The Case of Monochorionic Twin Gestation Complicated by Intrauterine Demise of One Fetus in the First Trimester

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he study investigated the clinical and pathomorphological characteristics of monochorionic diamniotic twin gestations complicated by the spontaneous loss of one fetus during the first trimester. Nine monochorionic diamniotic twin gestations were analyzed in which the demise of one fetus occurred during the first trimester. During the course of the study, 178 twin gestations were sonographically identified during the first trimester. Forty-three (24.2%) were complicated by the intrauterine demise of one fetus before the end of the 12th week of pregnancy. Nine cases were monochorionic diamniotic. The gestational age of the demised fetuses ranged between 5 and 11 weeks (mean 7.4 weeks). The prognoses for the surviving fetuses were rather poor with the surviving co-twin dying 1 to 3 weeks after the first twin, with abortion of both fetuses. In only one case was gestation concluded on the 40th week with delivery of a live neonate. No blood coagulation changes were observed in the affected pregnant women, and the one live newborn did not reveal any haematological or neurological abnormalities. A postnatal study provided evidence for the history of monochorionic diamniotic placentation in all of the cases. In two cases, probable causes of fetal death were established (congenital malformation, the presence of anastomoses between blood vessels). The obtained results suggest an extremely high risk of mortality for the surviving twin fetus following the co-twin's death in cases of monochorial placentation. The morphological evaluation of placentas and fetal membranes enables the establishment of the type of placenta and, in certain cases, the probable cause of the intrauterine demise of one twin fetus during the first trimester of monochorionic diamniotic twin gestation.

Twin gestation does not always result in the delivery of all the primarily conceived embryos as the intrauterine death of one or more fetuses may occur during the course of gestation. Such a death is regarded as an exceptional type of prenatal mortality. In such cases, besides the care of the pregnant woman, significant attention is focused on the further development of the surviving fetus remaining in the uterine cavity. Fetal death may occur at any timepoint of gestation, but it occurs most often during the first trimester. Out of all twin pregnancies diagnosed by ultrasound imaging before the 10th week, 21% to 75.9% result in the delivery of a single neonate (Benson et al., 1993; Blumenfeld et al., 1992; Legro et al., 1995).

There is a growing interest in such twin gestation complications. In the majority of cases, however, the authors concentrate on intrauterine fetal deaths which occur during the second and the third trimesters. Publications concerning the first trimester are rather scarce (Blumenfeld et al., 1992; Landy & Keith, 1998; Newton et al., 2003; Rudnicki et al., 1991), while reported evaluations covered all twin pregnancies in general without specifying monochorionic or dichorionic. Moreover, in most reported cases there have been no reports on either the postnatal placentation or the vanished intrauterine embryo or fetus.

This study evaluates the course of pregnancy and the development of the surviving fetus after the intrauterine demise of one fetus during the first trimester of a monochorionic diamniotic twin pregnancy. A detailed analysis of results obtained in the pathomorphological study of the dead embryo/fetus and placenta are also presented.

Materials and Methods

One hundred and seventy-eight twin pregnancies between 5 and 12 weeks' gestation were referred for ultrasonography to the Ultrasound Consulting Room at the Obstetric-Gynaecological Department of the Hospital of the Public Health Care Trust in Kutno between the years 1990 and 2003. Out of those preg-

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nancies, 43 (24.1%) were complicated by 'vanishing twin' syndrome before the 12th week of pregnancy. All pregnancies were scanned by the first author (Prof. W. Malinowski). The sonographic examinations were performed at first with a Hitachi EUB 40 device with 3.5 and 5.0 MHz convex probes, and then with a B&K Medical System 3535 device with a 6.5 MHz intravaginal probe.

The sonographic diagnostic criteria for monochorionic diamniotic pregnancy included a single disc of the placenta, a 'one-ring sign', yolk sacs not separated by septum, 'tau' signs (the placental site where the double membranes join is flat) and a thin (less than 2mm), two layer septum (Malinowski, 2001). Pregnancies regarded as twin pregnancies with the intrauterine death of one fetus included those with the coexistence of a live fetus with a dead fetus (no heart function), abnormal size or appearance of one of the yolk sacs (observed in the very early period of gestation) and the 'empty' gestational sac (in a later period of pregnancy) found in sonographic imaging (Malinowski, 1998). The assumed time of fetal death was assessed on the basis of the crown-rump length (CRL) or the size of the gestational sac (GS).

