CASE PRESENTATION

Management of Sacrococcygeal Teratoma as a Cause of Dystocia During Labour

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ABSTRACT

Sacrococcygeal teratoma is a rare cause of dystocia. In this paper, we report two cases of sacrococcygeal teratoma. In the first case, the infant was partially delivered up to the level of the umbilicus, cesarean section was performed, during which a large sacrococcygeal teratoma, partly cystic and partly solid, was aspirated; the foetus was delivered vaginally with an Apgar score of 8 and 10 at one and five minutes, respectively. The second case presented with acute polyhydramnios and was found to have a large solid sacrococcygeal teratoma at 29 weeks gestation. The patient ruptured her membranes spontaneously and went into labour. The baby was delivered by cesarean section with an Apgar score of 5 and 8 at one and five minutes respectively, however, the baby died after 48 hours due to severe prematurity.

The encounter of an unexpected obstruction of delivery by a foetal tumour after partial expulsion of the infant in the midst of an anticipated normal spontaneous vaginal delivery is an obstetrician's nightmare.

In this paper, we report two such cases; in the first case, dystocia was caused by a large sacrococcygeal teratoma which was first diagnosed after the head, shoulders, upper arms and trunk of the infant were delivered. This type of crisis has only rarely been reported in the literature. The second case was a sacrococcygeal teratoma complicated by polyhydramnios, diagnosed antenatally by ultrasound examination and the patient was delivered by cesarean section in order to avoid dystocia.

Prolonged arrest at the level of the umbilicus poses considerable stress on the mother and her attendant and is likely to provoke the use of undue force in the absence of the alternative of operative intervention. If the diagnosis of foetal sacrococcygeal teratoma is suspected during the antepartum period, delivery by cesarean section is usually the treatment of choice.[1] The presence of such a neoplasm may be unexpected however, until dystocia occurs during delivery, as in our first case, or may be diagnosed antenatally whereby management can be planned in advance as in our second case.

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In the past, the outcome of an obstruction occurring during a vaginal delivery up to the umbilicus was usually fatal for the infant and was associated with maternal morbidity. Destructive debulking procedures, such as incisional drainage or blind dissection of the tumour, have been associated with extensive haemorrhage and usually, foetal death.[2]

CASE 1

A 22-year-old lady, gravida 2, para 1, was admitted to King Abdulaziz University Hospital, Jeddah, in early labour. Her first pregnancy ended by cesarean section due to malpresentation two years earlier, and a baby weighing 2.8 kg was delivered.

Since early in her second pregnancy, she was regularly followed up in the antenatal clinic at King Abdulaziz University Hospital and no discrepancy was noted between the abdominal size and gestational age. Ultrasound was not performed during the follow-up.

Examination on presentation in labour revealed a mature foetus with a clinically estimated weight of 3.5 kg. The pelvis was found to be adequate and the progress of labour was carefully monitored. Labour progressed smoothly and the patient reached full cervical dilatation and started to push. There was very slow progress in the descent of the presenting part. As the maternal condition was stable and foetal heart was satisfactory, no intervention was thought to be necessary. The foetus was delivered up to the umbilicus, when progress was arrested. Gentle traction was applied, but no further descent was achieved. A tense mass of unclear nature was noticed along the back of the foetus.

A tentative diagnosis of obstructed labour due to foetal tumour was made and no further attempt of forceful traction or aspiration was made, due to the unclear nature of the mass and the scar in the uterus, which contraindicated any manipulation.

During this period, the infant's condition remained good; respiration was unimpeded and asserted with oxygen mask. The baby cried vigorously and maintained a pink color and a satisfactory pulse, in spite of the half-delivered status.

