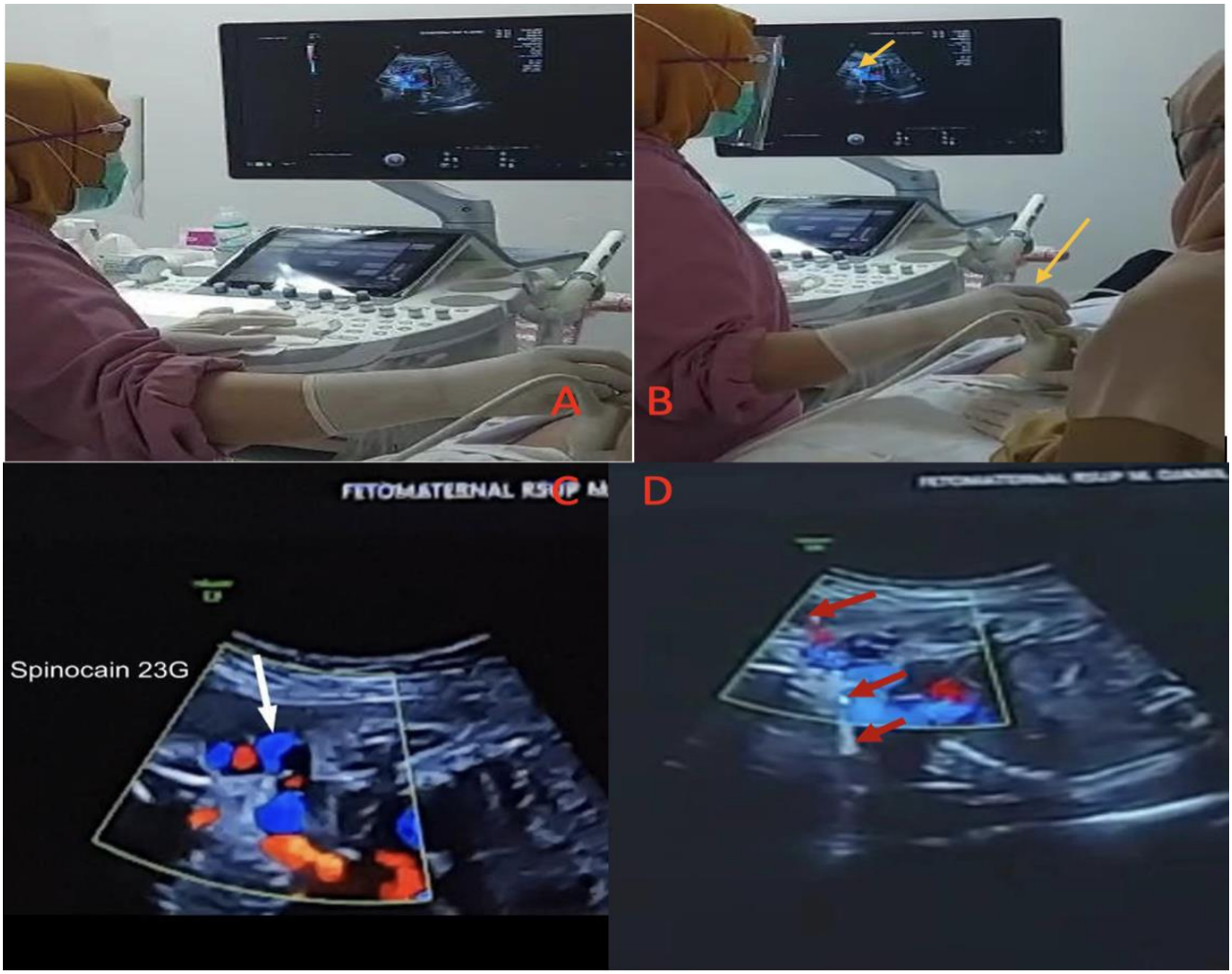

Figure 2: A; an ultrasound evaluation was performed and searched for access to the uterus for amnioinfusion, namely the part of the uterus that was free from the placenta and the fetal part, B; After the location is obtained (A), entry with spinocaine Fr 23 into the amniotic cavity with ultrasound and Doppler guidance (yellow arrow), C; spinocaine fr 23 into the amniotic cavity D; seen 0.9% NaCl fluid flowing into the uterine cavity through the spinocaine needle fr 23 with transabdominal ultrasound guidance (red arrow)

Received : January 10th, 2022
Accepted : January 22th, 2022
Correspondence: Rizka fadhillah Yusra, email: *rizkayusra@yahoo.com*