All pregnant women with multiple pregnancies complicated by the intrauterine demise of one fetus received extended prenatal care which included control examinations at an obstetric outpatient clinic for multiple pregnancy and sonographic examinations (every 1 to 2 weeks), as well as measurements of the blood serum concentrations of clotting factors (fibrinogen, platelets, APTF, FDP) repeated at 2- and then 4-week intervals.

Evaluations of live neonates included perinatal status in the first and fifth minute after birth (Apgar score), birth weight and birth length. Also transfontanellal sonographic imaging, blood group and platelet counts were performed together with acidbase balance, glucose and bilirubin concentrations and platelet numbers. The diagnostic series was completed by neurological examinations.

Macroscopic pathomorphological examinations of the placenta included searching within the placental tissue and fetal membranes for the presence of a gestational sac with possible vanished fetus remnants. All identified tissues were submitted for microscope analysis. The postnatal determination of the type of gestational chorionicity was obtained by the evaluation of the membranes surrounding the demised fetus.

Results

Out of 43 multiple pregnancies complicated by the intrauterine death of one embryo/fetus in the first trimester, nine were monochorionic diamniotic twin gestations (MC DA). The period of gestation in which the demise of one of the fetuses occurred was probably between the 5th and the 11th week (mean 7.4 weeks). The time interval between the fetal death and abortion or delivery was between 1 and 29 weeks.

Monochorionic Twin Gestation Complicated by Intrauterine Demise

In seven MC twin pregnancies, the intrauterine death of one fetus occurred between the fifth and the eighth week of gestation. In the eighth case, the death took place on the 10th week and in the ninth case on the 11th week of pregnancy. The prognoses for surviving fetuses were poor. In eight cases, the co-twin died 1 to 3 weeks after the death of the first twin. In only one case did the pregnancy result in the delivery of a live neonate at 40 weeks. A male neonate was born in a good condition (Apgar score: 10 in 5th minute) by spontaneous delivery without congenital malformations. Sonographic transfontanellal imaging, laboratory tests and neurological examinations did not reveal any deviation from normal values. The child remains under strict paediatric and neurological follow-up.

In women with monochorionic diamniotic twin pregnancies complicated by the intrauterine demise of one fetus in the first trimester, bleeding from the reproductive tract occurred in seven out of nine cases at such a time that may have coincided with the time of intrauterine fetal death. No blood coagulation disorders or any other type of complications were found in any of the affected women either during the course of pregnancy or after abortion or delivery.

Pathomorphological examination of demised fetuses and placentas were performed in nine of the cases. The presence of a monochorionic diamniotic pregnancy was confirmed in all cases. In seven cases, the attempt to unequivocally determine the cause of intrauterine fetal death ended with diagnostic failure. Regarding the other two cases, oedema was found in the nuchal region of the demised fetus in one case (death occurred at the 10th week of pregnancy approximately) which could be suggestive of congenital malformation (Figure 1). In the second case, the fetal death took place at the 11th week and the pregnancy resulted in the delivery of the remaining live twin at the 40th week. Detailed macroscopic evaluation revealed a placenta of 22.0×19.00 cm in size and 3.0cm thick with a central attachment of the umbilical

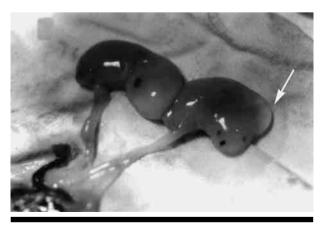


Figure 1

Oedema in the nuchal region of an intrauterinally demised fetus in the 10th week of monochorionic twin pregnancy.