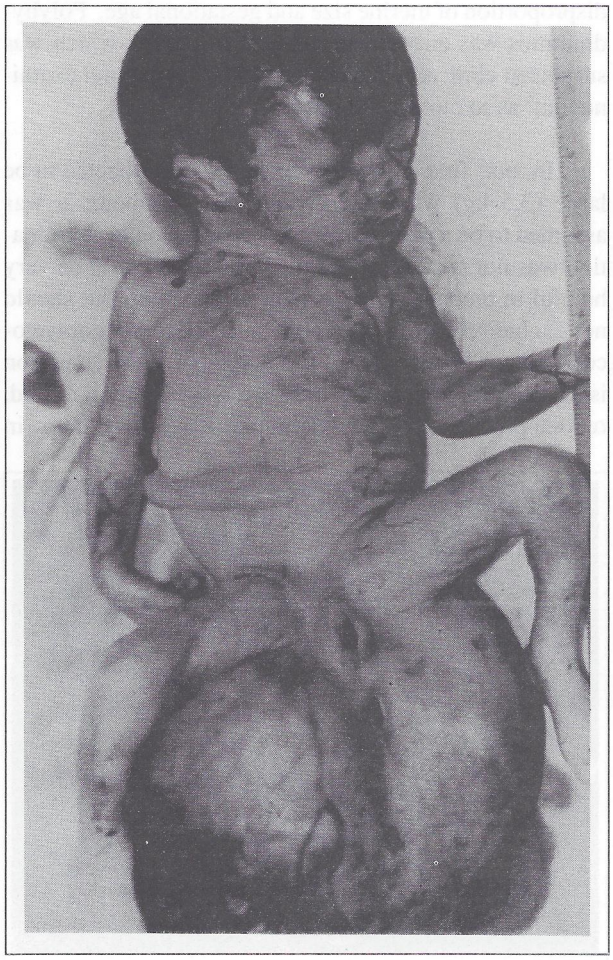


Figure 1a *Frontal view*

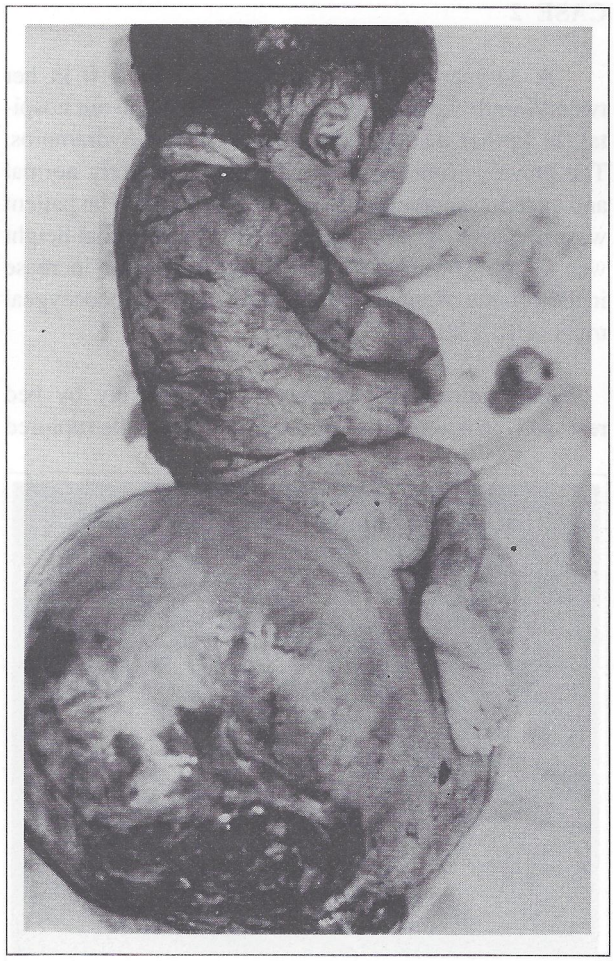


Figure 1b *Lateral view*

It was decided to proceed with the delivery by cesarean section and the mother was placed in a modified lithotomy position to allow a combined abdomino-perineal approach to the delivery of the infant. The abdomen was opened through a midline sub-umbilical incision. Transverse incision of the lower uterine segment revealed a tumour measuring 15 x 25 cm, which was attached to the foetal sacrum. It was found to be partly cystic, partly solid and tense. A paediatric surgeon was consulted and 1000 ml of serous fluid was aspirated by means of a 22-gauge needle. This led to a rapid collapse of the cystic mass and easy vaginal delivery of a mature, vigorous 3070-g female infant with an Apgar score of 8 and 10 at one and five minutes respectively (Fig 1a, b). The placenta was delivered through the abdominal incision, which was followed by the closure of the uterine, abdomino-perineal incision. The patient was commenced on antibiotics. The puerperium was marred by low grade pyrexia which was due to breast engorgement which responded to the expression of milk. Resection of the tumour was performed after two days and the baby was doing well at the time of this report (aged four months).

CASE 2

A 34-year-old lady, gravida 5, para 4+0 in her twenty-ninth week of gestation was referred to our hospital for further management because of polyhydramnios. The previous four pregnancies were completely normal and ended in spontaneous vaginal deliveries. The patient was in acute distress and her symphysis-fundal height was 43 cm. Ultrasound examination showed an increase in the volume of amniotic fluid and a solid sacrococcygeal mass of 15 x 20 cm (Fig 2a, b).

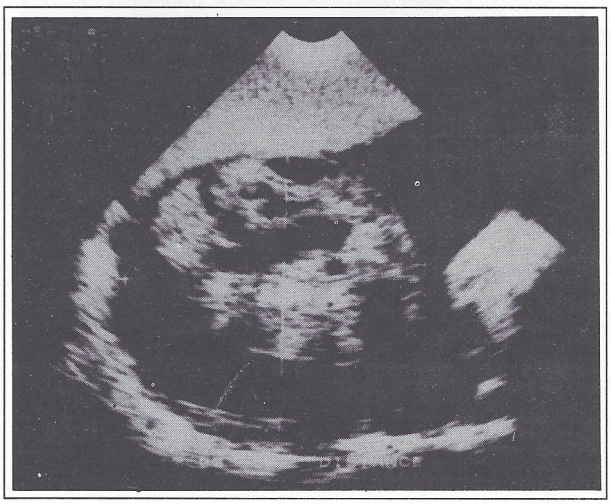
