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CASE REPORT

# Post-zygotic diploidization of triploidy in human is possible? – a case of triploid partial molar pregnancy resulting in a premature live-born diploid female infant

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## Abstract

**OBJECTIVE:** During the treatment of our patient we found that reports covering possible complications and their treatment are very scarce. Due to advancement in ultrasound diagnosis most of molar pregnancies are terminated in first trimester of pregnancy. There is the gap in knowledge concerning pregnancy complications in case of partial mole discovered in advanced pregnancy. This is why we incorporated extensive and up-to-date review of literature in our manuscript.
**METHOD:** We described a case of previously healthy, 25 year old primigravida who delivered live daughter at 27 weeks of gestation, complicated with unusual ultrasound appearance of the placenta, severe hypotrophy, and subsequent postpartum eclampsia.
**RESULTS:** Healthy diploid female infant, now two years old and healthy mother taking care of her.
**CONCLUSIONS:** In clinical practice early diagnosis of this complication usually lead to pregnancy termination. In modern medicine, decisions should be based on evidence and patient-doctor mutual understanding. Termination of pregnancy with suspicion of molar placenta can be specially difficult in gestation in older nulliparous women or after ART. We sincerely hope that this report will be useful for physicians across the world in counseling and treating their patients.

## MANUSCRIPT

Molar pregnancy was first systematically described by Vassilakos and Kajiiin 1976 (Vassilakos & Kajii 1976). Hydatidiform moles are categorized into partial or complete mole due to the cytogenetic, histological and clinical differences. Acomplete molar pregnancy occurs when an ovum not carrying any chromosomal genetic material is fertilized by either a haploid sperm, followed by the duplication of the chromosomes, or two different sperms. Then, the conceptus is diploid and contains exclusively paternally derived chromosomes. A partial mole is defined as excessive trophoblast proliferation with both components of normal and hydropic villi and a presence of a fetus with nucleated cells in its circulation. Its incidence is reported between 0.005 and 0.01% of all pregnancies (Teng & Ballon 1984). However a geographic variation of occurrence is observed: moles occur more often in Asia, Africa and Latin America, while in Europe, Australia and USA its rates of incidence are lower (Bracken 1987). Cytogenetic studies have shown that over 90% of all partial moles are associated with triploidy and malformed and nonviable fetus (Jauniaux 1999). Pregnancies with normal diploid fetus and triploid partial mole are very rare.We present a case of partial mole pregnancy coexisting with a live diploid fetus, complicated by fetal growth restriction and eclampsia.

## CASE REPORT

25 years old woman, gravida 2, para 1, was admitted to the 1st Department of Obstetrics and Gynecology, Medical University of Warsaw, at 25 weeks of pregnancy due to fetal intrauterine growth restriction (IUGR) and abnormal appearance of the placenta. Her family and past medical history were unremarkable. During the first trimester ultrasound scan no fetal abnormalities were detected. The risk of trisomy was calculated solely on ultrasound markers and no biochemical markers were assessed.The next ultrasound scan at 19 weeks revealed a single female fetus with no anatomical anomalies, however abnormal, thickened "jelly like placenta" with numerous cysts with diameter up to 2 cm was observed [Figure 1]. At 25 weeks IUGR with abnormal Doppler was diagnosed (estimated fetal weight 650g – 5th percentile, umbilical artery pulsatility index 1.8 - >90 percentile). Patient was admitted to hospital. During observation patient's blood pressure was normal. As deterioration in Doppler studies was observed (absent end-diastolic flow in umbilical artery) intramuscular betamethasone was administered and patient was carefully monitored.

At 27 weeks a pathological cardiotocograph tracing revealed an imminent fetal asphyxia (reduced variability with repetitive prolonged decelerations) and female newborn weighting 740 grams(1 percentile) was delivered via emergencycesarean section. The child was admitted to Neonatal Intensive Care Unit. She required mechanical high-frequency ventilation and administration of two doses of surfactant due to respiratory distress syndrome and catecholamines infusion due to circulatory insufficiency. She received a blood transfusion due to severe anemia (HGB 9.6 G/L) and steroid treatment due to bronchopulmonary dysplasia. Further improvement of cardiac and respiratory functions were observed and the child was discharged after 77 days of hospitalization. At the age of 18 months the infant is phenotypically and developmentally normal.

