

Double true umbilical cord knots coexisting with a nuchal cord with successful fetal outcome: A case report

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Abstract

Umbilical nodes and cords play a crucial role in fetal development and are essential for the transfer of nutrients and oxygen between the mother and the fetus. Sonographic diagnosis of umbilical nodes and cords has become an integral part of prenatal care, allowing for the early detection of abnormalities and potential complications. The umbilical cord is a vital structure connecting the fetus to the placenta, providing essential nutrients and oxygen for fetal growth and development. Sonographic examination of the umbilical cord and its associated nodes has become an indispensable tool in prenatal care, enabling the early detection of abnormalities and potential complications. This review aims to analyze the current literature on sonographic diagnosis of umbilical nodes and cords, highlighting the key points and advancements in this field. A 37-year-old booked G₄P₂+¹A₂ Nigerian woman was registered for prenatal tertiary health care at 12 weeks of gestation. The booking investigations were normal and the booking packed cell volume was 37%. She was compliant with scheduled clinic visits and routine drugs. Pregnancy was carried to term uneventfully. Elective cesarean section was successfully performed at 38 weeks of gestation owing to the patient's prior history of third-degree perineal tear. The intraoperative findings included a loose cord around the neck of the baby and double true knots along the length of the 65 cm umbilical cord. The baby was delivered with appearance,

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pulse, grimace, activity and respiration (APGAR) scores of 7 in the first minute, 9 in the fifth minute, and the birth weight was 3.0 kg. Mother and baby were discharged 48 h postpartum in stable clinical condition. Although the presence of true double umbilical knots is rare, its coexistence with the nuchal cord is even rarer. There are risk factors associated with true umbilical knots. The possible risk factor implicated in this index case is the gender of the fetus and maternal multiparity. True umbilical knots are usually associated with certain fetal negative outcomes of pregnancy. There are currently no evidence-based treatment options available.

Keywords

Placenta, nuchal cord, umbilical cord, umbilical cord knots

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Introduction

True umbilical cord knot, albeit not a frequently faced obstetric issue in clinical practice, has a vital emotional encumbrance on both the obstetrician and the affected pregnant women.¹ Fetuses with umbilical knots have a 4- to 10-time upsurge of stillbirth during pregnancy as well as perinatal ill health in up to 11% of cases.^{2,3} The frequency of occurrence of a solitary true knot in the umbilical cord is very low with frequency ranging from 0.3% to 2% of pregnancies. Although the incidence of the occurrence of a double true knot in a birth cord is not known, it is expected to be rarer than the single true knot.^{2,4} Quite often, non-hypoxic non-acidotic newborn deliveries ensue more regularly when the nuchal cords are single. This frequently can occur at term.⁵

True umbilical cord knots can decrease or even stop blood flow totally to the fetus when it gets tight. This is usually associated with various extents of hypoxia which could lead to fetal distress or demise in utero during the prenatal course of fetuses with umbilical true knots.^{6,7}

The gap in the knowledge on this case report was that there is a dearth of knowledge on the outcome of multiple umbilical cord abnormalities (coexistence of true knots and nuchal cord). Currently, there is a dearth of substantiation and agreement on the best management of umbilical cord knots detected prenatally.³⁻⁷ There is also a lack of prospective studies investigating the outcomes of double umbilical true knots. However, a previous systematic review and meta-analysis recommended future research on the effects of multiple umbilical cord abnormalities (combined true knots and nuchal cord) on adverse pregnancy outcomes.⁸ Therefore, it is of particular interest to the authors of the current study to explore the implications of true double umbilical knots with coexisting cords. We report a clinical occasion of true double umbilical knots coexisting with the nuchal cord which was detected and managed during an elective cesarean section due to a prior history of third-degree perineal tear. The baby had close neonatal monitoring, and we proposed a diagnostic and management algorithm based on our experience and a review of the existing literature.

Case presentation

Our patient is a 37-year-old booked G₄P₂+¹A₂ Nigerian woman whose last menstrual period was dated April 14, 2022, date of delivery estimated to be January 21, 2023, by Naegele's rule.⁹

Pregnancy was desired and achieved spontaneously. She booked for antenatal care at an estimated gestational age of 12 weeks. Booking investigations were done and there were no abnormal findings. The booking packed cell volume was 37%. She was compliant with scheduled antenatal clinic visits and routine medications. The obstetric transabdominal ultrasound scan done at 21 weeks of gestation revealed a viable intra-uterine pregnancy with no gross abnormality noted. The placenta was noted to be posteriorly located, liquor volume was normal for gestational age and the estimated fetal weight was 397 g.

