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Case Report

Expectant management in dichorionic diamnionic twin pregnancy discordant for anencephaly

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Abstract

Dichorionic Diamnionic Twin Pregnancy Discordant for Anencephaly is a rare occurrence with only very few reported cases. We report a case of dichorionic twin pregnancy discordant for fetal anencephaly delivered at 37 to 38 weeks of gestation when she had premature rupture of membrane and was managed expectantly.

Keywords: twin pregnancy, anencephaly, dichorionic, diamionic, discordant

INTRODUCTION

Anencephaly occurs in 1.4 to 4.7 per 10,000 singleton deliveries (Wilson et al., 2009), and is thought to result from failed closure of the anterior neuropore at 24 to 26 days post fertilization. However, in twin pregnancy wherein the co – twin is normal while the other twin is discordant for anencephaly, it occurs in 1 per 500 million live birth (Callen PW, 2008).
Twin pregnancy discordant for fetal anencephaly is a serious condition that can be detected by serial ultrasound examination (Johnson et al.,1997), and it poses a serious consequences. Twin pregnancies complicated with one anencephalic fetus poses an increased risk of either neonatal death due to severe preterm delivery secondary to polyhydramnios (due to lack of swallowing reflex) or even intrauterine death (Sebire et al., 1997; Leeker and Beinder 2004). About 55% of pregnancies are complicated with polyhydramnios in dichorionic twins discordant for anencephaly, and this is the main risk to the co – twin (Vandecruys et al., 2006). In monochorionic twins, polyhydramnios and hemodynamic consequences of the spontaneous death of the abnormal twin are the threats (Sebire et al., 1997; Ramos-Arroyo MA 1991). Therefore, early diagnosis of anencephaly is of essential in order to assure survival of the unaffected, normal twin.

In dichorionic twin pregnancies discordant for anencephaly, the general management options include; abortion of both fetuses, selective fetocide of the abnormal twin by intra – cardiac injection of potassium chloride, or continuation of the pregnancy without intervention (Sebire et al., 1997; Leeker and Beinder 2004). For monochorionic twin pregnancies discordant for anencephaly, selective reduction by cord occlusion has been suggested. Other options also include termination of pregnancy and expectant management (Sebire et al., 1997; Lim et al., 2005; Challis and Gratacos, 1991). Since this case is unusual, with no known published case in the Philippines, this paper presents our experience in managing a case of dichorionic diamniotic twin pregnancy with one twin discordant with anencephaly that was managed expectantly and the pregnancy reaches to term.

Case Report

This is a case of a 23 year old, G2P1 with twin pregnancy at 24 to 25 weeks gestation (based on last menstrual cycle)

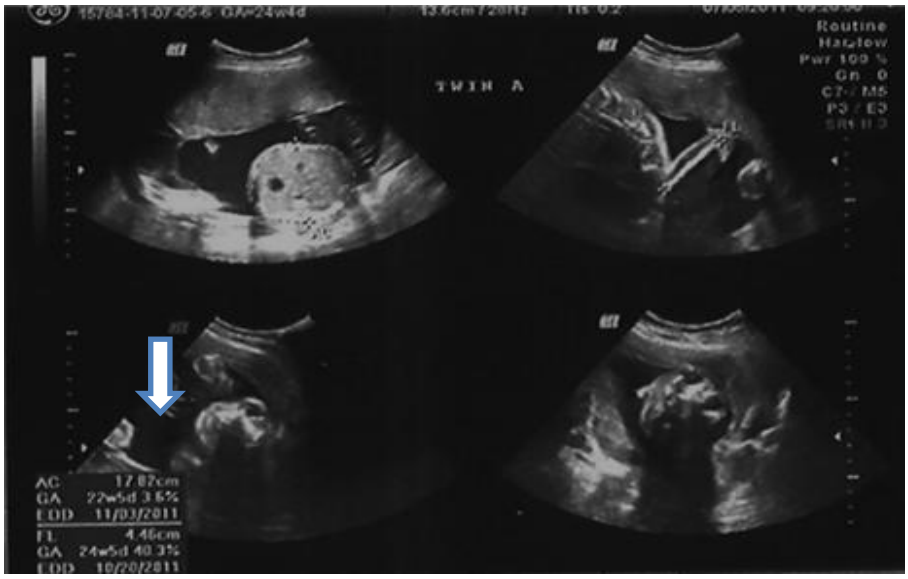

Figure 1. Sonographicgrayscale image of Twin A at 24 weeks shows absent upper head structure