The patient was managed conservatively by bed rest and sedation, however after two days, she ruptured her membranes spontaneously and went into premature labour. She had three uterine contractions every ten minutes and 4 cm dilation of the cervix with the mass as the presenting part. Emergency cesarean section was performed and a 1280-g baby girl with an Apgar score of 5 and 8 at one and five minutes respectively, was delivered with difficulty through a lower segment transverse incision. The baby required intubation and died the second day post delivery due to severe prematurity.

DISCUSSION

Sacrococcygeal tumours are rare, with a frequency of only 1 or 2 per 40,000 live births; the majority occur in females. Although rare, they account for 8 to 11% of solid tumours encountered in childhood, and it has been estimated that dystocia may occur in 6.0 to 11.7% of cases.[3] Unfortunately, sacrococcygeal teratomas, have few if any, external clues and may remain undiagnosed until failure of delivery occurs, as in these reported cases.[4]

In a series of 10 cases, 6 were diagnosed by ultrasound examination, which was performed due to disproportion of uterine size and gestational age. Polyhydramnios was encountered in 3 of the cases, which was suspected clinically and confirmed by ultrasound examination,[5] as in our second case.

In our first case, the foetus was suspected to be large (3.5 kg) when the patient was in labour; it was assumed to be a normal baby, hence ultrasound examination was not performed. Ultrasonography could be very helpful in prenatal diagnosis and assessment, but should not exclude the differential diagnosis of meningomyelocele and to a lesser extent, haemangioma. If the condition is suspected prenatally, the levels of amniotic fluid, α-foetoprotein and acetylcholinesterase may help in