As for the mother, the early postoperative period was complicated by hypertension of 200/100 mmHg and two eclamptic seizures attacks. The patient received hypotensive and anticonvulsive treatment (urapidyl and dihydrazaline) and magnesium sulfate infusion. She was transferred to Neurological Intensive Care Unit, where she was diagnosed with posterior

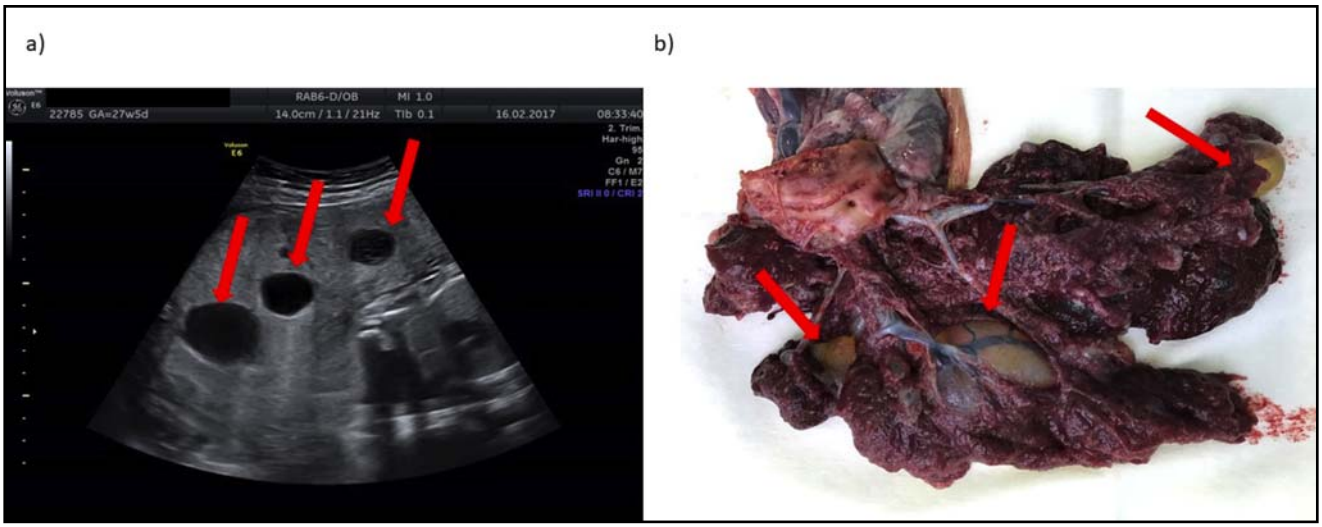


**Fig. 1.** a) Numerous cysts with diameter up to 2 cm observed by US at 19 weeks (red arrows). b) Multiple grape-like fluid filled cysts and cisterns up to 4.0 cm in diameter interspersed among normal-appearing villous tissue of placenta (red arrows)