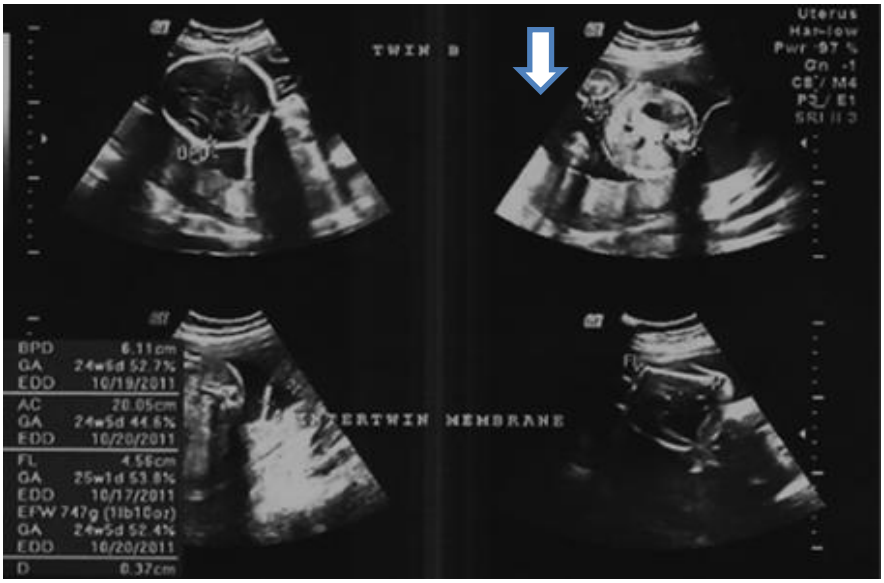

Figure 2. Sonographicgrayscale image of Twin B at 24 weeks shows normal upper head structure

presented to the Clinical Division of UST – Obstetric Department for her first pre – natal check – up. Upon history taking, she was known to have systemic lupus erythematosus since 9 years old and was maintained on corticosteroid therapy (prednisone). Her first pregnancy, ended up in preterm delivery at 24 weeks age of gestation by normal delivery in one of the hospitals in Quezon City, Philippines.

Ultrasound evaluation showed a live, diamniotic, dichorionic twin pregnancy at about 24 to 25 weeks of gestation, with good cardiac and somatic activity. In Twin A, scan result showed absent upper head structure, possibly presenting anencephaly (Figure 1). Twin B appeared to be healthy and no congenital anomaly seen at the time of scan (Figure 2). No additional findings were noted during the scan.

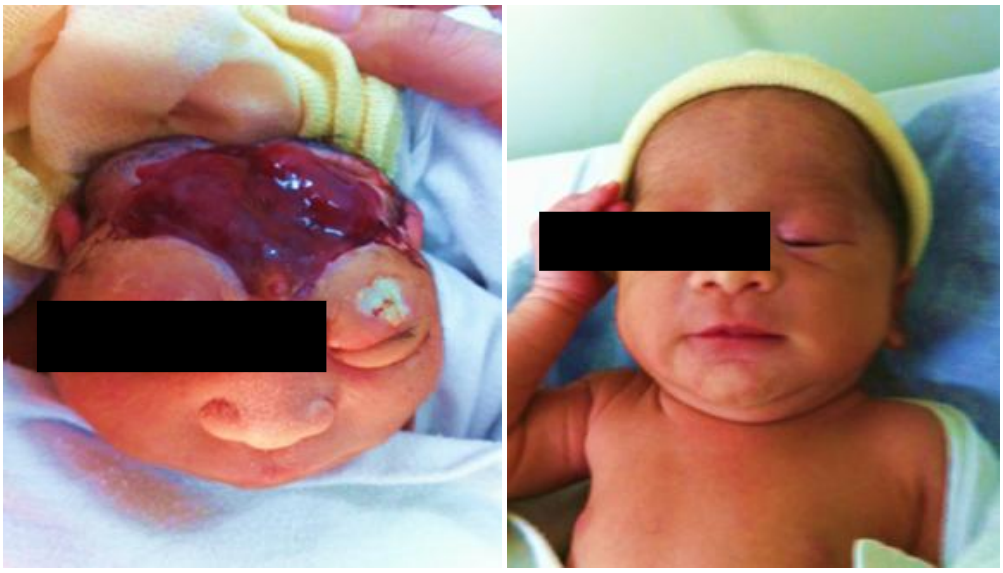

Figure 3. Postnatal image of Anencephalic and Normal Twins

The succeeding prenatal check – up was unremarkable. She was maintained on prednisone and was advised to take multivitamins and iron supplements. She was also co – managed with the rheumatology department of medicine. Ultrasound findings were the same on the follow up exam. Results still showed a growth discordancy.

