

CASE REPORT: ANENCEPHALY ON G2P1A0 GRAVIDA 16-17 WEEKS PREGNANT WOMEN WITH COVID 19 MILD SYMPTOMS

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ABSTRACT

Background: Anencephaly is severe congenital malformation of the central nervous system (CNS), with complete or partial absence of the cranium and brain. The incidence of this case is estimated to be between 1-5 events per 1000 births. Anencephaly arises from an embryonal defect before day 28 so that there is no closure of the anterior neuropore (cranial end of the neural tube). Case Summary: Case report about patient woman aged 21 years with COVID-19 infection who never had an ultrasound in the first trimester with complaints of abdominal pain a day ago, diminished fetal movement, there was not a release of blood show and mucus. An ultrasound examination performed in the second trimester and vaginal delivery is performed. Conclusion: Anencephaly is a malformation that has a high mortality according to the failure of the closure of the sefic part of the neural tube. The ISUOG guidelines recommend that ultrasonography be performed on pregnant women between 11 and 13 weeks. Through this paper, an explanation of the transabdominal and transvaginal 2D grayscale ultrasound examination to assess crown rump length and cranium at 11-13 weeks of pregnancy can detect anencephaly early.

INTRODUCTION

Anencephaly is a severe congenital malformation of the central nervous system (CNS), with complete or partial formation of the cranium and brain, absence of both cerebral hemispheres, absence of cranial bones and structures originating in the forebrain and skull. Anencephaly is a lethal condition, with a 100% mortality rate that can occur intrauterine or within hours to days after birth, and carries increased medical risks for the mother (Munteanu *et al.*, 2020).

The number of cases of anencephaly varies, estimated to be between 1-5 per 1000 births (Munteanu *et al.*, 2020). Although the etiology of anencephaly remains unclear, there are various risk factors that can be attributed, in addition to genetic factors (Agopian *et al.*, 2013). Risk factors include environmental factors, maternal factors as well as fetal factors (Muglia *et al.*, 2022). The risk of recurrence in subsequent pregnancies for cranial or spinal cord defects is 10% (Avagliano *et al.*, 2019). In families, the birth of anencephaly may be followed by the birth of a second child affected by the lumbar-sacral meningocele (Wilson *et al.*, 2021). (2023)

Anencephaly arises from an embryonal defect before day 28 and thus does not occur closure of the anterior neuroporus (cranial end of the neural tube).

The prenatal diagnosis of anencephaly can be determined by ultrasound as well as by assessing the concentration of α -fetoprotein in amniotic fluid and maternal serum (Aboughalia *et al.*, 2020). Pregnancy with a fetus with anencephaly carries an increased medical risk for the mother, so prospective parents should be offered the opportunity for termination of pregnancy. The percentage of termination of pregnancy is more than 83% (Jacobs *et al.*, 2015).

CASE REPORT

Pregnant woman, Mrs. N, age 21 years, a housewife, last education Elementary School, G2P1A0 gestational age 16-17 weeks with complaints of abdominal pain since 1 day ago felt not penetrating back accompanied by reduced fetal motion since 1 week. There were no complaints of discharge or bleeding from the birth canal. The patient denies any contact with the pet. History of falls and abdominal pain is absent. The history of antenatal care examination is incomplete, including not screening during pregnancy (TORCH, HIV, Syphilis, and HbsAg), because they do not know about the pregnancy until 8 weeks gestation. History of heartburn in mothers by getting drugs Antacids, Ampicillin and also Paracetamol. In early pregnancy, the mother had illnesses such as fever, cough and runny nose, but did not seek treatment. The mother denied having a history of hypertension, diabetes mellitus, heart disease, seizures, and infectious diseases or vaginal discharge during pregnancy. A history of allergies to food and drugs is also denied. Previous operation history is missing. A family history of systemic diseases and congenital abnormalities is denied.

The daily activity of mothers is as a housewife, by continuing to do ordinary activities such as cooking, washing clothes and also drawing water (Shahul Hamid *et al.*, 2023). Mother has a good relationship with her partner (Golombok *et al.*, 2023). The mother lives with her partner in the dormitory of an oil palm plantation company (Pulungan *et al.*, 2023). Mothers usually eat 1-2 times a day, with small portions due to complaints of nausea and also difficult to buy groceries in the area where they live (Harja *et al.*, 2023). The patient and partner deny having a history of smoking, drinking alcohol and illegal drugs (Board *et al.*, 2023).

