

Plastic reconstruction of fetal anatomy using three-dimensional ultrasound and magnetic resonance imaging scan data in a giant cervical teratoma. Case report.

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Abstract

Cervical teratoma is a rare congenital tumor that tends to be large and is usually solid/cystic. Estimation of the degree of tracheal compression or distortion allows multidisciplinary planning for delivery and neonatal resuscitation. We present a case of prenatal diagnosis of cervical teratoma at 29 weeks of gestation. The use of a physical model from 3D ultrasound and magnetic resonance imaging improved the understanding of spatial relationships of fetal anomaly and the adjacent structures, permitting better parent counselling. This technology can be used for educational purposes and as a method for parents to visualize their unborn baby.

Keywords: prenatal diagnosis, cervical teratoma, 3Dultrasound, magnetic resonance imaging, physical model

Introduction

Cervical teratomas represent 3–5% of all teratomas, and their incidence may range from 1 in 20,000 to 1 in 40,000 [1] or from 1 in 35,000 to 1 in 200,000 live births [2], with a female-to-male ratio of 3:1 [3]. Teratomas involve at least one type of tissue from each of the three embryonic layers (usually accompanied by solid areas) and are most often of benign origin. Subsequently, for better viewing and diagnostic clarification, and depending on the fetal malformation observed on ultrasonography, the imaging examination was supplemented with magnetic resonance imaging (MRI).

Case report

A 34 year-old woman, G2P1 referred for II level ultrasound examination at 29w0d due to a severe fetal cervical mass with associated polyhydramnios. The scan was carried out using a Voluson E8[®] ultrasound apparatus equipped with a transabdominal volumetric RAB 4-8-D probe (General Electrical Medical Systems, Milwaukee, WI). The fetal growth parameters were as follows: BPD (biparietal diameter), 74.4 mm (73.8th centile for gestational age); HC (head circumference), 264 mm (21st centile for gestational age); AC (abdominal circumference), 236 mm (22nd centile for gestational age) and FL (femur length), 53.1 mm (21st centile for gestational age). A septated, dyshomogeneous cervical mass measuring 74 mm in its greatest width was confirmed and a prenatal diagnosis of giant cervical teratoma was prompted. The three-dimensional (3D) ultrasound in the rendering mode allowed the visualization of a large mass in the cervical region with externalization to the oral cavity (fig 1). MRI was performed using an apparatus with a 1.5-T scanner (Magnetom Avanto and Aera; Siemens, Erlangen, Germany). The protocol used was T2-weighted sequence (half-

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Fig 1. Three-dimensional ultrasound in the rendering mode shows a large mass in the cervical region with externalization to the oral cavity.

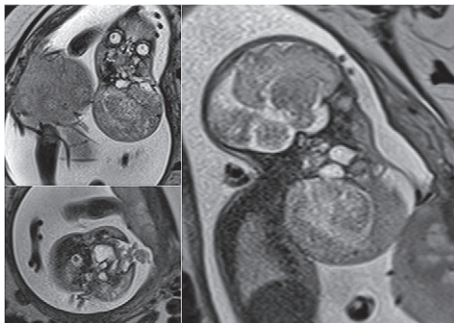


Fig 2. Magnetic resonance imaging T2-weighted sequence (coronal, axial and sagittal) shows a heterogeneous cervical mass extending from the oral cavity to the cervical region.

Fourier acquisition single shot turbo spin echo; repetition time shortest, time to echo 140 milliseconds; field of view = 300Y200 mm; matrix 256 _ 256; slice thickness 4 mm, 40 slices, and acquisition time 18 seconds in 3 planes of the fetal body). In addition, we applied 3D T2-weighted true fast imaging with steady-state precession (True FISP) sequence in the sagittal plane (repetition time 3.02 milliseconds, time to echo 1.34 milliseconds, voxel size 1.6 _ 1.6 _ 1.6 mm³, fractional anisotropy 70, parallel acquisition techniques 2) with 96 to 136 slices of thickness 1.0 to 1.6 mm and acquisition time of 26 seconds. The total duration of the examination did not exceed 40 minutes. The fetal MRI confirmed a heterogeneous cervical mass of 82 x 77 mm, extending from the oral cavity to the cervical region with a bulky external component in the transition from face to neck to the right with externalization of the lesion to the oral cavity. The inner portion of the lesion which was located in the oropharynx presented poorly defined boundaries, an infiltration of the adjacent soft tissues and the upper airway was not bounded (fig 2).

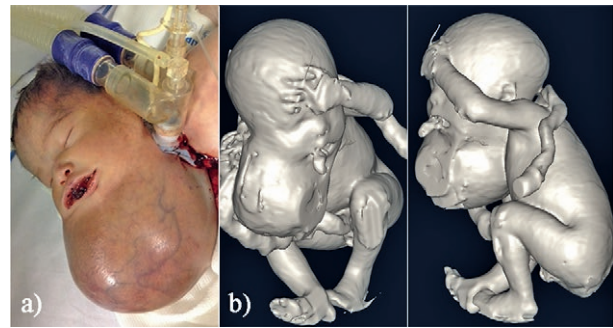


Fig 3. a) Neonate after tracheostomy; b) three-dimensional virtual reconstruction after magnetic resonance imaging showing cervical mass.

