

Sudden Fetal Death Managed by Cesarean Delivery: Case Report and Literature Review

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Abstract

Background: Sudden fetal death (stillbirth) is common in low-income countries although it has always been underreported. There are few published studies in medical literature regarding the situation in Cameroon. It is often difficult to determine with certainty the cause of fetal death in Cameroon. We are describing the case of sudden fetal death managed at the Douala General Hospital and highlighting the pitfalls of etiologic diagnosis.

Case presentation: MC, a 30-year-old G2P0010, blood group O rhesus positive, unemployed, Jehova witness and married lady of the Bassa tribe of Cameroon was admitted to our department at 32 3/7 weeks gestation because of reduced fetal movements of two days onset. This was her second antenatal care visit and she had no previous ultrasound scan.

Her medical history is consistent with a spontaneous abortion at 6 weeks' gestation (no pathology or genetic studies done) and threatened abortion at 19 2/7 weeks gestation during the current pregnancy that was managed with bedrest. She has secondary level of education.

On examination the blood pressure was 108/85 mmHg and body mass index (BMI) was 25 kg/m². Her temperature was 37.5°C and pulse rate was 80 beats per minute. There was milk letdown on breast expression. The uterine height was 30 cm and the lie was longitudinal. The fetal heart tones were absent. The diagnosis of Sudden Fetal Death (SFD) with myoma previa was confirmed at ultrasonography. Her hemoglobin level was 9.2 g/dL and she underwent cesarean delivery with myomectomy. The etiologic diagnosis was never obtained despite verbal autopsy and post mortem clinical examination of the macerated fetus. No autopsy was done on fetus.

Conclusion: Sudden fetal death is a tragic obstetric accident. In our setting, verbal autopsy, ultrasonography and post mortem examination of the fetus are the cornerstones for an etiologic diagnosis. Management should be with a multidisciplinary team for better outcomes. Maternal anemia may be a contributing factor of stillbirth but the etiologic diagnosis of SFD is seldom made in our setting.

Keywords: Stillbirth, Verbal autopsy, Ultrasonography, Cesarean, Macerated, Fetus

Background

Sudden Fetal Death (SFD) is fetal demise in utero (stillbirth) at age of fetal viability; 28 weeks gestation according to WHO [1,2] or 22 weeks in high income countries [3]. An estimated 2.6 million stillbirths occurred worldwide in 2009 despite a 14.5% decline in stillbirth rate between 1999-2009 [2,4]. Majority, 98% stillbirths, occur in low-middle income countries [5]. The overall stillbirth rate in low-middle income countries was 28.9 per 1000 births, ranging from 13.6 in Argentina to 56.5 per 1,000 births in Pakistan [5]. An estimated 45% of stillbirths occur intrapartum [6]. Stillbirths are also common in Cameroon although they are for the most part underreported; there are few published studies that have reported a 1.3-2% stillbirth rate [7,8]. In another study at the Regional Hospital Buea Laboratory through implementation of the Strengthening Laboratory Management Toward Accreditation (SLMTA) project, the number of stillbirths decreased from 5% to < 1% [9].

It is often difficult to determine with certainty the cause of fetal death even after extensive evaluation. First, many risk factors that are associated with fetal death in epidemiologic studies are present in numerous apparently normal women with

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uncomplicated pregnancies. Second, most studies of fetal death do not include controls, making it difficult to ascertain the contribution of a potential abnormality to the stillbirth. Third, several conditions may be present simultaneously. Sometimes fetal death may be due to the interaction or additive effect of two or more disorders and conventional post mortem diagnostic systems fail to identify a specific cause in about half of SFDs [10].

We are describing the case of sudden fetal death managed at the Douala General Hospital and highlighting the pitfalls of etiologic diagnosis.

Case Presentation

A 30-year-old G2P0010 married lady is a Jehova witness of the Bassa tribe in Cameroon. Her blood group is O rhesus positive and was admitted to our department at 32 3/7 weeks gestation because of reduced fetal movements of two days onset. This was her second antenatal care visit and she had no previous ultrasound scan.

Her medical history is consistent with a spontaneous abortion at 6 weeks' gestation (no pathology or genetic studies done) and threatened abortion at 19 2/7 weeks gestation during the current pregnancy that was managed with bedrest. She is not working and has secondary level of education. She has never received blood transfusion and does not take alcohol or substance abuse. Furthermore, there is no family history of hypertension, diabetes or sickle cell disease.