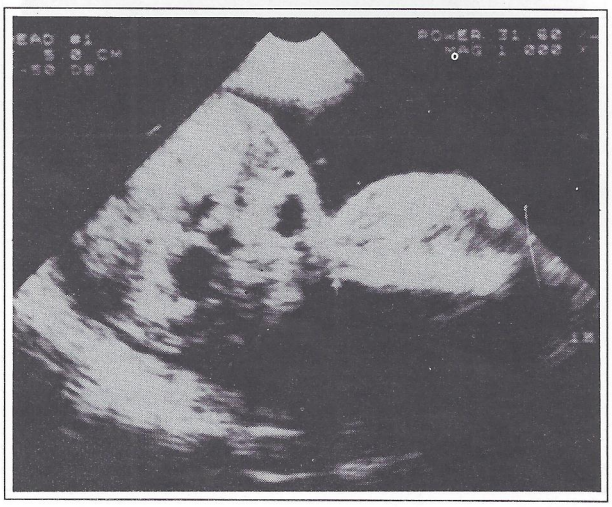


Fig 2a & b : *Ultrasound examination showing an increase in the volume of amniotic fluid and a solid sacrococcygeal mass.*

establishing the diagnosis, as these are found to be elevated in sacrococcygeal teratomas, especially in the second trimester.[2-5]

The main problem with this type of dystocia is that it is often not recognised until the patient is in the second stage of labour and the baby partially delivered. Careful re-assessment of the situation and vaginal examination allowed us to suspect a foetal tumour causing obstruction to further progress and delivery. However, it is not always easy to differentiate by palpation between sacrococcygeal teratoma and other sacral masses, such as meningocele, meningomyelocele or hamartoma.[3]

The circumstance which made this case unique was the presence of a cesarean section scar. We were aware that dystocia could also be caused by rupture of the uterus with partial extrusion of the foetus in the peritoneal cavity,[5] however, palpation of a large solid cystic mass made us ascertain that the presence of a foetal tumour was the cause of dystocia.

While the modes of management of partially delivered infants have been well-documented and individualised in the literature,[1,3-6] tumour size is critical in deciding which mode to employ. There are two reasons for this: first a large sacrococcygeal teratoma can cause serious obstetric complications, such as abnormal lie and dystocia, and second it can be highly vascular, thereby causing exsanguination or rupture on aspiration. Gross has reported cases of neonatal death after rupture of sacrococcygeal teratoma during vaginal delivery.[5]

Prolonged arrest at the level of the umbilicus causes considerable stress on the mother and may lead to foetal asphyxia. During normal delivery of the head, a volume of fluid equivalent to one third of the functional residual lung capacity is squeezed through the nose and mouth. The remaining lung fluid is absorbed by lymphatics and capillaries, presumably from pressure generated by glossopharyngeal respiration and chest recoil before the first breath and after delivery of the thorax.[7] Since the head and most of the thorax were delivered in this situation and several gasps were observed, it is reasonable to assume that most of the foetal lung fluid was replaced by air. If the foetus is adequately ventilated, undue haste is unnecessary and can cause injury. However, survival in this partially delivered state is unknown, therefore unnecessary delay should be avoided as this is likely to increase the chances of morbidity and mortality.

Open foetal intervention in animal models has revealed to be physiologically sound and effective for a variety of congenital anomalies, including urinary tract obstruction and diaphragmatic hernia.[6] In addition, early experience in humans with open foetal surgery for the correction of hydronephrosis has demonstrated the risk to the mother to be low and yielded encouraging results with appropriate patient selection.[8] Measurement of foetal aortic blood flow and cardiac output with a Doppler probe[6-9] may be used to follow the development of high output cardiac failure before the appearance of foetal hydrops, and might potentially be used to predict the outcome. Early intervention and open foetal surgery may become the answer to this problem.

CONCLUSION

Our first case highlights the occurrence of unexpected complications of labour and the necessity to ensure rapid access to optimal obstetric resources for parturient patients. The second case shows that prenatal diagnosis by early ultrasound is helpful. The role of Doppler echocardiography in predicting the outcome and early intervention is very promising because of the rarity of these cases.

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