Vol. 6, No. 1, Jan-Jun 2022
ANDALAS OBSTETRICS AND GYNECOLOGY JOURNAL
Address for Correspondence:
Editorial Room Andalas Obstetrics and Gynecology Journal, 3rd floor of KSM of Obstetrics and Gynecology, RSUP DR. M. Djamil Padang, Jl. Perintis Kemerdekaan Padang, Sumatera Barat 25127
eISSN : 2579-8324 pISSN : 2579-8323
Website:
http://jurnalobgin.fk.unand.ac.id/index.php/JOE

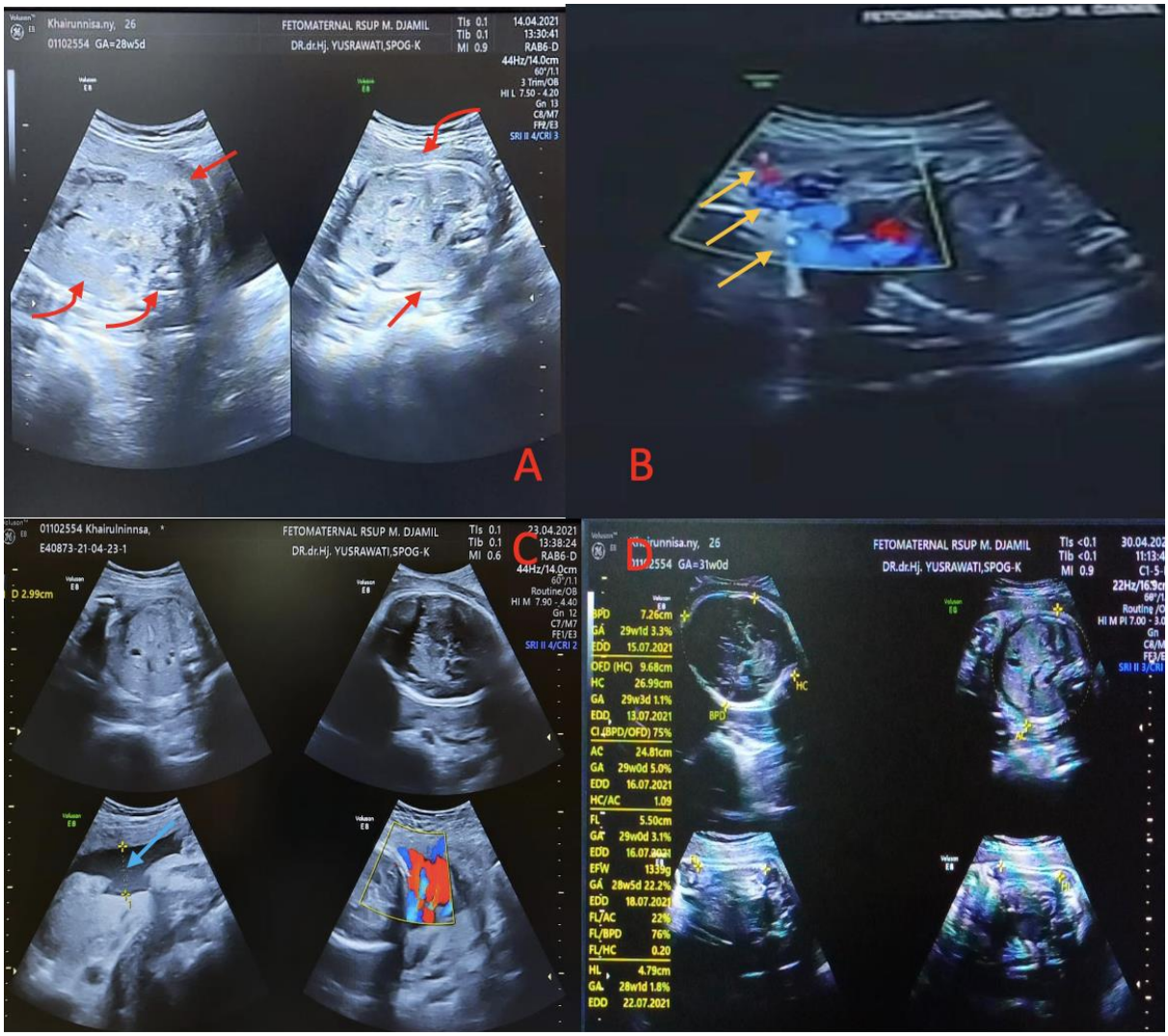

Figure 3: A; anhydramnios (red arrow) pre-amnioinfusion, B; Amnioinfusion process; picture of fluid entering the amniotic cavity (yellow arrow), C; post amnioinfusion; SDP 2.99 cm (blue arrow), D; Ultrasound on the 3rd day after the 2nd amnioinfusion, again found anhydramnios, SDP is difficult to assess, but the fetal organs can be visualized well for biometric measurements.