History of obstetrics of the first child born at home in 2017, born term, female gender, spontaneous birth with the back of the head assisted by health workers, BBL 3100 gr. There is no history of taking pregnancy vitamins (folic acid and iron) until 8 weeks gestation (Guye *et al.*, 2023). Mom had time to do an examination Ultrasound in the first hospital 4 days before and said fetus with anencephaly (Mirabal-Beltran *et al.*, 2023). Then, the mother wants to check her uterus to another doctor to get a second opinion.



Figure 1. Ultrasound at the First Hospital (JTHIU, DJJ: 161x/min, TBJ: 205 grams, fetus with anencephaly)

Physical examination obtained the general state of moderate pain, compos mentis consciousness. Anthropometric status, weight 70 kg, height 160 cm, with body mass index 27.65 kg/m². Vital signs examination blood pressure 120/70 mmHg, pulse 74x/min, respiration 20 x/min and temperature 37.8°C. Generalized status within normal limits. Obstetric examination, enlarged abdomen with striae gravidarum, TFU at 2 fingers below the center (16 cm). On inspection inside, it was found that the soft portio had no opening. In laboratory examination, WBC 18.3 x 10³ / uL, RBC 3.6 x 10⁶ / uL, HGB 10.6 g / dL, and PLT 219 x 10³ / uL, GDS 89 mg / dl, CT 7", BT 2", as well as SARS Cov-2 Antigen and PCR Positive. Supporting examinations carried out ultrasound examination found fetuses with anencephaly.

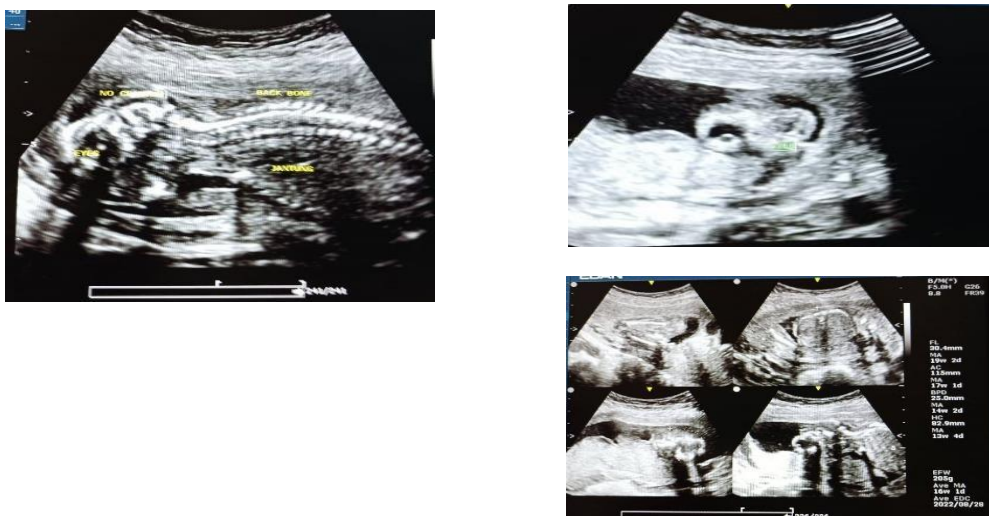


Figure 2. Ultrasound in the Second Hospital (Fetus with anencephaly)

Based on the history, physical examination, and supporting examinations, the working diagnosis of this case is G2P1A0 gravid 16 weeks with polyhydramnios e.c anencephaly. In these patients, management is carried out by termination of pregnancy by induction of vaginal delivery. Pregnancy was induced by administration of misoprostol 400 µg / 12 hours / oral, and also given the drug Favipiravir (Avigan) 600 mg / 12 hours / oral, calcium lactate 500mg / 12 hours / oral. The mother gave birth to a stillborn anencephaly baby girl with intact amniotic membranes per vagina, and

added Cefixime drugs 200 mg / 12 hours / oral, paracetamol 500mg / 8 hours / oral if fever. The patient's postpartum condition was good and then the patient was allowed to go home.

RESEARCH METHODS

This research uses qualitative methods with a literature review approach. Literature review is a systematic, explicit and reproducible method for identifying, evaluating and synthesizing research works and thoughts that have been produced by researchers and practitioners. The step in writing this review literature begins with the selection of topics. Search libraries or sources to gather relevant information from Google Scholar, CINAHL, Proquest, Ebsco, or National Library databases. Determine keywords or keywords for journal searches. After the data is collected, it is processed, analyzed and conclusions drawn.

