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Trapped after Coming Buttocks: A Consequence of Sacrococcygeal Teratoma: A Case Report

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Authors' contributions

This work was carried out in collaboration between all the authors. All the authors designed the study and wrote the first draft of the manuscript. Author SMI managed the literature searches and all authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Sacrococcygeal teratoma (SCT) is the commonest fetal tumour but a rare cause of obstructed labour. If undiagnosed during antenatal period, high index of suspicion is required during labour to make a diagnosis. In this case report, we present a case of sacrococcygeal teratoma (SCT) which was diagnosed at the time of delivery. The fetus, with a massive ruptured sacrococcygeal mass, was delivered by traction and suprapubic pressure. Histology report revealed malignant sacrococcygeal teratoma. The diagnosis of sacrococcygeal teratoma (SCT) should form part of the differential diagnoses when there is obstructed labour after the delivery of the fetal shoulders so that appropriate management can be instituted.

Keywords: Sacrococcygeal teratoma; primigravida; obstructed labour; suprapubic pressure; stillbirth; Nigeria.

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1. INTRODUCTION

Obstructed labour due to fetal tumours are rare but when it occurs sacrococcygeal teratoma (SCT) is considered most common cause with a reported incidence of 1 in 35,000-40,000 live births [1,2]. The female to male preponderance is 4:1 and inheritance is generally sporadic although familial forms have been reported [3-5].

SCT arises from the Hensen's node which is made up of totipotent primitive cells that differentiate into ectoderm, endoderm and mesoderm [2,3]. Most patients present during the neonatal period with a sacral mass but the few with intrapelvic tumours usually present late outside the neonatal period [2]. It has a malignant potential which parallels the age of the patient at presentation [2].

Antenatal ultrasonography can effectively screen SCT, thus decreasing the incidence of labour complications arising from it [6]. However**,** in Nigeria where only 58% of women receive some antenatal care from skilled birth attendants and 65% of the total deliveries take place outside health facilities [7], a high index of suspicion and appropriate management are required as these patients may not benefit from antenatal screening but are encountered unexpectedly in emergency situation after unsuccessful attempts at delivering at home.

2. CASE REPORT

A 20-year old unbooked primigravida at a gestational age of 29 weeks was referred to our hospital due to inability to deliver the waist and lower limbs of the fetus that was trapped after delivery of the head and trunk. General physical examination was unremarkable. The abdomen was enlarged with symphysio-fundal height of 30cm; pelvic examination revealed a fetus delivered to the waist region with no cord pulsation.

Abdominal ultrasound examination at the time of presentation revealed a massive tissue and fluid collection with no identifiable fetal parts in the uterus. A matured female fresh stillbirth baby with a massive ruptured sacrococcygeal mass (Fig. 1) was subsequently delivered by traction and suprapubic pressure. A diagnosis of SCT with fetal death was made.

The post mortem revealed the body of a fresh stillbirth, matured female fetus with a huge sacrococcygeal mass revealing solid and cystic areas and extensive skin ulceration. There was atelectasis of both lungs. The brain, heart, kidneys, gastrointestinal tract, liver, spleen, bladder, ovaries and uterus are unremarkable. The histology of the sacrococcygeal mass shows malignant primitive germ cells tumour which are features of malignant teratoma (Fig. 2).

Fig. 1. Delivered neonate with ruptured sacrococcygeal teratoma

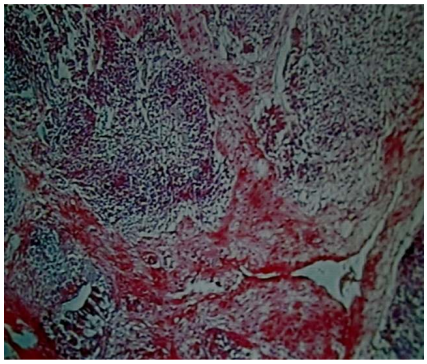

Fig. 2. Photomicrograph showing primitive malignant cells of teratoma (H&E. X132)

3. DISCUSSION

Although very rare, SCT is the commonest solid neoplasm of newborns [2,8]. To the best of our knowledge, the case presented was the first documented SCT in the centre where the patient was managed. The rarity of this condition is supported by the report of only 21 cases over a period of 18 years from North-east Nigeria, the same region with the study centre [9]. In addition, only few cases of SCT as a cause of obstructed

labour have been reported as most published cases were patients managed after delivery, following presentation in neonatology or paediatric surgery unit [6,10-15].