The second amnioinfusion was carried out 3 days after the first amnioinfusion, by inserting 500 ml of 0.9% Nacl, 3 days after that an ultrasound was re-evaluated and the condition of anhydramnios with SDP was difficult to assess (Figure 3D). The patient is then planned for periodic monitoring and amnioinfusion until the fetus is viable for delivery

DISCUSSION

Amniotic fluid is an important component for fetal growth and development during pregnancy. Early in embryogenesis, the amnion is an extension of the extracellular matrix and there is bidirectional diffusion between the fetus and the amniotic fluid. At 8 weeks of

Received : January 10th, 2022
Accepted : January 22th, 2022
Correspondence: Rizka fadhillah Yusra, email: *rizkayusra@yahoo.com*

Vol. 6, No. 1, Jan-Jun 2022
ANDALAS OBSTETRICS AND GYNECOLOGY JOURNAL
Address for Correspondence:
Editorial Room Andalas Obstetrics and Gynecology Journal, 3rd floor of KSM of Obstetrics and Gynecology, RSUP DR. M. Djamil Padang, Jl. Perintis Kemerdekaan Padang, Sumatera Barat 25127
eISSN : 2579-8324 pISSN : 2579-8323
Website:
http://jurnalobgin.fk.unand.ac.id/index.php/JOE

gestation, the urethra is formed and the fetal kidneys begin to produce urine. Then the fetus begins to swallow. Excretion from the urine, respiratory system, digestive system, umbilical cord and placental surface are sources of amniotic fluid. Fetal urine production begins at between 8 and 11 weeks of gestation, but it does not become a major component of amniotic fluid until the second trimester, which explains why fetuses with severe renal impairment are asymptomatic until 18 weeks of gestation.[7,15]

Oligohydramnios is defined by an abnormal decrease in the amount of amniotic fluid.[4] The amount of amniotic fluid changes with gestational age and the accurate way of estimating the amount of amniotic fluid has changed in recent years. Oligohydramnios can be defined by; 1. The amount of amniotic fluid is less than 500 ml at 32-36 weeks of gestation; 2. Deepest vertical pocket (MVP) less than 2 cm in late 2nd trimester of pregnancy; 3. The amniotic fluid index (AFI) is less than 5 cm or less than the 5th percentile in the late 2nd trimester of pregnancy.[4,8,16]

Anhydramnios is a condition where there is no amniotic fluid due to excessive fluid loss or reduced urine production or urinary excretion.[7] Amniotic fluid is required to maintain lung fluid within the lungs to promote alveolar distention and growth and to maintain a transpulmonary gradient. The low amniotic fluid volume allows lung fluid to flow from the trachea and causes alveolar compression, affecting lung distention and growth by compressing the chest cavity and allowing fetal lung fluid to escape from the lungs.[9] Pulmonary fluid volume increases with lung weight and by the third trimester, epithelial secretions in the fetal lung produce about 25 mL/kg of lung fluid, which makes up about 90% of the lung weight. During fetal breathing movements, the fluid passes through the trachea and is swallowed or mixed with the amniotic fluid. During the non-respiratory period, the positive pressure of the amniotic fluid in the upper respiratory tract inhibits the outflow of lung fluid, keeping it in the trachea by the glottis. This creates the transpulmonary pressure gradient necessary to maintain alveolar distention above the functional residual capacity of the newborn and promote lung growth.[2,10,17]

On ultrasound, bilateral renal agenesis is suspected if no part of the kidney is found or only a small portion of tissue is found where the kidney should be. Examination is done at 18 weeks of gestation, usually will find a small amount of amniotic fluid, bladder that is not visible, or looks very small. This deficiency can cause malformations in the baby due to lack of space. The most common malformations are lungs that are too small and joints that are too stiff.[5,18,19]

The mechanism of anhydramnios can be related to bilateral renal agenesis, where the bladder and kidneys are not found after the second trimester of pregnancy, bilateral renal agenesis is one of the primary causes of Potter's syndrome. Potter syndrome and the Potter phenotype is a complex condition associated with congenital renal failure, one of which is bilateral renal agenesis and is associated with oligohydramnios [4,7]. Potter's syndrome is described as a characteristic condition of the newborn, in which there is very little or no amniotic fluid.[11] Oligohydramnios causes the baby to have no cushion against the uterine wall. Pressure from the uterine wall causes a characteristic facial appearance (Potter's face). In