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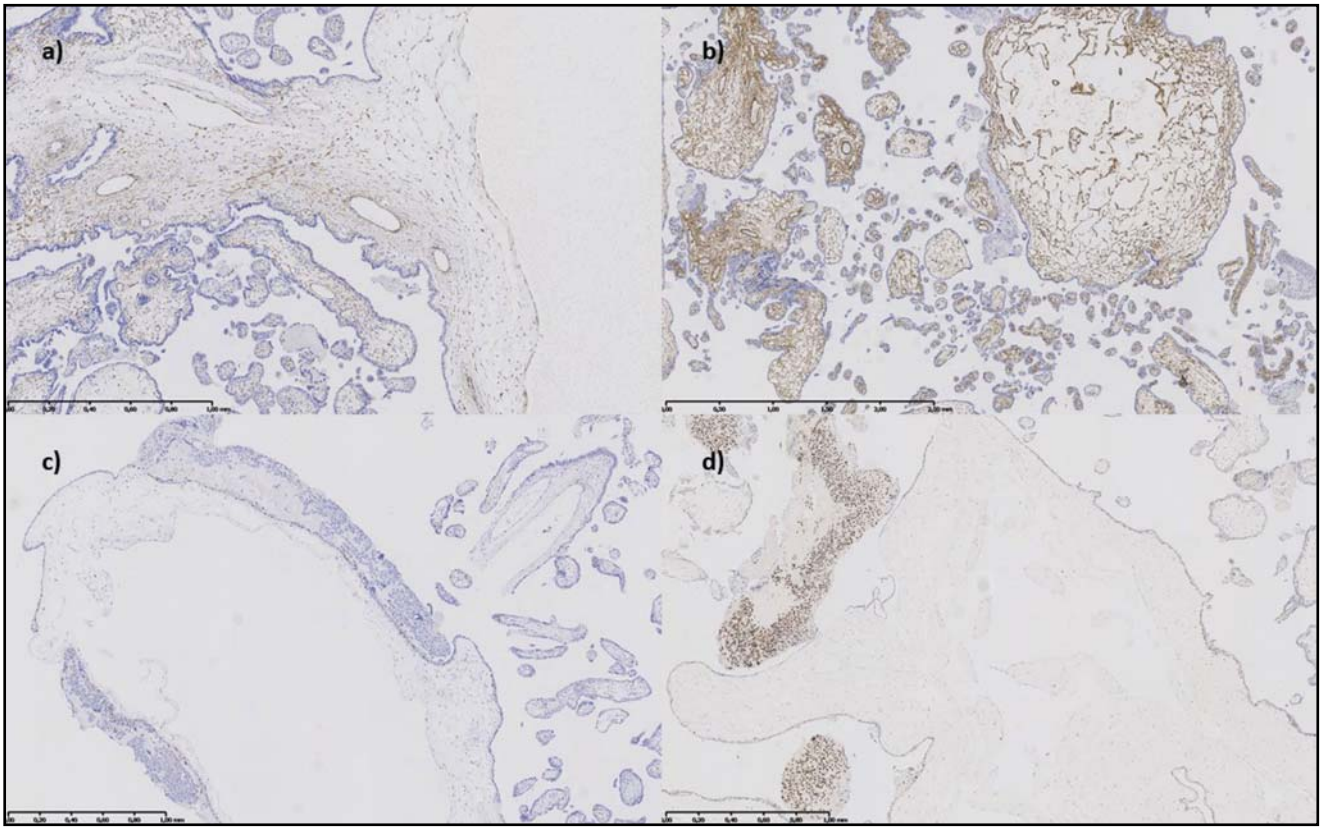


**Fig. 2.** Histopathologic findings compatible with a partial molar pregnancy. Immunohistochemical staining demonstrated that desmin (a) and vimentyn were strongly expressed in stromal cells of normal and dysmorphic villi. Smooth Muscle Actin's expression in stroma of normal villi was high but in dysmorphic villi was low (b). There was low detectable Ki67 expression in stromal cells and villous trophoblast, only slightly higher in focal trophoblastic proliferation (c).Nuclear expression p57 immunostaining in villous stromal cells and cytotrophoblast was focally positive (d).

reversible encephalopathy syndrome (PRES)(Garg, Kumar *et al.* 2018). Treatment with dexamethasone and mannitol was successful with normalization of the blood pressure. Maternal postpartum serum βHCG concentration was not elevated (5 mIU/mL, <0.1 mIU/mL 2 weeks later). She was discharged home 3 weeks after delivery.

The placenta weighted 630g, which was above the 97$^{th}$ percentile for gestational age. Distended/dilated tortuous vessels were present on chorionic plate. The umbilical cord with three vessels was eccentrically inserted close to the border of chorionic plate. The maternal plate and cut section of the placenta showed multiple grape-likefluid filled cysts and cisterns up to 4.0 cm in diameter interspersed among normal-appearing villous tissue [Figure 1].

Representative samples/sections ofumbilical cord, membranes and placental tissue were histologically processed to the microscopic slides. Microscopic examination revealed an admixture of normal terminal and steam villi, enlarged dysmorphic villi with hydropic changeand cystic edematous steam villi. Dysmorphic immature villi with irregular villous contour shoved focal aberrant vascular pattern, mild trophoblast proliferation and trophoblast inclusions.

Microscopically, proximal enlarged steam villi demonstrated hydropic changes with cistern formation. These villi were characterized by loose, myxoid, edematous stroma with fewer and smaller thick-walled vessels then their normal counterparts. Immunohistochemical examination demonstrated that desmin and vimentyn were strongly expressed in stromal cells of normal and dysmorphic villi. Smooth Muscle Actin's expression in stroma of normal villi was high but in dysmorphic villi was low, patchy. There was low detectable Ki67 expression in stromal cells and villous trophoblast, only slightly higher in focal trophoblastic proliferation.

Nuclear expression of p57 immunostaining in villous stromal cells and cytotrophoblast was focally positive [Figure 2]. The histopathologic findings were compatible with a partial molar pregnancy.

The placenta karyotype on cultured cells revealed a 69,XXY karyotype in all cells examined, unfortunately no interphase FISH studies on the uncultured cells were performed. The newborn karyotype (cultured blood lymphocytes) was diploid (46, XX).

