



Case Report

Fetus papyraceous disguised as compound presentation: A case report



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ABSTRACT

Introduction: and importance: Fetus papyraceous (FP) is a rare condition that describes a mummified fetus in a multiple gestation pregnancy in which one fetus dies and becomes flattened between the membranes of the other fetus and uterine wall. Compound presentation occurs when the fetus's arm or leg is next to the main presenting part, very often the vertex is combined with arm presentation. A severe complication can occur in mother and child in such cases.

Case presentation: We report a case of incidental finding of fetus papyraceous disguised as a compound presentation during normal delivery which was managed successfully without any complication.

Discussion: Regular antenatal chorionicity assessment is essential for early diagnosis of fetus papyraceous and reduces mortality and morbidity in the surviving fetus.

Conclusion: Early identification of such cases is necessary as it is associated with severe complications like pre-term labor, infection from a retained fetus, severe puerperal hemorrhage, consumptive coagulopathy like disseminated intravascular coagulation, and obstruction by a low-lying fetus papyraceous producing dystocia leading to cesarean birth.

1. Introduction

Compound presentation is defined as the presentation of a fetal extremity (hand, arm, and foot) alongside the presenting part either the vertex or the breech. It may involve one or more extremities. The majority of compound presentations are represented by the fetal hand or arm presenting with the vertex [1]. Management for compound hand presentation is expectant because the extremity will often retract as the head descends and the manipulation of the extremity is usually avoided [1]. For term deliveries, compound presentations with parts other than the hand are unlikely to result in safe vaginal delivery that may lead to complicated compound presentations which include cord prolapse and injury to the presenting limb. Fetus papyraceous (FP) is the condition where the intrauterine fetal demise of a twin early in pregnancy occurs, with retention of the fetus resulting in mechanical compression of the small fetus such that it resembles parchment paper [2]. We report a fetus papyraceous disguised as compound presentation. The CARE Criteria were followed in reporting this case study [3].

2. Presentation of case

An unbooked 28 years G3P2L2 lady at 40 weeks 4 days period of gestation presented to Dhulikhel Hospital antenatal clinic (ANC) with a complaint of post-dated pregnancy. The USG done in the second trimester revealed a live, singleton pregnancy with no anomaly detected.

On abdominal examination, we found a term-sized uterus present in a longitudinal lie with adequate contractions, cephalic presentation, engaged head, and fetal heart rate of 148 beats per minute. On per vaginal examination, the cervix was parous and uneffaced. She was admitted for induction of labor.

On a detailed evaluation in the labor room for induction of labor, a sharp and solid body was felt on the right side of her cervix and was diagnosed as compound presentation. She was allowed for spontaneous progress of labor. Bishop's score was found to be 6 and she was having mild contractions.

The next morning Artificial rupture of membranes (ARM) was done with the intention to manipulate the hand supposed to be the compound

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presentation, and if the manipulation failed, she was planned for cesarean section. After ARM, on manipulation the feeling of the supposed hand was different from the usual feeling of the fetal hand, so per speculum was done, a whitish structure similar to the fetal feet was noted which was corresponding grossly to that of around 13/14-week fetus. So, a case of fetus papyraceus was suspected and with the hooking method, the fetus papyraceus, about 16cm in length, mummified, flattened, yellowish with identifiable head and limbs were taken out along with its thin umbilical cord (Fig. 1). The cervix was 4 cm dilated with 60% effacement and with the vertex of the healthy fetus at zero station. She was then augmented with Oxytocin, followed by vaginal delivery of a live 3200 g male neonate (Fig. 2) with no perinatal or maternal complication.

3. Discussion

The term fetus papyraceus is used when intrauterine fetal demise of a twin early in pregnancy occurs, with retention of the fetus for a minimum of 10 weeks resulting in mechanical compression of the small fetus such that it resembles parchment paper. Its incidence is estimated to occur between 1:184 and 1:200 [4].

The specific etiology of fetus papyraceus is unknown, however it has been suggested that twin-to-twin transfusion syndrome, fetal genetic and chromosomal abnormalities, and umbilical cord insertion anomalies (such as velamentous insertion and deadly nuchal cord) may play an important role [5,6]. In most cases death occurs in the second trimester and a co-twin who dies early may be totally absorbed but fetuses who die later are frequently macerated but not compressed [7]. In the late second and third trimesters of a twin pregnancy, a single fetal demise is associated with to severe morbidity and mortality in the surviving co-twin



Fig. 1. Fetus papyraceus.



Fig. 2. Healthy surviving twin.

[4]. The prognosis for a surviving dichorionic twin is reasonably fair, whereas the prognosis for a surviving monochorionic twin is poor [8]. As a result, antenatal ultrasonographic chorionicity assessment is critical in determining the possible risk [4].

Congenital anomalies in the second twin are found with intestinal atresia, gastroschisis, absent ear, aplasia cutis, central nervous system damage and heart anomalies in the child [9]. Maternal consequences of FP includes preterm labor, infection from a retained fetus, severe puerperal hemorrhage, consumptive coagulopathy like disseminated intravascular coagulation and obstruction by a low-lying fetus papyraceus producing dystocia leading to cesarean birth [10]. In contrast to our case where no such maternal complications were reported.