The parturient had unsupervised pregnancy and only presented in labour at a community hospital from where she was referred due to inability to deliver the fetal waist and lower limb after the delivery of the head and trunk. If the patient had availed herself of antenatal care, diagnosis of SCT might have been made with ultrasound scan and early management instituted [3]. Similarly if the baby was delivered alive, surgical intervention would have been instituted [16]. However in our environment where 58% of women receive antenatal care from skilled birth attendants and only 35% of them have supervised delivery [7], this late presentation is bound to be a common finding. In recent years, the practice of routine obstetrical ultrasound scan has led to a significant increase in the number of SCT diagnosed in utero and reduction in obstructed labour caused by SCT [3,11]. Fetal MRI has been reported to be superior to sonography in assessing the intrapelvic and intraspinal extent of tumours and compression of pelvic organs by the tumour [3]. Antenatal intervention is warranted in only specific circumstances and most fetuses do not need antenatal intervention. Intervention is needed whenever there is a high risk of developing hydrops. The aim of antenatal intervention is to halt or reverse the in utero physiologic changes that occur [3]. SCT can be resected in utero [3]. The first successful open fetal surgery to debulk the tumour was performed by Adzick et al.in 1997 [17]. Since then, a number of fetal surgeries have been done.

Tanaree in 1982 and Mistri et al. in 2012 in separate reports have documented their experiences in obstructed labour caused by SCT, and the delivery of the fetus [6,10]. Our patient was delivered vaginally by slight downward traction combined with application of suprapubic pressure. Accomplishing delivery this way was easier than recourse to caesarean section for a dead fetus who had already been delivered up to the trunk. This mode of delivery was utilised by Krishan et al. in 2004 [11]. Vaginal delivery is possible for uncomplicated tumours smaller than 10 cm even if the fetus is alive but when the tumour is 10 cm or more, caesarean section through a vertical incision is recommended [3]. In some of the reported cases, Caesarean section was combined with vaginal delivery because the fetus was alive and there was need to decompress the tumour to facilitate subsequent vaginal delivery [6,10]. Another delivery option is percutaneous drainage for cystic masses of SCT followed by vaginal delivery, which can reduce the risk to both mother and fetus since caesarean section requires a large uterine incision that is associated with increased maternal morbidity and caesarean delivery in subsequent pregnancies [9].

Differential diagnoses of SCT which must be excluded particularly if diagnosis is made antenatally include myelomeningocoele which is cystic and always associated with spinal dysraphism, and lymphangiomas, chordomas, and ependymomas which are usually smaller and seldom have a large external component [11].

The index case had preterm labour at 29 weeks gestational age probably due to increasing size of the tumour. The gestational age at diagnosis and delivery of the fetus closely affects the perinatal outcome [11]. Most fetuses with large SCT are born prematurely, with resultant increased morbidity due to lung immaturity and other complications associated with prematurity [11].

The finding of malignant teratoma is not surprising considering the poor perinatal outcome. Malignant SCT is rare and constitute 10% of SCT [3]. Abubakar et al. [9] and Sebire et al. [14] in separate reports from North-east Nigeria and London found malignant SCT in 14.3% and 20% of SCT cases studied. This implies that a good proportion of SCT is malignant.

Although the patient presented had a fresh stillborn, in the event of delivery of a live fetus with SCT, perinatal management requires an interdisciplinary team of radiologists, obstetricians, neonatologists and paediatric surgeons [11]. Surgical removal of SCT should be performed without delay to avoid complications such as haemorrhage and ulceration and to minimize the risk of malignant degeneration [11].