In order to determine the parental origin of the extra haploid set of chromosomes STR (shorttandem repeats) analysis was performed on DNA samples from placenta, both parents and a child.The results wereconsistent

**Tab. 1.** Microsatellite results of DNA sampling

| Locus | father | placenta | child | mother | Sperm 1 | Sperm 2 | Oocyte |
|---|---|---|---|---|---|---|---|
| AMEL | X ; Y | X; X ; Y | X ; X | X ; X | Y | X | X |
| D3S1358 | 16 ; 17 | 16 ; 17; 17 | 16 ; 17 | 15 ; 17 | 17 | 16 | 17 |
| D1S1656 | 13 ; 15 | 15 ; 15; 15 | 15 ; 15 | 14 ; 15 | 15 | 15 | 15 |
| D2S441 | 11 ; 14 | 11 ; 11 ; 14 | 11 ; 14 | 10 ; 14 | 11 | 11 | 14 |
| D10S1248 | 15 ; 17 | 16 ; 17 ; 17 | 16 ; 17 | 13 ; 16 | 17 | 17 | 16 |
| D13S317 | 8 ; 12 | 8 ; 9 ; 12 | 9 ; 12 | 9 ; 9 | 8 | 12 | 9 |
| PENTA E | 10 ; 12 | 10 ; 12; 12 | 10 ; 12 | 12 ; 18 | 12 | 10 | 12 |
| D16S539 | 11 ; 13 | 12 ; 13 ; 13 | 12 ; 13 | 12 ; 12 | 13 | 13 | 12 |
| D18S51 | 13 ; 15 | 12 ; 13 ; 15 | 12 ; 13 | 12 ; 14 | 15 | 13 | 12 |
| D2S1338 | 22 ; 25 | 17 ; 22 ; 22 | 17 ; 22 | 17 ; 20 | 22 | 22 | 17 |
| CSF1PO | 11 ; 15 | 10 ; 15 ; 15 | 10 ; 15 | 10 ; 11 | 15 | 15 | 10 |
| PENTA D | 10 ; 10 | 9 ; 10 ; 10 | 9 ; 10 | 9 ; 9 | 10 | 10 | 9 |
| TH01 | 8 ; 9 | 6 ; 8 ; 9 | 6 ; 8 | 6 ; 7 | 9 | 8 | 6 |
| vWA | 14 ; 18 | 14 ; 17 ; 18 | 14 ; 17 | 14 ; 17 | 18 | 14 | 17 |
| D21S11 | 29 ; 30 | 29 ; 30 ; 30 | 29 ; 30 | 29 ; 29 | 30 | 30 | 29 |
| D7S820 | 8 ; 8 | 8 ; 8 ; 12 | 8 ; 12 | 12 ; 12 | 8 | 8 | 12 |
| D5S818 | 12 ; 12 | 11 ; 12 ; 12 | 11 ; 12 | 11 ; 12 | 12 | 12 | 11 |
| TPOX | 8 ; 9 | 8 ; 8 ; 9 | 8 ; 9 | 8 ; 11 | 8 | 9 | 8 |
| D8S1179 | 9 ; 10 | 9 ; 10 ; 10 | 9 ; 10 | 10 ; 15 | 10 | 9 | 10 |
| D12S391 | 18 ; 19 | 18 ; 19 ; 20 | 18 ; 20 | 20 ; 23 | 19 | 18 | 20 |
| D19S433 | 14 ; 15.2 | 14 ; 14 ; 14 | 14 ; 14 | 14 ; 14 | 14 | 14 | 14 |
| SE33 | 13.2 ; 34.2 | 11.2 ; 13.2 ; 34.2 | 11.2 ; 34.2 | 11.2 ; 20 | 13.2 | 34.2 | 11.2 |
| D22S1045 | 11 ; 11 | 11 ; 11 ; 15 | 11 ; 15 | 15 ; 15 | 11 | 11 | 15 |
| DYS391 | 10 | 10 | - | - | 10 | - | - |
| FGA | 19 ; 23 | 19 ; 21 ; 23 | 19 ; 21 | 21 ; 22 | 23 | 19 | 21 |
| DYS576 | 19 | 19 | - | - | 19 | - | - |
| DYS570 | 19 | 19 | - | - | 19 | - | - |

with diandric triploidy (due to dispermy) in placenta andbiparental diploidy in the child. Table 1 presents the microsatellite results.

