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A Multiparity Woman with Pregnancy of Recurrent Anencephaly: A Rare Case Report

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Abstract:- Anencephaly is birth defect of neural tube.The recurrent of anencephaly is a rare case. This study reports of a 35-years old woman gravida who detected her second fetal anencephaly in seventeen-weeks gestation. She had her first fetal anencephaly seven years before which was late known in fourty weeks gestation. The awareness of high-risk maternal group of anencephaly is low. Management strategies should be promoted for preventing the recurrent case of anencephaly pregnancies.

Keywords:- Anencephaly, Recurrent Anencephaly, Neural Tube Defect, Birth Defect, Rare Case.

I. INTRODUCTION

Neurulation is formed at tenth of carnegie stage and formed of the brain at thirteenth of carnegie stage.[1]Anencephaly is caused by this phasemal formation that caused by multi factorial, such as folic acid deficiency, genetic, maternal comorbid, and nutrition.[2]Although it is preventable, recurrent anencephaly that is rare, have been reported in several studies. The rate in Europe during 2000-2010 is 3.52 per 10,000 live births. In America during 1991-2001, it is 9.4 per 100,000 per live births.[3] Indonesia has no data for anencephaly prevalence. However, neural tube defect prevalence is 18.4% from 956 congenital abnormalities during 2014-2018.[4]

II. CASE HISTORY

A 35-years old woman came to outpatient clinic with 17 weeks gestation. She reffered from another obstetrician who diagnosed fetal anencephaly. It was her fourth gestation and second anencephaly pregnancy. The first gestation was delivered 10 years ago with section cesarean as indicated by premature rupture membrane. The second gestation with late-known anencephaly had delivered seven years before with section cesarean in fourty weeks gestation. Five years ago, the third gestation was delivered, however he was gone at two months-old because of aspiration. The patient had negative ToRCH(*Toxoplasma gondii, Rubella, Cytomegallo virus, Herpes simplex virus*) test result at six years before and no history of contraception. During antenatal care, the blood pressure and the fetal heart rate were normal. Based on ultrasonography examination in seventeenth weeks gestation, there were frog sign and brain parenchym (fig 1). The patient had ultrasonography evaluated every two weeks. After four weeks from her first visit, the ultrasonography result was cerebellum out of the cranium which is indicated of fetal anencephaly (fig 2). Patient was advised to termination. Pervaginam termination was choosen with misoprostol 200mcg combined with catether pendulum.The anencephaly baby was delivered after two days of cervix rippening. During post-partum control, she was tested of cobalamine (339 pg/mL; standard level: 187 – 883 pg/mL), folic acid (17.94 ng/mL; standard level: 7.20-19.40 ng/mL) and homocysteine (9,52 umol/L; standard level: 4,44 – 13,56 umol/L). In this case there is no MTHFR gene (*methylenetetrahydrofolate reductase*) evaluation because of the limited resources of hospital.

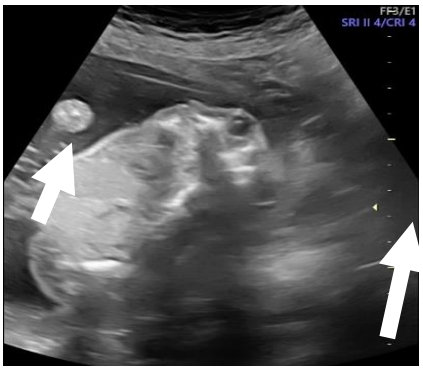

Fig 1. ultrasonography at 17 weeks gestation. White arrow showed the frog sign and brain parenchym.

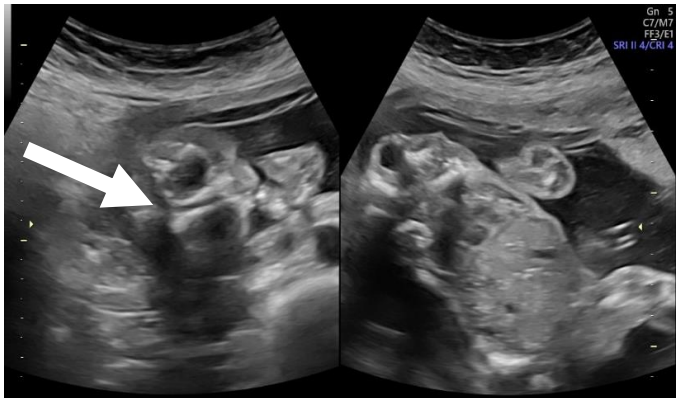

Fig 2. ultrasonography evaluation at 19 weeks gestation. White arrow showed the cerebellum outside the cranium

III. DISCUSSION

Anencephaly is a rare and low recurrent case. It is multifactorial inherited,[5]such as bad nutrition, comorbid status, supplement deficiency and teratogenic ingredients.[6]In this case, the patient has no comorbid.Therecurrent rate of

anencephaly in one parent sibling is 1.8% and different parent sibling is 0.8%. Recurrent rate of anencephaly is 3.15% at subsequent child if the first child with anencephaly.[7]Based on epidemiology data, female fetus increases the prevalence, still there is no evidence.[8] In this case, the patient had twice anencephaly and both of them are female.

Anencephaly screening should be started in the first trimester gestation with regular evaluation. Termination can be plan if ultrasonography confirmed the case. High AFP (alpha-feto protein) serum testing in first and second trimester can be a screening test. The test considered if there is cranium lucent at ultrasonography of suspected fetus.[9] Folic acid should be tested in suspected cases. low of folic acid can inhibit the availability homocysteine metabolism. it will result of high amount level in extracellular that can be teratogenic and increase the excitotoxicity through simulation of NMDA receptor, then DNA neuron will be damaged apoptosis.[9]The prevention of recurrent anencephaly should be started from periconception stage (28 days before fertilization) because the forming of neurogenic system starts in 3-4 weeks early gestation. Moreover, in high risk maternal, the supplement should to consumed at 3 months before conception.[6]In this patient, there is no prevention program of recurrent anencephaly, because the lack of awareness and the unpredictable gestation. The lack of preconception counseling service facilities, and the lack of detail in providing explanations and prescribing folic acid during pregnancy also contribute to the rate of recurrent anencephaly.

IV. CONCLUSSION

Anencephaly is a neurulation failure that occurs in the third or fourth week after fertilization. Anencephaly caused by multifactorial, so it should be prevented. In this case, a patient with recurrent anencephaly was spontandelivered with a pendulum catheter. Evaluation of maternal folate, cobalamin and homocysteine were within normal limits and the ToRCH test was negative. In this case, it is not known the underlying risk factors for recurrent anencephaly, while no ToRCHinfection and nutritional status are within normal limits.

V. CLINICAL MESSAGE

Anencephaly is a preventable congenital abnormality because of the multi factorial. preventable program should be massively informed to the maternal group who have high risk of it. primary health care should contribute to this case to educate, advocate and encourage to the maternal group about this issue.

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CONFLICT OF INTERESTS

The authors state that this article has no competing interest.

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