4. CONCLUSION

Sacrococcygeal teratoma (SCT) should form part of the differential diagnoses when there is obstructed labour after the delivery of the fetal shoulders so that appropriate management can be instituted. However, with improvement in diagnostic and therapeutic procedures and

antenatal care, more cases of sacrococcygeal teratoma can be managed early.

CONSENT

All authors declare that 'written informed consent was obtained from approved parties for publication of this case report and accompanying images'.

ETHICAL APPROVAL

The author obtained ethical clearance from the research and ethic committee of the institution, regarding the retrieval of the patient's case file and publication of the case report.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

REFERENCES

1. Rao KA. Obstructed labour. In: Textbook of midwifery and obstetrics for nurses. Gurgaon: Elsevier Health Sciences India. 2011;480-482.
2. Amoah M, Boateng N, Abantanga FA. Sacrococcygeal teratoma: A 4-year experience at Komfo Anokye Teaching Hospital. Postgraduate Medical Journal of Ghana. 2012;1(1):1-5.
3. Parlakgumus HA, Tarim E, Ezer SS. Antenatal diagnosis of sacrococcygeal teratoma two different case reports. Ginekol Pol. 2009;80:445-448.
4. Onuigbo WIB. Sacrococcygeal teratoma in a developing community. J Case Rep Stud. 2015;3(5):501.
   DOI: 10.15744/2348-9820.3.501
5. Yekula MSKK, Yekula A. Adult sacrococcygeal teratoma: The third leg, a rare case report in a 25 year old man. International Journal of Surgery Case Reports. 2015;14:146-148.
6. Mistri PK, Patua B, Alam H, Ray S, Bhattacharyya SK. Large sacrococcygeal teratoma hindering vaginal delivery attempted at home. Reviews in Obstetrics and Gynaecology. 2012;5(2):65-68.
7. Demographic and health survey. Nigeria Demographic and health survey; 2008. Available:http://www.measuredhs.com/pubs/pdf/GF15/GF15.pdf
   (Accessed 13th March, 2012)
8. Vital RM, Valenzuela JMS, Barraza RCL. Sacrococcygeal teratoma: Case report. Medwave. 2015;15(4):e6137.
   DOI: 10.5867/medwave.2015.04.6137
9. Abubakar AM, Nggada HA, Chinda JY. Sacrococcygeal teratoma in Northeastern Nigeria: 18-years of experience. Pediatr Surg Int. 2005;21:645-648.
10. Tanaree P. Delivery obstructed by sacrococcygeal teratoma. Am J Obstet Gynecol. 1982;142(2):239.
11. Krishan S, Solanki R, Sethi SK. Sacrococcygeal teratoma- role of ultrasound in antenatal diagnosis and management. J HK Coll Radiol. 2004;7: 35-39.
12. Drut RM, Fontana A, Grosso JJ. Mature presacral sacrococcygeal teratoma associated with a sacral "epignathus". Fetal Pediatr Pathol. 1992;12:99-103.
13. Bare JB, Abramowsky CR, Hayes LL, Shehata BM. Congenital immature teratoma of the central nervous system: Three case reports with literature review. Fetal Pediatr Pathol. 2007;26:109-118.
14. Sebire NJ, Fowler D, Ramsay AD. Sacrococcygeal tumours in infancy and childhood; a retrospective histopathological review of 85 cases. Fetal Pediatr Pathol. 2004;23:295-303.
15. Albert JR, Munoz MC, Castel V. Neonatal sacrococcygeal teratoma: Prolonged survival after malignant relapse. Pediatr Haematol Oncol. 1995;12:91-3.
16. Shahjouei S, Hanaei S, Nejat F, Monajemzadeh M, Khashab ME. Sacrococcygeal teratoma with intradural extension: Case report. Journal of Neurosurgery: Pediatrics. 2015;15(4): 380-383.
    DOI: 10.3171/2014.10.PEDS1445
17. Adzick N, Crombleholme T, Morgan M, et al. A rapidly growing fetal teratoma. Lancet. 1997;349:538.

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