## DISCUSSION

We have described a unique case of triploid partial molar pregnancy resultingina diploid normal live-born femaleinfant. Pregnancies with hydatiform mole and a viable fetus are very rare, occurring in 1 in 20,000 to 1 in 100,000 gestations (Sebire *et al.* 2002). However,fetuses in those cases have usually chromosomal abnormalities. It is even rarer for a newborn to be born with a normal diploid karyotype from a molar pregnancy, although there are reports of live, term deliveries of diploid fetuses with partial mole in primarily diploid placenta (Sak *et al.* 2012). Up to date 6 cases of triploid partial mole with diploid fetus were described (Sarno *et al.* 1993; Zhang *et al.* 2000; Yoneda *et al.* 2013; Kawasaki *et al.* 2016; Booth & Eskandar 2018; Loza and Victor Fang 2018). In 2 of them therapeutic termination of pregnancy was performed and one case was complicated by intrauterine fetal demise at 20 weeks of pregnancy (Zhang *et al.* 2000; Yoneda *et al.* 2013; Booth & Eskandar 2018). Only three viable diploid newborns born form a triploid partial molar pregnancy were reported (Sarno *et al.* 1993; Kawasaki *et al.* 2016; Loza & Victor Fang 2018). The first one was described in 1993 by Sarno *et al.* A 32-year-old women, diagnosed with a partial molar pregnancy, delivered at 33 weeks of gestation a healthy, female infant, weighting 1935 grams. The neonatal peripheral blood karyotype was 46,XX,while the karyotype from the villous core mesenchyme was triploid (68, XXX, (-11)) in all analyzed cells (Sarno *et al.* 1993). The second case was described by Kawasaki *et al.* in 2016. A 27-year-old primigravida


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with partial molar pregnancy delivered at 25 weeks of gestation. The neonate was a 576-g girl, hospitalized for 134 days due to severe prematurity, but finally discharged home in a good general condition. Her peripheral blood karyotype was diploid (46,XX), while the chromosome complement of the placenta was 69,XXX (Kawasaki *et al.* 2016). Most recent case was described in 2018 by Loza and Fang. A 34-year old primigravida delivered healthy boy at 32 weeks due to preterm labor, both mother and the child were discharged without further complications (Loza & Victor Fang 2018). Our case is the fourth one with normal viable newborn with diploid karyotype and a triploid karyotype of the placenta. The female infant weighting 740g at birth was discharged home at 77th day of life in a good general health. On the contrary to the three previously reported cases of live-born infants from partial molar gestations, where triploid karyotype of the placental tissue had 3 X chromosomes, in our case the placenta had a 69,XXY karyotype. No such case was described before. Booth &Eskandar presented a case of a partial molar pregnancy with a placental diploid/triploid mosaicism (46,XX/69,XXY) associated with a viable fetus, but the pregnancy was terminated at 21 weeks of gestation due to severe pre-eclampsia (Booth & Eskandar 2018).

Molar pregnancy has a high risk of fetal and maternal complications, like abnormal fetal karyotype, fetal anemia and intrauterine growth restriction, vaginal bleeding, hyperthyroidism, pre-eclampsia, placental abruption or persistent gestational trophoblastic disease (Sanchez-Ferrer *et al.* 2009; Yoneda *et al.* 2013; Kawasaki *et al.* 2016; Booth & Eskandar 2018).The prevalence of pre-eclampsia in molar pregnancies is nowadays reduced due to early detection and first trimester terminations, however in 1970s, when most molar gestations were left without intervention until the second trimester, pre-eclampsia occurred in 27% of those pregnancies (Kohorn 1984; Soto-Wright *et al.* 1995; Koga *et al.* 2010). Severe pre-eclampsia has been reported in women with molar pregnancy with a viable fetus (Yoneda *et al.* 2013,; Kawasaki *et al.* 2016). Among 5 described cases of triploid partial mole with diploid fetus pre-eclampsia occurred in 3 of them (Yoneda *et al.* 2013; Kawasak *et al.* 2016; Booth & Eskandar 2018). In our case the patient had two eclampsia attacks postpartum. Placental growth factor (PlGF) and soluble fms-like tyrosine kinase 1 (sFlt1), which acts as a potent antagonist for vascular endothelial growth factor (VEGF) and PlGF, play an important role in the etiology of pre-eclampsia. In women with molar gestation serum concentration of sFlt-1 are significantly higher, while levels of PlGF significantly lower than in women with normal pregnancies (Koga *et al.* 2010). In a partial molar pregnancy with triploid placenta and diploid viable fetus, described by Yoneda *et al.*, maternal serum levels of sFlt-1, sFlt-1:PlGF ratio and soluble endoglin (another anti-angiogenic factor) were more than 3-times higher than in women with pre-eclampsia (Yoneda *et al.* 2013). Such elevated concentrations of anti-angiogenic circulating factors may contribute to the early-onset pre-eclampsia with symptoms even before 20 weeks of gestation, or eclampsia (Melody 1946; Cox & Klein 1993; Yoneda *et al.* 2013; Booth & Eskandar 2018).