RESULTS AND DISCUSSION

Anencephaly babies with external examination appear to be babies without craniums, short necks and protruding eyeballs. The thoracic skeleton appears normal, and no spina bifida appears. The abdomen was abnormal, and no anomalies were found. The extremities do not appear abnormal, with fingers that have separated. This anencephaly baby is classified as a type of meroanencephaly, which is there is rudimentary brain tissue and some skull formation.

In normal human embryos, nerve plates begin to form approximately 18 days after fertilization. At the development of the 4th week, the nerve plate invaginates along the midline forming a nerve groove. The closing process starts from the middle and then towards both ends of the neural tube. The cranial part closes on the 25th

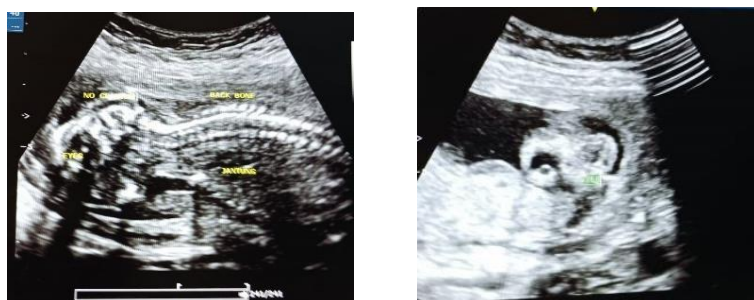


day and the caudal part closes on the 28th day. Anencephaly arises from an embryonal defect before day 28 so that there is no closure of the anterior neuroporus (cranial end of the neural tube). The brain and calvaria may be only partially or completely absent.⁴

Figure 3. Photos of Babies with Anencephaly After Vaginal Delivery Development of pregnancy in the period

Critical neurulation, namely the first 3-4 weeks of pregnancy, can be evaluated from the book Maternal and Child Health (MCH) and supporting examinations carried out. The mother's MCH book is not completely filled and the mother does not get folic acid tablet supplementation periconception, so we cannot assess fetal development at that gestational age. Prenatal diagnosis can be established by ultrasound examination (ultrasound) and elevated levels of α -fetoprotein.³ However, this α -fetoprotein examination is not performed on the mother in cases due to limited resources. Early detection by ultrasound examination can be done from 11-13+6 weeks gestation, using 2-dimensional grayscale ultrasonography (USG), transabdominal and transvaginal.⁵

Transabdominal ultrasound examination is performed on the mother and a characteristic picture of anencephaly is found. Ossification of the cranium bone is complete at 11 weeks gestation, so that at 16 weeks gestational ultrasound should have seen the head with cranium bone, but on ultrasound examination the case found the head but without the cranium bone on it, which showed signs of anencephaly. On ultrasound examination also found other images such as "frog eye" and obtained AFI > 24 mm due to the presence of polyhydramnion, this is due to amniotic fluid formed, routinely removed and replaced with a new one. One way of production is swallowed by the fetus, absorbed by the intestine then flowed into the placenta to finally enter the mother's blood circulation. This amniotic fluid excretion will be disturbed if the fetus cannot swallow. In the case of anencephaly there is an increased transudation of fluid from the exposed meninges into the amniotic cavity.^{4,8}



Gambar 4. Ultrasonografi Janin Anensefali UK 16-17 minggu

After the prenatal diagnosis in this case of anencephaly is established, the choice for termination of pregnancy should be communicated to the married couple. For couples who choose to continue the pregnancy, the possibility of preterm labor, polyhydramnios, non-advanced labor, and delayed onset of labor until past its time should also be discussed. In these patients, the option to terminate the pregnancy

has been given to the family since the first ultrasound but the family wants a second opinion and is finally willing to do the termination.

Anencephaly is caused by multifactorial factors including genetic and environmental factors. Specific genes associated with anencephaly have not been identified.² However, research suggests that the presence of 5,10-Methylene Tetrahydrofolate Reductase (MTHFR), an enzyme associated with folate synthesis increases the risk of anencephaly and also spina bifida in the fetus.^{3,4,6} For genetic factors requiring further examination, while resources and examination are very limited in the rural area of this case so we exclude this factor.

Many studies have reported on the protective effects of folic acid supplementation in the first trimester of pregnancy.^{1,7} In fact, folic acid supplementation during the periconception period is said to lower the risk by 40-80%.⁶ Mothers do not get periconception folic acid supplementation and also inadequate daily food intake, so the mother can develop folic acid deficiency anemia, which is supported by laboratory test results (Hb 11,6), which will increase the risk of anencephaly in the fetus.