She was counseled on the need for an elective cesarean section at 38 weeks of gestational age due to a previous history of a third-degree perineal injury in her last childbirth. Adequate preparations were made for resuscitation at a designated area in the theater prior to delivery. Resuscitation equipment was arranged and tested and roles were assigned to the neonatologist. Intraoperative findings included a clean peritoneal cavity, normal fallopian tubes, and ovaries seen bilaterally. The umbilical cord seen hanging loosely around the neck of the baby was gently eased out. The umbilical cord and placenta were removed by controlled cord traction after the delivery of the baby. The placenta was normal, whereas the umbilical cord which was 65 cm long had two loose true knots along its length. The first knot was 10.4 cm from the baby and the second one was 7.5 cm distal from the first knot (Figures 1 and 2). The cord was found to be inserted centrally in the posterior placenta. A male baby was delivered with APGAR scores of 7 in the first minute, and 9 in the fifth minute, with a birth weight of 3.0 kg. The baby was wrapped in dry linen, dried alongside providing tactile stimulation. The baby had copious secretions from the mouth and nose which were suctioned. The umbilical stump with the true double knot was clamped and cut. The baby's oxygen saturation was 68% and random blood glucose was 93 mg/dl.

By the 10 min of life, oxygen saturation was 79% and the baby was crying vigorously on stimulation. Free-flow oxygen was given via nasal cannula at 1 liter per minute for about 5 min. By the 15th minute of life, the baby became pink all over with vigorous activity. Oxygen saturation was 93% in room air. The baby was observed for the next 15 min and remained stable with oxygen saturation between 93% and 94% in room air.

Fortunately, the neonate did not meet the criteria for admission into our neonatal intensive care unit (NICU).

Immediate and remote post-partum conditions were satisfactory for both the mother and neonate. Breastfeeding was initiated within 2 h post-delivery following adequate recovery. Maternal postoperative packed cell volume (PCV) was 34%. Following satisfactory post-partum conditions, both mother and neonate were discharged 48 h post-partum.

Discussion

Main findings and interpretation

True knots of the umbilical cord are an infrequent obstetric manifestation and they are known to be linked with neonatal asphyxia during delivery and increase the risk of stillbirth.⁷ Some factors significantly increase the danger of developing a true umbilical knot. These are increased cord length, monoamniotic twins, male babies, advanced maternal age, polyhydramnios, multiparity, and mothers who are diabetic.⁷ The index patient is an older obstetric patient and she had a male neonate.

Several studies have detected associations between the double umbilical knot and increased risk for intrauterine growth restriction, preterm delivery, and emergency cesarean section even in the absence of other anomalies.¹⁻³ True umbilical cord knot can significantly alter the outcome of the pregnancy negatively, hence leading to low APGAR scores, preterm birth, and low birth weight. Intra-uterine fetal death and NICU admission can occur.⁸

In a report by Carter et al.¹⁰ that determined the relationship between electronic fetal monitoring and neonatal outcomes, true knot-associated neonates at term births had analogous electronic fetal monitoring physiognomies in comparison with newborns lacking true knots. Furthermore, no significant differences were observed for neonatal morbidity. The occurrence of a true knot was not related to electronic fetal monitoring variations, repetitive late cardio decelerations, or newborn ill health, further buttressing that a true knot can be well-thought-out as an obstetric phenomenon without malignant implications.¹⁰

Current data on the contributions of umbilical cord in gestational hurdles are inconsistent and the guesstimates of the percentage of stillbirths as a result of umbilical cord hitches range between 3.4% and 26.7%.²⁻⁴ In one meta-analysis by Hayes et al. that aimed at determining which umbilical cord aberrations are linked with the risk of stillbirth as well as

other similar bad prenatal consequences revealed a 22% prevalence of nuchal cords and a 1% prevalence of true umbilical cord knots.⁸ In Hayes et al.'s study, evidence has not shown any relationship between stillbirth and the nuchal cord but the odds of stillbirth were considerably greater for a true knot of the umbilical cord.⁸ Similarly, Agarwal and Singh,¹¹ in their case series, stated that true knots are of obstetric import as they could lead to an eclectic continuum of adverse fetal outcomes such as intrauterine growth restriction, poor Apgar score at birth, fetal hypoxia, and even fetal death. Conversely, a study by Lichtman et al.¹² revealed a dearth of the relationship between true umbilical cord knots and long-standing nervous system ill health in the progeny. In a case where there is a coexistence of double true umbilical knots and nuchal cord, it is expected to have an increased snowballing threat of these negative outcomes.^{3,13}