Received : January 10th, 2022
Accepted : January 22th, 2022
Correspondence: Rizka fadhillah Yusra, email: *rizkayusra@yahoo.com*

Vol. 6, No. 1, Jan-Jun 2022
ANDALAS OBSTETRICS AND GYNECOLOGY JOURNAL
Address for Correspondence:
Editorial Room Andalas Obstetrics and Gynecology Journal, 3rd floor of KSM of Obstetrics and Gynecology, RSUP DR. M. Djamil Padang, Jl. Perintis Kemerdekaan Padang, Sumatera Barat 25127
eISSN : 2579-8324 pISSN : 2579-8323
Website:
http://jurnalobgin.fk.unand.ac.id/index.php/JOE

addition, because the space in the uterus is narrow, the limbs become abnormal or contracture and are fixed in an abnormal position. [8,9,18,20]

In this patient no risk factors were found from the mother, so it was suspected that the cause of oligohydramnios in this patient came from fetal factors. Where the results of ultrasound of the fetal kidneys are difficult to assess, it is suspected that renal agenesis or congenital abnormalities in the urogenital are suspected. Pathophysiologically oligohydramnios occurs because of a condition that causes excessive amniotic fluid expenditure or reduced fetal urine production. In this case, the patient had no history of ruptured membranes or vaginal discharge, so the suspicion of reduced urine production due to fetal congenital abnormalities was increasing.

Amnioinfusion is a prenatal action by inserting fluid into the amniotic cavity, either transabdominally or transvaginally.[6] Amnioinfusion is an attempt to restore physiological conditions of the intrauterine environment for the fetus. Amnioinfusion reduces uterine pressure resulting from anhydramnios and maintains alveolar distention to promote fetal lung growth. Amnioinfusion also aims to prevent pulmonary hypoplasia and promote fetal survival. Considering pulmonary hypoplasia is a very lethal condition for the fetus, currently no treatment has been found for pulmonary hypoplasia after the baby is born, so intrauterine surgery is the only way that is expected to overcome this pulmonary hypoplasia.[2,12]

Amnioinfusion is both diagnostic and therapeutic. Amnioinfusion can be performed antepartum or intrapartum. Diagnostic amnioinfusion is performed antepartum with the aim of improving sonographic assessment in the interest of prenatal diagnosis.[6,18] In this patient amnioinfusion was performed to improve the diagnostics, whereas there were no renal and bladder features after amnioinfusion which confirmed the diagnosis of bilateral renal agenesis: Potter's syndrome in this patient. The therapeutic goal of amnioinfusion in this patient is to minimize contractures in the fetus, provide space for the fetus to move and minimize the incidence of pulmonary hypoplasia that may be experienced by the fetus. After the first amnioinfusion, the SDP increased to 2.99 ml, but when ultrasound was re-evaluated 3 days later, anhydramnios was found again, this proves that the physiology of amniotic flow continues but insufficient production of amniotic fluid results in persistent anhydramnios if not performed. intervention. Amnioinfusion is done periodically to improve the prognosis of live birth of the fetus.

CONCLUSION

Anhydramnios in this case was caused by bilateral renal agenesis; Potter's syndrome. Amnioinfusion aims to assist in diagnosis and as a therapy to prevent contractures and pulmonary hypoplasia in the fetus and increase life expectancy when the fetus is born.

Received : January 10th, 2022
Accepted : January 22th, 2022
Correspondence: Rizka fadhillah Yusra, email: *rizkayusra@yahoo.com*

Vol. 6, No. 1, Jan-Jun 2022
ANDALAS OBSTETRICS AND GYNECOLOGY JOURNAL
Address for Correspondence:
Editorial Room Andalas Obstetrics and Gynecology Journal, 3rd floor of KSM of Obstetrics and Gynecology, RSUP DR. M. Djamil Padang, Jl. Perintis Kemerdekaan Padang, Sumatera Barat 25127
eISSN : 2579-8324 pISSN : 2579-8323
Website:
http://jurnalobgin.fk.unand.ac.id/index.php/JOE