In mammalian both paternal and maternal genomes are essential for embryonic development (Hsu *et al.* 2008). The presence of paternal chromosomes is associated with hyperplasia of the trophoblast, while maternal chromosome contribution is associated with an initiation of the embryo development (Booth & Eskandar 2018). There are several hypothesis of a partial mole origin: a)fertilization of a haploid ovum by a single haploid sperm, which subsequently duplicates its chromosomes after fertilization, b) fertilization of a haploid ovum by two haploid sperms, c)fertilization of a diploid ovum by one haploid sperm or d)fertilization of a haploid ovum by a diploid sperm (Sanchez-Ferrer *et al.* 2009). The prevalence of reported sex chromosome constitutions XXX, XXY and XYY in triploid placentas are 27%, 69% and 3% respectively (Sanchez-Ferrer *et al.* 2009).

In two previously described partial molar pregnancies with triploid karyotype of the placenta (69,XXX) and diploid karyotype of the fetus (46,XX), the presumed hypothesis was the fertilization of a haploid ovum by a haploid sperm and a further duplication of the paternal chromosomes in only a trophectodermal cell (Yoneda, *et al.* 2013). This way triploid cells occurred only in trophoblast, while the fetus had a diploid karyotype. It is worth noticing that all reported viable infants from molar pregnancy were female. Another possible mechanism is a non-segregation of paternal chromosomes at stage II of meiosis and fertilization of a haploid ovum by a diploid sperm (Kawasaki *et al.* 2016). According to Zaragoza *et al.* around 8% of all diandric triploids originate from a diploid sperm (Zaragoza *et al.* 2000). In oligospermic men most triploid embryos develop from diplospermy, while in normospermic men from fertilization of two haploid sperms.

Sarno *et al.* proposed another mechanism of partial molar pregnancy with diploid embryo development. In their case they assumed that placental mosaicism of triploid and diploid cells (68,XXX (-11)/46,XX) resulted from a loss of a haploid set in both embryonic and trophoblastic progenitors, leaving the majority of trophoblastic cells triploid. Therefore triploidy affected only extraembryonic mesenchyme in the chorion and villous core tissue. The mosaic placenta developed as a partial molar gestation, while the fetus with a normal diploid genetic constitution developed normally (Sarno *et al.* 1993). Post-zygotic diploidization of triploidy has been proposed to explain the developmental mechanism of molar pregnancy by other authors. It can occur during the first cleavage in either diplospermic or dispermic zygote to form two cell lines (Golubovsky 2003). In those cases triploids

may divide into two daughter cells with a loss of complete chromosome set in one of them. It has been demonstrated in mouse species hybrids that paternal and maternal genetic contributions remain separated up to the four-cell preimplantation stage (Mayer *et al.* 2000). Therefore, it is possible that formerly triploid embryo may lose one complete set of chromosomes in early development. The loss of paternal chromosome set, including the chromosome Y, in the first cleavage of the triploid zygote, is a probable mechanism of pregnancy development in our case. As fetus develops from a small number of progenitor cells, a preferential partitioning of the triploid cells to the extra-embryonic cell lineages may lead to confinement of abnormal cells to the placenta. The presented case is the one illustrating the above theory. Alternative possibility is of pentaploid conceptus also discussed in the literature. In this hypothesis incorporating of a polar body leads to a tetraploid ovum and its fertilization by a haploid sperm. The pentaploid conceptus subsequently divides into diploid and triploid sets (Zhang *et al.* 2000). Whether this theory can actually occur in humans is not known. However, in such case diploid fetus would have two maternal chromosome sets, while the placenta two maternal and one paternal sets (digynic triploidy). This hypothesis can be excluded in our case as placenta had a diandric triploidy.

In conclusion we present the first case of a live born female infant with diploid biparental karyotype born from a pregnancy complicated with partial mole due to diandric triploidy. The biological mechanism pathomechanism behind its development remain unclear. Our data, supported by other scarce reports, suggests increased vigilancein care of patients with placental malformations, but proves the possibility of live birth even in cases of severe genetic disorders in placenta.

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