Routine ultrasound examination with better training and use of modern ultrasound machines with good resolution is important for detection of multiple gestations [10]. This will allow the diagnosis of FP early in pregnancy and may prevent future obstetrical complications and reduce the risk of mortality and morbidity for the surviving fetus [10]. In the case presented herein, the patient was unbooked and previous sonographic evaluation done during second trimester revealed only a singleton pregnancy. Another case report is available where, a case of FP was diagnosed during labor as a result of arrested descent [11]. On the other hand, our patient had a normal vaginal delivery post augmentation with Oxytocin without any such associated obstructed labor. When FP is diagnosed early, expectant management with close fetal and maternal surveillance is advised [11]. This could be done by performing serial ultrasound preferably every 2–3 weeks and monitoring of coagulation profile every fortnight [11].

A vanishing twin may cause an elevated maternal serum alpha-fetoprotein, an elevated amniotic alpha-fetoprotein level and a positive amniotic fluid acetyl choline esters. Serial evaluation of the

surviving fetus by sonography, biophysical profile, doppler and maternal serial evaluation by coagulation profile, FDP and D-dimer should be done. Unfortunately, there was no previous record of monitoring BPP, Doppler, maternal coagulation profile, FDP and D-dimer in our case.

Neonatal cranial ultrasound or magnetic resonance imaging is recommended after delivery and fetal MRI provides more detailed information about brain lesions in the surviving fetus [12]. However, in many cases of fetus papyraceus, there are no complications to the mother or to the surviving twin, as described in the two cases presented by Dahiya et al. which is quite similar to our case in the sense that no congenital anomalies were detected in the surviving fetus and no maternal complications were reported during delivery or postpartum [12].

In our case, this was an uneventful term pregnancy, presenting in OPD for a post-dated fetus misleading the case as a compound presentation. A prompt decision for the cesarean section was not made. ARM was planned for the next morning with the intention to let compound presentation be autocorrected and if the descent of the head failed, she was planned for cesarean section. After ARM the feeling of the supposed hand was a bit different from the usual feeling of the fetal hand so on performing a per speculum examination, a small paper whitish fetal limb-like structure was noted. Immediately, a diagnosis of fetus papyraceous was made and the dead fetus with its cord was taken out post which augmentation of labor was done which progressed to successful vaginal delivery of a live male baby. The patient and baby were observed for 24 hours and discharged.

4. Conclusion

Fetus Papyraceous may be misdiagnosed as compound presentation due to missed periodic ultrasound scans, putting the mother and the live fetus at immense risk. Thus frequent antenatal monitoring of the baby and the mother is essential, along with routine placental examination post-delivery, to avoid potential complications.

Ethical approval

N/A.

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Author contribution

Neeta Timilsina: Surgical procedure, patient care, manuscript writing. Suman Raj Tamrakar: Surgical procedure, Patient care, manuscript writing, guarantor. Sabina Thapaliya: Manuscript writing and editing. Chhavi Sachdeva: Concept, Manuscript writing, editing and review. Ashish Tamang: Concept, Manuscript writing, editing and review.

Registration of research studies

N/A.

Guarantor

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Consent

Written informed consent was taken from the patient. The patient's information has been de-identified.

Provenance and peer review

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Patient perspective

The patient agreed to consent to the case report after explaining that her information would be de-identified, and her report would be published in a scientific journal to add to existing literature about the disease.

Declaration of competing interest

None.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.amsu.2022.104481>.

References

- [1] D.P. Cruikshank, C.A. White, Obstetric malpresentations: twenty years' experience, *Am. J. Obstet. Gynecol.* 116 (1973) 1097–1104.
- [2] R.P. Dickey, S.N. Taylor, P.Y. Lu, et al., Spontaneous reduction of multiple pregnancy: incidence and effect on outcome, *Am. J. Obstet. Gynecol.* 186 (2002) 77–83.
- [3] D.S. Riley, M.S. Barber, G.S. Kienle, et al., CARE guidelines for case reports: explanation and elaboration document, *J. Clin. Epidemiol.* 89 (2017) 218–235.
- [4] H.H. Woo, S.Y. Sin, L.C. Tang, Single foetal death in twin pregnancies: review of the maternal and neonatal outcomes and management, *Hong Kong Med. J.* 6 (2000) 293–300.
- [5] N. Neelima, U. Gopalan, Fetus papyraceus: a rare case report. *International Journal of Reproduction, Contraception, Obstet. Gynecol.* 10 (2021) 1727–1728.
- [6] E. Daw, Fetus papyraceus—11 cases, *Postgrad. Med.* 59 (1983) 598–600.
- [7] K. Csécei, Z. Tóth, G.T. Szeifert, et al., Pathological consequences of the vanishing twin, *Acta Chir. Hung.* 29 (1988) 173–182.
- [8] L. Fusi, H. Gordon, Twin pregnancy complicated by single intrauterine death. Problems and outcome with conservative management, *Br. J. Obstet. Gynaecol.* 97 (1990) 511–516.
- [9] I. Upadhyaya, M. Pradhan, R. Sharma, Twin pregnancy with fetus papyraceous, *JNMA J Nepal Med Assoc* 48 (2009) 246–248.
- [10] M. Bozkurt, D. Kara, Fetus papyraceous in a twin pregnancy: a case report without any maternal and fetal complications, *Proc ObstetGynecol* 3 (2012) 1–5.
- [11] D. Matovelo, E. Ndaboine, Fetus papyraceus causing dystocia in a rural setting: a case report, *J. Med. Case Rep.* 9 (2015) 178.
- [12] P. Dahiya, R. Bains, Conservative management of fetus papyraceus: a report of two cases, *Oman Med. J.* 29 (2014) 132–134.