The firmness of the true knot of the umbilical cord can lead to the barricade of the fetal blood exchange. This may result in intra-uterine growth restriction or fetal demise. This circumstance might predispose to unrestricted and disproportionate fetal movements. While it is a known fact that not all true knots lead to perinatal complications, they have been associated with adverse pregnancy outcomes.^{2-5,14}

To our knowledge, there are only four previous studies that reported cases of double umbilical cord knots in the literature.^{7,15,16} The first case was in Germany by Hönigl et al.¹⁵ The second case was in Iran as reported by Haghighi et al.¹⁶ and the third case was in Portugal as reported by Laranjo et al.⁷ while the fourth one was in Italy as reported by Stabile et al.¹⁷ Our findings concur with previous studies of the double umbilical cord in Germany, Iran, Portugal, and Italy because of the baby survival.^{7,15-17} As in the previous four reported cases of double umbilical cord, our diagnosis was confirmed post-delivery, since there were no symptoms of fetal compromise during pregnancy. Similar to Laranjo et al.'s⁷ report in Portugal, our case was unique and reportable because there was a coexistence of a double knot and nuchal cord with successful neonatal outcomes. A few authors adopt that 3D power sonography may be valuable in the identification of true umbilical cord knots.^{1,8} Nevertheless, 3D power Doppler cannot be well-thought-out as an ultimate method and is not routinely performed during pregnancies.¹⁶ However, no documented precise antenatal signals exist to persuade the physician to look for ultrasound signs suggestive of the umbilical true knot. Some authorities have described a characteristic sonographic "hanging noose" sign when a transverse section of the umbilical cord is surrounded by a loop of the umbilical cord, for diagnosis of a true knot of the umbilical cord.^{18,19} Prenatal diagnosis of a true umbilical cord knot is very challenging because of the lack of ability to assess the umbilical cord length in its entirety.¹⁴ It should be noted that even if the diagnosis is made prenatally, there is difficulty in choosing the right time for delivery, and also the route of delivery as an intervention may worsen the outcome. There is no evidence of any preferred ideal delivery mode in



Figure 1. Baby immediately after delivery (the first minute of resuscitation) with umbilical cord showing areas of true knot.

this kind of case. Although we are not certain about the route of delivery of the first reported case of double true knot, the second case and ours had cesarean section while the third case by Laranjo et al.⁷ and the fourth case by Stabile et al.¹⁷ had vacuum extraction. However, some authorities contend that cases of fetal demise and fetal menace are directly related to the number of knots.^{1,7,8}

Our case study, however, reports a normal perinatal and post-natal course of a neonate associated with the true umbilical cord knots that measured 65 cm in length and this coexisted with the cord around the neck at delivery. True umbilical knots may act in a strictness-dependent manner, meaning that the damage caused by the presence of the cord knot may be affected by the level of compression. In other words, a tight umbilical cord knot is more likely to result in acute hypoxia, which ultimately may lead to intrauterine fetal demise. However, a loose knot may lead to chronic mild hypoxia and fewer upsetting outcomes. Along this reasoning, a few fetuses with knots may remain unaffected. This could explain neonatal survival in the case reported in the literature by Laranjo et al. where the fetus was delivered by spontaneous vertex but survived.⁷

In a previously reported case of a true knot of the umbilical cord without a nuchal cord in Nnewi, Nigeria, the outcome was unfavorable as the baby died immediately following delivery.²⁰ The umbilical cord length was 126 cm, which is almost double the length of the index case being reported ours. There was no gross fetal abnormality seen on the neonate. In their case, a true knot was found 7 cm distal to fetal insertion, and the cord was found to insert centrally