Pregnancy continued to term at 37 to 38 weeks of gestation when she had premature rupture of membrane which prompted admission. On internal examination, the cervix was already open at 3 to 4 cm cervical dilatation. An emergency low transverse cesarean section was performed. At birth, twin B weighted 1860 grams, with a normal assessment, while twin A weighted 1420 grams, and anencephalic. Apgar score for twin B was 9, while the later was 6 (Figure 3). Twin A expired twenty one (21) hours after birth. Twin B was discharged together with the mother three (3) days without any complications.

DISCUSSION

Anencephaly has worldwide incidence and affects all races and ethnic groups. Its prevalence ranges from < 1/10,000 to 2.5/10,000 births and shows a female to male predominance of 3:1. (Lumenta et al., 2010; Mills et al., 2003).The prevalence of anencephaly in twins is higher than in singletons, and much higher in monochorionic than in dichorionic pregnancies (Sebire et al., 1997). Recent studies show that there is a higher rate for anencephaly among women who take insulin for diabetes, as well as those who take certain epilepsy medicines. Taking folic acids throughout the pregnancy is linked in preventing anencephaly, and those women who have anencephalic pregnancies who routinely takes folic acid decreases the risk of another anencephalic pregnancy (Mills et al., 2003; Hall and Solehdin 1998).

In cases of singleton pregnancies, having no chance to survive, management involves termination of pregnancy. However, in cases of twin pregnancies with one anencephalic fetus and one is normal, management remains vague. Normally, in dichorionic twins, serial ultrasound examinations for early diagnosis of polyhydramnios, which can be treated by either selective fetocide or amnio drainage are recommended (Vandecruys et al., 2006). Selective fetocide of anencephalic fetus before 15 weeks of gestation prevents the development of polyhydramnios and reduces prematurity, thus increasing survival rate of the health fetus (Leeker and Beinder 2004; Evans et al., 1999). However, in monochorionic twin pregnancies, selective fetocide is not possible because of the high risk of subsequent death of the normal co – twin (Sebire et al., 1997; Leeker and Beinder 2004). Selective reduction by cord occlusion has been an option in managing monoamniotic twins. Other options include termination of entire pregnancy, expectant management, use of non – steroidal anti – inflammatory drug, and selective termination by fetoscopic techniques (Sebire et al., 1997; Milner and Crombleholme 1999; Peek et al., 1997). In the presented case, fetocide was not performed due to refusal of the couple, thus expectant management was chosen.

In the previous years, retrospective and prospective studies showed that expectant management maybe one of the

best options for dichorionic twins discordant for anencephaly (Tasci et al., 2012; Langman et al., 2013). For example, in a retrospective study conducted in five Israeli Perinatal Center, fourteen (14) cases were managed expectantly and none miscarried. Meanwhile, Lust, noted a higher survival rate in dichorionic group treated and managed expectantly (Lust et al., 2008). In a retrospective study conducted by Sebire et al., the authors remarked that expectant management may not warrant survival of the normal twin in cases of monochorionic twin pregnancies discordant of anencephaly, since this option can result in the death of the normal twin. However, in much recent study and literature review of Lim et al., suggest that expectant management is a reasonable option that may result in successful outcomes (Lim et al., 2005). In the case presented, expectant management of a twin gestation discordant for anencephaly yield positive outcomes. The pregnancy reached its term and the normal, unaffected twin survived without any complications.

Anencephaly is a lethal diagnosis and may have serious consequences. However, this case is frequently detected with prenatal ultrasound at a routine 10 to 14 week sonogram (Johnson et al.,1997; Souka and Nicolaides 1997).Therefore, early ultrasound scanning as part of routine antenatal care can facilitate early detection and management for the normal, unaffected twin.

Conflict of Interest

Authors declare no conflict of interest.

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