

LESSONS FROM A CASE OF TUBAL TWIN PREGNANCY



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Unilateral tubal twin pregnancy illustrates and amplifies fundamental phenomena in developmental and reproductive anatomy. Knowledge of this condition is also important to practicing obstetricians and gynaecologists because it may constitute a diagnostic challenge, management dilemma, complex ethical issues and increased risk for maternal morbidity and mortality (Benn et al., 2016). Previously considered a rare occurrence, recent data suggest that the condition is not uncommon (Svirsky et al., 2010). Indeed, there are many reports (Vohra et al., 2014), including several from Sub-Saharan Africa (Makinde and Ogunniyi, 1990). The case reported by Pulei et al., in *Anat J Afr* 2017; 6 (1) reveals several unique features in the profile of risk factors, location, chorionicity and amnionicity, mode and time of presentation, condition of the tube, diagnosis and fetal viability. There was no evidence of the conventional risk factors (Sivalingam et al., 2011). This is consistent with several other reported cases where it occurred spontaneously (Abi Khalil et al., 2016). In the present case, however, the patient had multiple intramural and subserosal leiomyomata. Intramural myomata are known to disrupt uterine contractility which may interfere with transport of the ovum and hence predispose to ectopic pregnancy (Ajibade et al., 2012). It is probable, therefore, that this was the predisposing factor. Accordingly, it may be useful to monitor patients with uterine fibroids for potential to suffer ectopic pregnancy. Family history of twinning may have been useful, and is advocated, in view of the fact that it is one of the major predisposing factors.

In the case presented, the gestational sac was located in the fimbrial region, similar to that reported by Al – Quraan et al. (2007) and Adesule et al. (2010). The commonest site of tubal pregnancy is the ampulla followed by isthmus, with the fimbria being involved in only 5 – 11 % of cases (Almeida, 2011). The location of the present case in the relatively unusual position is concordant with reports of twin pregnancy in rarer tubal sites like cornual (Ragsdale et al., 2014) and in other ectopic positions including the cervix (Pascual et al., 2001), ovary (Garg et al., 2009) and abdomen (Mpogoro et al.,

2013). This calls for thorough examination of the abdomen and pelvis when scanning for ectopic pregnancy.

The fetuses shared a placenta but were in separate amniotic sacs. This condition is referred to as monochorionic diamniotic twins. It arises when the blastocyst divides between the 3rd and 8th gestational days and is the most common type of monozygotic twins (Fox, 2006). Cases similar to the present one have been described in tubal twin pregnancy (Erol et al., 2013). These twins have higher rates of fetal loss and growth discordance than dichorionic ones because of often precarious vascular anastomoses and unequal distribution of placental utility (Mascarenhas and Ghanaprabha, 2016). The death of one twin is consistent with this disadvantage. Accordingly, it is important to determine the amnionicity and chorionicity of the fetus to optimize care.

The most common presentation is abdominal pain with vaginal bleeding (Ghanbarzadeh et al., 2015). The present case was atypical in that there was no vaginal bleeding. Similar atypical cases have been reported (Indiran, 2016). Such atypical presentation may mimic other causes of acute abdomen, confuse diagnosis, delay treatment and lead to high morbidity and mortality (Sivalingam et al., 2011). Accordingly, twin tubal pregnancy should be considered a differential diagnosis in cases of persistent lower abdominal pain in amenorrhic women, even in the absence of vaginal bleeding. Most cases of ectopic pregnancy present in the first trimester, usually between the 5th and 10th weeks (Ghanbarzadeh et al., 2015), with only a few reaching the 11th week (Panelli et al., 2015). The current case presented at almost 13 weeks, although the pain had started at about the 8th week. Early presentation and diagnosis reduces the risk of rupture and maternal morbidity and mortality (Ghanbarzadeh et al., 2015).

The amazing observation in the present case is that despite the advanced gestation, the tube was not ruptured, only displaying slight leakage. Since the first report of unruptured twin tubal pregnancy by Santos et al (1986), there have been other reports in literature (Goswami et al., 2014). Tubal rupture is usually attributed to hemorrhagic necrosis consequent to trophoblastic invasion (Stock et al., 1991). The absence of overt rupture suggests that other factors are involved, including probably the location of the gestational sac. A pertinent observation in this case is that both twins were in the fimbrial region, which is relatively wide and may therefore accommodate a bigger conceptus before attaining the threshold for rupture. Identification of unruptured ectopic pregnancy is important as it provides an opportunity for tube preserving intervention (Goswami et al., 2014). Accordingly, whenever possible, effort should be made to diagnose ectopic pregnancy before tubal rupture.

Tubal twins are usually diagnosed by transvaginal ultrasound, markedly elevated hCG, laparoscopy (Ghanbarzadeh et al., 2015) and magnetic resonance imaging (Indiran, 2016). In the present case, one sac was diagnosed preoperatively by abdominal ultrasound while the second twin was diagnosed intraoperatively, and hCG levels were not determined. It is important that all the gestational sacs are identified and managed to minimize complications of inadvertent retention of ectopic products of conception. Another intriguing feature of the present case was that one fetus was viable. Since the first case of live unilateral tubal twin pregnancy by Gualandi et al (1994), several other cases have been described (Benn et al., 2016). This is important as it may influence the options of management – surgical, medical or expectant (Sivalingam et al., 2011).

In conclusion, tubal twin ectopic pregnancy may occur in the absence of any overt risk factor, be located anywhere along the tube, show atypical late presentation, intact tubes and viable fetus. These features call for heightened index of suspicion and sagacity in management of amenorrhic women with abdominal pain.

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