REFERENCES

1. Abuhamad A. Ultrasound Obstetric and Gynecology , Chapter 9 : Amniotic Fluid Assessment.: Davies Publishing, Incorporated; 2014. 178-84
2. Elizabeth., Angie. OH, Rodrigo. JJLM, Ruanoe, Meredith., Ahmet. A, et al. Amnioinfusions to Treat Early Onset Anhydramnios Caused by Renal Anomalies: Background and Rationale for the Renal Anhydramnios Fetal Therapy Trial. Fetal Diagn Ther 2019;45:365–72.
3. Chauhan N, Namdeo P, JN. Evidence Based Management of Oligohydramnios. . Modi Open Access Journal of Gynecology Medwin Publishers 2018;Vol.3(3).
4. Cunningham. Williams obstetrics, 25 th, Chapter 11: Disorders of Amniotic fluid.: McGraw-Hill Education; 2018.
5. Fred E, Laulom B, Cassart M, Eurin D, Nassez A, Hall M. Ultrasonography In Obstetrics and Gynecology; Callen. . United States of America Elsevier:; 2008.
6. Vikraman SK, Chandra V, Balakrishnan B, Batra M, Sethumadhavan S, Patil SN, et al. Impact of antepartum diagnostic amnioinfusion on targeted ultrasound imaging of pregnancies presenting with severe oligo-and anhydramnios: An analysis of 61 cases. European Journal of Obstetrics & Gynecology and Reproductive Biology. 2017;212:96-100.
7. Ross MG, Beall MH. Physiology of amniotic fluid volume regulation. UpToDate, Wolters Kluwer. 2017;Mey.
8. Marva Moxey-Mims MD, F.A.S.N.1 and Tonse N. K. Raju, M.D., D.C.H. Anhydramnios in the Setting of Renal Malformations: The National Institutes of Health Workshop Summary. Obstet Gynecol. 2018;131 (6):1069–79.
9. Bhandari J, Thada PK, Sergent SR. Potter Syndrome. StatPearls 2020.
10. Tisekar, Owais R, Kumar A. Hypoplastic Lung Disease. StatPearls 2020.
11. Yusrawati, Andi Friadi. Diagnosis Prenatal Hidronefrose dengan Ultrasonografi (laporan kasus). INAJOG, 2007;31 (1): 42-8
12. Cunningham GF. 2018. Disorders of Amniotic fluid. Williams obstetrics, 25th ed., Chapter 11, Hal: 236-238.
13. Micheal GR, Marie HB. 2017. Physiology of Amniotic Fluid Volume Regulation. UptoDate. Wolters Kluwer.
14. Moise KJ Jr; Toward consistent terminology: assessment and reporting of amniotic fluid volume. Semin Perinatol. 2013 Oct;37(5):370-4. doi: 10.1053/ j.semperi. 2013.06.016.

Received : January 10th, 2022
Accepted : January 22th, 2022
Correspondence: Rizka fadhillah Yusra, email: *rizkayusra@yahoo.com*

Vol. 6, No. 1, Jan-Jun 2022
ANDALAS OBSTETRICS AND GYNECOLOGY JOURNAL
Address for Correspondence:
Editorial Room Andalas Obstetrics and Gynecology Journal, 3rd floor of KSM of Obstetrics and Gynecology, RSUP DR. M. Djamil Padang, Jl. Perintis Kemerdekaan Padang, Sumatera Barat 25127
eISSN : 2579-8324 pISSN : 2579-8323
Website:
http://jurnalobgin.fk.unand.ac.id/index.php/JOE

15. Cicily TJ, Sherin S, Anitha KG.2017. Effect of hydration therapy on oligohydramnios. Int. J. Reprod Contracept Obstet Gynecol. May;6(5):1800-1805.
16. Krishna J, Nisha S, Sachin P. 2013. Maternal and Fetal Outcome in Oligohydramnios : a studi 100 kasus. International Journal of Medical Science and Publoic Helath. Vol 2; 3.
17. Royal Cornwall Hospitals. 2016. Amniotic Fluid Volume Abnormality : Diagnosis and Management.
18. Sonkhya P, Gupta N, Mittal P. 2017. Pregnancy Outcomes of Oligohydramnios by Ultra Sonography (USG): Penelitian Case Control; International Multispecialty Journal of Health (IMJH). Vol.3(2).
19. Elizabeth AD, Everett FM. 2013. Amniotic Fluid as a Vital Sign for Fetal Wellbeing. AJUM 16(2)
20. Nimra Dad, Mandy Abushama, Justin C. Konje, and Badreldeen Ahmed. 2016. What is the role of amnioinfusion in modern day obstetrics?. J Matern Fetal Neonatal Med; 29(17): 2823–2827

Received : January 10th, 2022
Accepted : January 22th, 2022
Correspondence: Rizka fadhillah Yusra, email: *rizkayusra@yahoo.com*