Hyperthermia has been suspected as a neuroteratogen for embryos, as it interferes with important developmental events such as cell proliferation, migration, differentiation and apoptosis.⁸ Maternal hyperthermia and maternal exposure to non-physiological high temperatures (e.g. in saunas, hot tubs) during critical periods of neurulation are associated with an increased incidence of NTDs (odds ratio 1.95) suggesting that the neural tube of human embryos is sensitive to heat.^{6, 7} The mother had a fever in early pregnancy that may be related to the occurrence of anencephaly. However, it is not known the cause of the mother having a fever, due to the mother who did not check herself at a health facility.

The mother stayed in the oil palm plantation area to accompany her husband. Exposure to nitrates and pesticides can double the risk of NTDs, especially fetal anencephaly. Nitrate exposure can be found in agricultural communities, where wells can be contaminated by fertilizers and animal waste. In fact, paternal exposure to pesticides during the periconception period is associated with an increased risk of having children with anencephaly, given the fact that these products have toxic effects on male gametes, altering the migration and transfer of deoxyribonucleic acid (DNA) during meiosis.¹ Low education levels and socioeconomic status are also associated with an increased risk of having babies with anencephaly.^{1,9} The mother's last education was primary school, is associated with a lack of knowledge and awareness of important things in pregnancy e.g. vitamin supplementation, and also the possibility of exposure to toxic substances.

Continuous exposure to amniotic fluid (polyhydramnios) and certain mechanical trauma is associated with degeneration and disorganization of nerve tissue, which is covered by edematous and angiomatous membranes.⁷ Mother carries out usual activities at home such as lifting heavy items on her own. This activity can increase the risk of trauma during pregnancy.

It has been reported that anencephaly is more common in female fetuses than males with a ratio of 0.75.^{1,6,7} The fetus in cases is also female. The reason

predominant anencephaly occurs in women is still unclear, but it is thought to be due to genetic or epigenetic effects associated with sex, and this requires further research.¹⁰

Other factors associated with fetal anencephaly are Diabetes Mellitus, obesity, infections during pregnancy (TORCH, syphilis), drug use (anti-epileptic), radiation, stress.^{1,6,8} These factors were not found in this case.

Anencephaly is a lethal condition, with a 100% mortality rate, either dying intrauterine (about 65% of cases) or within hours to days after birth. Detection of anencephaly during early pregnancy is generally followed by termination of pregnancy, which is more than 83%.^{1,3,11} The decision of termination is taken by the mother and family on the explanation of two obstetricians. After termination, the baby is stillborn with the amniotic membrane intact.

The risk of having another child with anencephaly with a positive family history varies between 2% and 10%, compared with 0.006% in women without a previous history.¹ Adequate folic acid intake, at least 400 µg per day, especially before pregnancy is necessary as a preventive measure. Although expectant mothers have gene mutations that are risk factors for anencephaly, adequate folic acid intake can reduce the risk.⁴ In addition, it is also necessary to detect abnormalities as soon as possible during pregnancy.³ It is necessary to educate mothers about the level of recurrence and the importance of adequate nutritional intake, especially periconception folic acid. Pregnancy must be planned with adequate preparation, as well as early detection of abnormalities during pregnancy by routinely conducting antenatal care examinations and recommending transabdominal and transvaginal 2D grayscale ultrasound examinations to assess crown rump length and cranium bones at 11-13 weeks of pregnancy.

CONCLUSION

Anencephaly is a serious failure of the development of the central nervous system in which the brain or cranium is largely unformed. Anencephaly is part of the spectrum of neural tube defects (NTDs), which arise from an embryonal defect before day 28 where there is no closure of the anterior neuroporus (normally closed on day 25), which can eventually lead to intrauterine fetal death or neonatal death. The risk factors that play a role in causing this anencephaly include genetic abnormalities (mutations), folic acid deficiency, maternal hyperthermia, history of disease in the mother, the use of certain drugs such as anti-epileptics, and various substances that are teratogenic. Anencephaly is a lethal condition with increased medical risks to the mother, so after detection of anencephaly is done, the mother and partner should be informed of the termination of pregnancy. The most important prevention is the adequate intake of folic acid during pregnancy, at least 400 µg per day, especially since before pregnancy and the early stages of pregnancy. In addition, it is also necessary to detect abnormalities as soon as possible during pregnancy, with transabdominal and transvaginal 2D grayscale ultrasound examining the crown rump length and cranium bones at 11-13 weeks gestation, to provide proper preparation and management.

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