Her physical examination showed blood pressure of 108/85 mmHg, weight 70 kg and height 1.68 metres (BMI=25kg/m²). The temperature was 37.5°C and pulse rate was 80 beats per minute. Her conjunctivae were not pale, her chest examination was normal and there was milk letdown on breast expression. The abdomen was distended by a gravid uterus with irregular surface. There were visible striae gravidarum and linea nigra at the midline. The uterine height was 30 cm and the lie was longitudinal but the presentation was difficult to appreciate because of an isthmic mass. The fetal heart tones were absent using the hand-held Fetal Doplex II FD2 Huntleigh Healthcare Doppler containing a 2 MHz probe. On vaginal examination, the vulva was clean, the cervix was long, closed and posterior.

During hospitalization, blood samples were taken for full blood count (Hemoglobin 9.2 g/dL, platelet count 180000/mm³), coagulation studies were normal but we could not have the results for fibrinogen and Fibrinogen Degradation Products (FDP). The Human Immunodeficiency Virus (HIV), HBsAg, syphilis, toxoplasma, and rubella serum titres were also normal. Furthermore, thick and thin film for malaria, C-reactive protein, urinalysis and culture, electrolytes were normal and fasting blood glucose levels (0.74 g/dL). High vaginal swabs and cultures for gonococci, chlamydia, mycoplasma, beta-haemolytic streptococcus, and bacterial vaginosis were taken and a transabdominal ultrasound scan was done to ascertain fetal viability, estimate gestational age, fetal weight, position, amniotic fluid volume, study the placenta, and confirm the mass found on physical examination. The ultrasound scan (GE Healthcare LOGIQ, USA) result showed a normal foetus in breech presentation and absent fetal heart pulsations. There was no placental detachment or retro-placental hematoma. There was also a uterine mass located at the anterior isthmic region measuring 83x 69 mm (probable leiomyoma in pregnancy)



Figure 1: Placenta, umbilical cord and macerated fetus.

The diagnosis of Sudden Fetal Death (SFD) with myoma previa was made. Further tests were then requested to get an etiologic diagnosis of the SFD. These tests included the indirect Coombs test (irregular agglutinins), antiphospholipid, lupus anticoagulant and anticardiolipin antibodies. Unfortunately, because of technical and financial reasons, these tests could not be carried out. We finally decided on caesarean delivery because of the myoma previa after concerting with other colleagues.

The patient was then referred to the anesthesiologist who validated the results of tests already done and requested for 1000 ml of typed, screened and cross-matched blood. The patient underwent a lower uterine segment transverse caesarean delivery of a macerated male fetus with no visible fetal anomalies who weighed 1700 gm under epidural anesthesia the following day. Furthermore, myomectomy was performed alongside the cesarean. The placenta was normal and weighed 410 gm. The umbilical cord was 1.5 m long and had 45 spirals (Figure 1).

The estimated blood loss was 800 ml and the patient did not undergo blood transfusion. The family did not consent to the histo-pathologic examination of the macerated fetus.

Her immediate post operative management included taking of vital signs (blood pressure, pulse, respiration, temperature, urine output). 2500 ml intravenous fluid (5% dextrose), antibiotic prophylaxis with ceftriaxone 1g q 12 hr x 48 hrs, enoxaparine 40 mg/day (lovenox™) for Deep Venous Thrombosis (DVT) prophylaxis, tramadol and paracetamol infusions (perfalgan™) were used for post-operative pain relieve and bromocriptine (parlodel™) for suppression of milk letdown. The patient was discharged on the 5th post-operative day on iron supplementation for 3 months. She was seen one month post-cesarean for check-up during which she was counselled on probable causes of the accident and advised on contraception. However, the patient opted for the natural method of contraception.

Discussion

This diagnosis of sudden fetal death may be associated with a cessation of previously perceived fetal movements or a decrease in pregnancy-related symptoms. In some cases, women will

present with bleeding, milk letdown, cramping, or labor. Clinical examination is of paramount importance in case of SFD because it enables us to decipher signs that could be associated with a probable etiology: fever, elevated blood pressure, signs of trauma, hydrorrhea, signs of rupture of amniotic membranes. Some specific signs of SFD such as milk letdown on breast expression, discordance of the uterine height and gestational age of pregnancy, a soft uterus, absence of fetal heart tones, and in cases where the presenting part is vertex and accessible (the cervix is open) one can actually palpate the collapsed and overlapping of the fetal skull bones if SFD occurred several days before. However, many patients with fetal death have no bleeding or contractions, and fetal death may precede clinical symptoms by a variable and often extended period of time. In such patients there will be reduction of abdominal size. In the index case, the clinical signs suggestive of SFD were reduction of fetal movements, milk letdown and absence of fetal heart tones. The definitive diagnosis of the index case was by ultrasonography, confirming the presence of a fetus and the absence of visible fetal heart pulsations [11-13]. Furthermore, this patient felt reduction of fetal movements two days earlier before coming to hospital. Maybe if she came earlier we could have saved the life of the fetus, making it a "near miss", thus corroborating other studies [14].