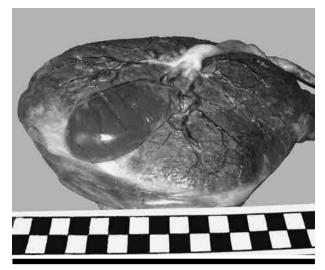


Figure 2

Placenta with gestational sac containing a fetus demised in the 11th week of monochorionic twin pregnancy.

cord. The umbilical cord was 65.0cm long and 1.5cm in diameter. Microscopic examination of the umbilical cord demonstrated the presence of three vessels. Lumen obliteration was found in one of the umbilical arteries. A sac-like formation was found on the fetal surface of the placenta $7.0 \times 5.0 \times 2.5$ cm in size containing a small volume of brown fluid and a macerated fetus. The long axis of the fetus was 2.5cm and the short axis 1.5cm (Figure 2 and Figure 3). The necropsy of the demised fetus revealed the presence of macerated homogenous masses, anatomical structures were impossible to identify. Two blood vessels, each 0.2cm in diameter, were visible on the chorionic surface of the placenta ending at the base of the sac with the demised twin (Figure 2 and Figure 3). This may indicate the presence of direct vascular (arterio-arterial and veno-venous) anastomoses between the two fetuses. Detailed macroscopic and microscopic analyses demonstrated the presence of a

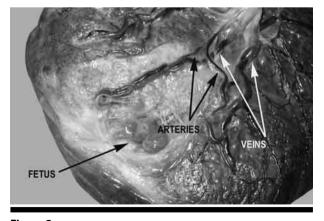


Figure 3 Macerated fetus and direct arterio–arterial and veno–venous anastomoses.

two-layer sac wall (monochorionic diamniotic pregnancy) and a 'blind' umbilical cord. No pathological changes were found within the fetal membranes. Microscopic evaluation of the placental plate revealed numerous, multifocal infarcts, the largest cluster of which was close to the previously mentioned sac-like formation. Moreover, focal oedema and placental chorionic villa degeneration features were found.

Discussion

A precise assessment of the incidence of the intrauterine demise of one fetus during the first trimester of twin pregnancy is rather difficult as the diagnoses of these events is affected to a large extent by the type of sonographic equipment, the gestational age of death, during which pregnancy week the first sonographic examination was performed and lastly, the size of the population examined. Moreover, postnatal examinations of placentas from surviving fetuses usually demonstrate no macroscopically visible remnants of the atrophied twin.

It has been estimated that the incidence of twin pregnancies at conception may amount to 1:8 pregnancies. This would mean 1 pair of live twins delivered per 6 newborns from single pregnancies, which in fact are single surviving fetuses from primarily twin pregnancies (Boklage, 1995). Blumenfeld et al. (1992) sonographically examined a group of 5000 pregnant women during the first trimester and diagnosed 116 multiple pregnancies. Out of the whole group, 88 (75.9%) were complicated by a 'vanishing twin' syndrome.

In our material, the spontaneous loss of one embryo or fetus during the first trimester of a twin pregnancy was observed in 24.1% of cases. This percentage may indeed be higher, as only 30% of the entire group of pregnant women were submitted to sonographic examination at an early stage of pregnancy.

An episode of mild vaginal bleeding, which was observed in between 25% and 76.5% of all cases (Landy et al., 1986; Yoshida, 1995), may be a clinical symptom of the intrauterine demise of one fetus during the first trimester of twin pregnancy. In the present study of nine monochorionic pregnancies, bleeding of various intensity from the uterine cavity was observed in the majority of cases (seven of nine).

At present, diagnosis of the spontaneous loss of one fetus during the first trimester of twin pregnancy is possible only via ultrasound imaging. Oddly enough, in the case of the intrauterine demise of one fetus in the first trimester of twin pregnancy, the dead fetus undergoes slow resorption ('vanishing twin' syndrome), while the amniotic fluid retains its volume until late pregnancy. The etiology of this phenomenon is yet to be explained.

In addition, in one of the cases, the gestation sac of the atrophied fetus was visible in the ultrasound examination until the end of the pregnancy at the 40th week. This facilitated identification of the sac in the postnatal period as well as thorough examination.