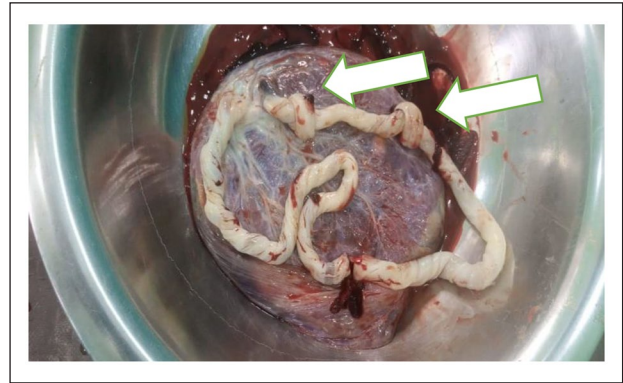


Figure 2. The placenta and umbilical cord show areas of true knots after it has been cut from the baby.

in the posterior placenta.²⁰ There was no associated umbilical cord prolapse.²¹

Sonographic evaluation of umbilical nodes

Umbilical nodes, also known as Wharton's jelly cysts, are common findings during routine prenatal ultrasound examinations. Sonographic evaluation of umbilical nodes involves assessing their size, location, and morphology.²² Recent studies have highlighted the importance of differentiating between normal umbilical nodes and pathological conditions, such as umbilical cord cysts or tumors. Advanced sonographic techniques, including color Doppler and three-dimensional ultrasound, have shown promising results in improving the accuracy of diagnosing umbilical node abnormalities.²³

Sonographic assessment of umbilical cords

Sonographic evaluation of umbilical cords primarily focuses on assessing their length, number of vessels, and presence of abnormalities. The detection of a single umbilical artery, for instance, may indicate an increased risk of fetal anomalies and adverse pregnancy outcomes. Doppler ultrasound has emerged as a valuable tool for assessing umbilical cord blood flow and detecting conditions such as umbilical cord compression or true knots. In addition, advancements in ultrasound technology, such as elastography, have shown potential in evaluating umbilical cord stiffness and elasticity.²⁴

Clinical significance and management

Accurate sonographic diagnosis of umbilical nodes and cords is crucial for appropriate clinical management and counseling. Identification of umbilical cord abnormalities can help guide obstetricians in determining the optimal timing and mode of delivery. Furthermore, early detection of umbilical cord complications, such as umbilical cord

prolapse or nuchal cord, allows for timely interventions and reduces the risk of adverse perinatal outcomes. Sonographic surveillance of umbilical nodes and cords throughout pregnancy is essential to monitor any changes or progression of abnormalities.²⁵

Sonographic diagnosis of umbilical nodes and cords has revolutionized prenatal care by enabling early detection and management of abnormalities. This analytical review has provided a comprehensive overview of the current literature on sonographic evaluation of umbilical nodes and cords, highlighting key points and advancements in this field. The adoption of advanced sonographic techniques, such as color Doppler and elastography, has significantly improved the accuracy of diagnosing umbilical cord abnormalities.²⁶ Further research and technological advancements are warranted to enhance our understanding and management of umbilical node and cord pathologies.

Strengths and limitations of the report

The strengths of our report are that we are the first to provide data on double umbilical cords with coexisting nuchal cord placement in low- and middle-income settings and the comprehensive literature review. However, it would have been nice to see a picture showing a double knot and nuchal cord before delivery, but this was overtaken by events since we could not make the diagnosis of a double umbilical cord prenatally.

Conclusion

True umbilical cord knots are very rare and even rarer are true double umbilical knots and nuchal cord in coexistence with good fetal outcome. This case revealed a good neonatal outcome following double true knots coexisting with a nuchal cord which was not detected on routine ultrasound during prenatal evaluations. There are currently no evidence-based treatment options available. There is a need for more research on how and when the diagnosis of true umbilical cord knots can be made prenatally.

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Author contributions

G.U.E., G.O.U., C.G.O., O.A.O., C.C.O., and C.C.O. conceived, supervised the study, and performed the surgery; C.B.N., K.O.N., E.P.I., E.U.N., T.K.N., O.C.E., E.C.E., D.E.M., and O.O.D. analyzed data; E.C.I., E.S.E., C.I.O., K.E.O., C.N.O., A.V.E., H.C.N., O.D.U., and C.O.E. wrote the manuscript. G.U.E., G.O.U., C.G.O., E.P.I., E.U.N., T.K.N., and O.C.E. made manuscript revisions. All authors reviewed the results and approved the final version of the manuscript.

Availability of data and materials

Data sharing is not applicable to this article as no datasets were generated or analyzed during this study.

Declaration of conflicting interests

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Ethical approval

Our institution does not require ethical approval for reporting individual cases or case series.

Informed consent

Written informed consent was obtained from the patient to publish patient information.

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