Demographic factors for fetal death include race, low socioeconomic status, inadequate prenatal care, less education, and advanced maternal age [15]. Our patient came for antenatal care for the first time at 19 2/7 weeks because of threatened abortion. One study in Cameroon reported that the mean gestational age of first antenatal care visit was 19.2 (SD 4.2) weeks; range 8 weeks to 31 weeks especially among rural dwellers [16]. There was a strong negative correlation between gestational age at start of antenatal care and overall adequate care in the study ($p < 0.001$). These pregnant women are likely to have <3 antenatal care visits and majority, (51.2%) pregnant women's prenatal care visit is by nurses and midwives. They are also unlikely to have a vaginal, stool examination or ultrasound compared to those followed-up by doctors. Inadequate care, being an adolescent and single were associated with adverse pregnancy outcomes including preterm birth, postterm birth, stillbirth, labour induction and augmentation, labour dystocia, low Apgar score and low birthweight [16-18]. Other factors associated with inappropriate prenatal care in Cameroon include: rural dwellers, low educational level, high parity, low monthly income of women, health institution attended, qualification of healthcare provider, low partner's educational status and income [16,17]. Late ANC visits is common in most sub-Saharan African countries. Similarly, African-American women have rates of IUFD that are more than twice the rate for white mothers [19,20]. In part, this may be due to secondary risk factors such as socioeconomic status and lack of prenatal care [19,20].

Increasing maternal age after 35 years is associated with an increased risk for fetal death [21]. Advanced maternal age is an independent risk factor of stillbirth among nulliparous women and not for parous women. This may be as a result of maternal adaptation in pregnancy [22]. However, our patient was only 30 years old and she had a miscarriage in her first pregnancy where no genetic studies were done. This may be a risk factor for the SFD she just had which may have warranted the use of low dose acetyl salicylic acid (Aspirin™) early in pregnancy for the prevention of SB [23].

The rate of fetal death is also higher among obese women. Numerous studies have shown a consistent doubling in the risk of fetal death in cases of overweight and maternal obesity; BMI of 30 kg/m² or more. High BMI increases the risk of several conditions known to increase the risk of stillbirth, such as diabetes mellitus, hypertensive disorders including preeclampsia, socioeconomic status, and smoking. Nonetheless, obesity remains associated with fetal death after controlling for these confounders. The association between obesity and fetal death is of particular concern given the dramatic and persistent increase in the rate of maternal obesity [24,25]. The index case was overweight with a BMI of 25 Kg/m². Stephansson et al. 2001 in their Swedish case control study of 649 cases and 690 control (lean) nulliparous pregnant women found that maternal overweight BMI 25-29.9Kg/m² increased the risk of antepartum stillbirth (OR 1.9: 95% CI 1.2-2.9) especially term antepartum stillbirth (OR 2.7: 95% CI 1.5-5.0) whereas weight gain during pregnancy was not associated with risk [26]. The relationship between overweight/obesity and stillbirth is not well understood. However, metabolic disorders such as diabetes and pre-eclampsia (syndrome X) are associated with overweight/obesity and are in turn associated with stillbirth [27]. Furthermore, the odds of being overweight/obese are 80.8% higher for micronutrient deficient mothers than for non-deficient mothers [28]. Finally, Obesity has also been associated with altered perception of fetal movements, with more overweight and obese women presenting with reduced fetal movements compared with women of normal weight [29]. Overweight/Obese pregnant women have significantly more sleep-related disordered breathing than normal weight women [30]. Snoring has been associated with fetal growth restriction and pregnancy-induced hypertension [31]. A case study report suggested a link between obstructive sleep apnoea and stillbirth [32].

It is estimated that maternal diseases play a role in fetal deaths. The median late fetal death rate was 6.6 per 1000 deliveries (interquartile range 4.2-26.8 per 1000 births). Hypertensive disorders were the most common (2.7%), followed by other complications/diseases (2.5%), hemorrhagic disorders (1.1%), and infective disorders (0.6%) [33].