It is still unclear whether the intrauterine demise of one twin during the first trimester has a negative influence on the further development of the surviving fetus. Boklage (1995) claims that the majority of newborns from single fetal pregnancies delivered with congenital malformations, may, in fact, have come from primarily multiple pregnancies. This may also apply to left-handed individuals, the highest percentage of whom are found among twins. This have not been confirmed by results from other studies (Blumenfeld et al., 1992; Yoshida & Matayoshi, 1990), as no increased risk of congenital malformations has been found in the surviving fetus following the intrauterine demise of the other twin. However, these observations refer exclusively to dichorionic twin pregnancies. Only in one of the rarely reported cases of monochorionic diamniotic twin pregnancies complicated by the intrauterine death of one fetus were congenital malformations observed after delivery (including atrophy of brachial muscles atresia of the jejunum and of the transverse colon; Baldwin, 1994). It was suggested that the presence of anastomoses between the twin fetuses may have resulted in haemodynamic disturbances of circulation in both fetuses, which led to the death of one fetus and congenital malformations in the other. Such a possibility can not be excluded in our case either, as in the majority of cases observed, the other twin died soon after the death of the first one.

Data from literature (Anderson et al., 1990; Blickstein, 1990; Boklage, 1995; Eglowstein & D'Alton, 1993; Petersen & Nyholm, 1999; Prompeler et al., 1994) demonstrate that the intrauterine demise of one fetus in the first trimester of twin pregnancy does not create any significant risks to the life or health of the mother. Neither was the development of any blood coagulation disorders observed in any of the mothers in the study group. However, the observations comprised only a small population and the time period between fetal death and pregnancy termination was relatively short (less than 3 weeks) in all but one case. Therefore, it seems both appropriate and reasonable to provide enhanced obstetric care to such patients.

In most cases, it is not possible to determine the unequivocal causes of the intrauterine twin demise in the first trimester. Anderson et al. (1990) found fetal growth disturbances to be the etiological factor in 78% and congenital malformations in 22%, the latter mostly resulting from chromosomal errors. Also indicative was the considerable prevalence of female fetuses and monochorionic pregnancies (Benson et al., 1993).

In this study, one of the observed fetuses demonstrated a distinct oedema in the nuchal region. This observation may be suggestive of the existence of serious congenital malformations already occurring in the division process of the primarily single zygote. In the second case, the presence of superficial direct arterio-arterial and veno-venous anastomoses may indicate acute 'twin-to-twin transfusion' syndrome or 'twin-reversed arterial perfusion' syndrome as probable causes of fetal death.

Regardless of the increasingly frequent sonographic diagnoses of the intrauterine demise of one fetus, its occurrence during the first trimester remains the least recognised type of fetal mortality in multiple pregnancies. There are associated difficulties in confirming early losses in postnatal examinations, as, usually, very little is left of the demised twin in the placenta of the surviving fetus delivered many weeks later. Nevertheless, the determination of the type of twin pregnancy and the diagnosis of cause of fetal death is important. Unquestionably, pathologists play the key role in this diagnostic process with thorough and precise pathological study enabling the diagnosis in many cases regarded previously as impossible. Moreover, such an unequivocal diagnosis of a monochorionic twin pregnancy provides an opportunity for the diagnosis and the prognosis of the live delivered twin and, in the case of abnormal development, the opportunity to reassure parents that no similar malformations are expected in subsequent pregnancies.

In all cases in this study, detailed pathomorphological study performed in the postnatal period provided unequivocal evidence of monochorionic diamniotic twin pregnancies, while in two cases, it allowed definition of the probable cause of fetal death.

Conclusions to be drawn from the study are: (1) The intrauterine demise of one fetus in the first trimester of a monochorionic diamniotic twin pregnancy does not create any health risks for the pregnant woman; (2) the prognoses for the surviving fetuses in monochorionic diamniotic twin pregnancies are extremely unfavourable; and (3) postnatal pathomorphological study provides information on the type of twin pregnancy and, in some cases, on the assumed cause of fetal death.

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