Numerous retrospective and prospective studies have linked recurrent pregnancy loss, especially fetal death, with antiphospholipid syndrome. The two best characterized antiphospholipid antibodies are lupus anticoagulant and anticardiolipin antibodies [34]. Fetal death also has been associated with heritable thrombophilias [35]. Several case series and retrospective studies reported an association between the factor V Leiden mutation (associated with abnormal factor V resistance to the anticoagulant effects of protein C) [36]. However, these tests are not done in our health facility. The index case was moderately anemic Hb=9.2 g/dL. Others have reported that maternal anemia Hb<10 g/dL was associated with stillbirth (adjusted OR=3.8; 95% CI 1.7-8.6, $p < 0.001$) [37,38].

In case of SFD, verbal autopsy is very important. Here we interview the patient meticulously, and relatives where possible, and do a good autopsy on the fetus. Examination of the fetus in the index case was difficult to carry out because of maceration. We had to look for malformations, lesions caused by infection or hemorrhage and karyotype. The placenta and its adnexes were examined to look for causes linked to the placenta and umbilical

cord. In our patient the umbilical cord was 1.5 metres long with 45 spirals. This finding alone could not explain the SFD because there was no nuchal cord.

Testing should be limited to cases wherein clinical history or other testing raises suspicion for a particular disorder. Ideally, the clinician should discuss clinical details as well as physical and laboratory findings with the pathologist so that the workup is tailored for each individual loss.

The limitations to having appropriate management of SFD in our health facility is the lack of financial and technical know-how to appropriately investigate patients with SFD. Furthermore, there is no possibility to carry out genetic studies on fetuses after demise. This is compounded by the fact that parents in Cameroon will hardly consent to an autopsy. Other studies have reported that among 144 cases of stillbirth examined, 104 (72%) underwent autopsy and these cases constitute the cohort of study. The clinical and laboratory information alone identified a cause of death in 35 (24%). After placental pathologic examination, 88 (61%) cases had a probable cause of death identified. The addition of autopsy resulted in 78 (74%) cases having an identifiable probable cause of death. Placental examination alone changed clinical management in 52 (36%) cases. Autopsy led to additional clinical management changes in 6 (6%) cases [39]. There is no psychologist in our hospital to manage post-SFD psychological problems and we did not have an etiologic diagnosis of the case.

The clinical implication of this case is that we should sensitize parents to consent to autopsy. This will be valuable to alleviate their psychological bereavement after the loss and elucidate preventable causes in future pregnancies. There is need for a resident psychologist in our hospital.

After an IUFD about 75%-90% of cases will get into spontaneous labour and deliver vaginally within 15 days. However, we always resort to induction of labour because of psychological stress although our patient did not have any psychological stress. It is also important to have a multidisciplinary team and to associate parents in management decisions.

This patient had a cesarean birth because of the myoma previa. Other studies reported that women with stillbirth usually delivered vaginally regardless of whether labor was spontaneous or induced or whether they had a prior cesarean delivery. However, 15% underwent cesarean delivery, often without a documented obstetric indication [40].

Conclusion

Sudden fetal death is a tragic obstetric accident. In our setting, verbal autopsy, ultrasonography and post mortem clinical examination of the fetus are the cornerstones for an etiologic diagnosis. Management should be multidisciplinary for better outcomes. Maternal anemia may be a contributing factor for stillbirth in this case but the etiologic diagnosis of SFD is seldom made in our setting. CARE guidelines/methodology were adhered to in the preparation of the manuscript.

Abbreviations

BMI: Body Mass Index

DVT: Deep Venous Thrombosis

FDP: Fibrinogen Degradation Products

Hb: Hemoglobin Level

HIV: Human Immuno-deficiency Virus

IUFD: Intrauterine Fetal Death

OR: Odds Ratio

SB: Still Birth

SFD: Sudden Fetal Death

SLMTA: Strengthening Laboratory Management toward Accreditation

WHO: World Health Organization

Declarations

Ethics approval and consent to participate: Ethical Clearance was sought from the Ethics Committee of the Douala General Hospital and Authorization was obtained from the Director General of the Douala General Hospital. Written informed consent was obtained from the patient to report the case.

Consent to publish: Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the editor-in-chief of this journal.

Availability of data and material: The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Competing interests

The authors declare that they have no competing interests.

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Author's contributions

TOE wrote the manuscript. TOE, FGMN, CNT and MNJA were in the surgical team. All authors read and approved the final manuscript.

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