Cesarean section was planned at 34w0d and a male baby weighing 2755 g with 1-min and 5-min Apgar score of 1 and 4, respectively, was delivered. Multispecialist team for possible EXIT (ex utero intrapartum treatment) procedure was available at the time of birth. Tracheostomy was performed. The baby was then transferred to the NICU (neonatal intensive care unit) and post-natal surgery performed (fig 3).

Discussions

Thurkow et al [4] reported an extremely rare case of malignant teratoma of the neck, with mature and immature metastatic lesions in the lungs, in an immature fetus. Bauman and Nerlich [5] reported a metastasizing cervical teratoma of the fetus, and Shoenfield et al [6] found only six reports of malignant teratoma in the literature thus far. Teratomas of the head and neck account for only 6% of all teratomas [7], with the majority involving the sacrococcygeal region in the fetus and the newborn [8]. Teratomas are most frequently found in the anterior and midline areas of the neck, whereas lymphangiomas, hemangiomas and bronchial cysts are found in the posterior and lateral region [9]. Polyhydramnios that occur in approximately 30% of cases [9] and is usually associated with epignathus is most likely secondary to the obstruction of the fetal mouth and swallowing due to local mass effect, and causes severe respiratory compromise at delivery. In cases of giant cervical teratomas and/or giant epignathus, obstruction of circulation may also lead to high-output cardiac failure, with subsequent development of polyhydramnios and non-immune hydrops fetalis with certain intrauterine fetal death [10]. Hirose et al [11] concluded that fetal giant cervical teratoma in fetuses with hydrops is best managed through fetal resection, while in fetuses with airway obstruction, an EXIT procedure provides a generous amount of time to obtain airway control by intubation, tracheostomy, or, if necessary, by resecting the tumor on placental support (OOPS).

Although ultrasound is the diagnostic in these cases, 3D ultrasound and fetal MRI may enhance the accuracy of the antenatal diagnosis by improving location, extension. MRI provides more accurate information in cases of intracranial spread of the lesion and may aid in the selection of patients requiring treatment such as the EXIT and/or the OOPS procedure.

Werner et al [12] published a pictorial essay about the manufacturing physical models from 3D ultrasound, computed tomography (CT) and MRI scan data in fetuses with different malformations. They concluded that, despite being a more complex and relatively costly method, the techniques for 3D viewing show the possibilities for manipulating the physical characteristics of malformed fetuses along with their spatial relationships. Recently, Werner et al [13] described four cases of fetal cervical tumors, being three lymphangiomas and one teratoma, between 26 and 37 weeks. These authors developed a virtual bronchoscopy by means a 3D model created from MRI scan data. In all fetuses, fetal airway patency was demonstrated by virtual bronchoscopy and this was confirmed postnatally.

In our case, the plastic anatomic model provided by 3D MRI reconstruction enabled an important means of communication both for doctors and for the parents, and especially for visually impaired individuals, for whom the physical model would represent an important means for comprehending the fetal abnormality [14].

To construct the physical model from 3D ultrasound and MRI, the first step was to create the 3D virtual model. All the images generated through 3D ultrasound and MRI, were exported to a workstation in DICOM format. Following this, segmentation was performed by a 3D modelling technician, under supervision by the doctor responsible for the case using a digital high definition screen tablet (Cintiq Wacon, Tokyo, Japan). The 3D structure of the fetus was reconstructed by generating its surface using software with the capacity to convert the images obtained into numerical models (Mimicsv.12; Materialize, Leuven, Belgium). These reconstructed images were then exported using STL (standard triangular language) and were converted to the file extension "OBJ" touse 3D polygonal modeling software (Autodesk Mudbox, San Francisco, CA, USA). This software determined the volumetric surface of the image for analysis and subsequent topographic comparison. Following this, the 3D model was converted back to the "STL" extension and was exported to the Mimics software, which correlated the shapes and outlines observed from 3D ultrasound, MRI, and CT with the 3D images that were created. The final process to determine the physical model of the fetus consisted of guiding UV spot laser beams through a reservoir

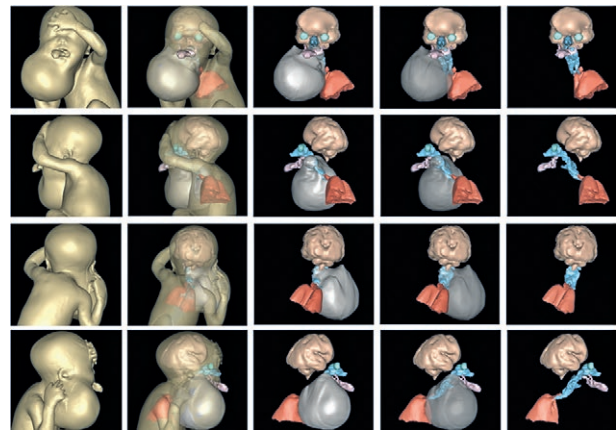


Fig 4. Three-dimensional virtual reconstruction (external and internal view of the fetus) showing the cervical mass invasion and the absence of the fetal airway patency.

of photosensitive resin to model the fetal shape based on the data stored in the 3D geometry software, which were sliced into individual transverse planes of predefined thickness, of between 0.1 and 0.2 mm [15] (fig 4).

In summary, we presented a case of a fetus giant cervical teratoma using a physical model from 3D ultrasound and MRI scan data. This manufacturing model allowed better understanding of the relationships between the cervical mass and the adjacent structures, permitting better parent counselling.

References

1. Nascimento GC, De Souza AS, Lima MM, et al. Intrapartum management strategies for congenital cervical teratoma: the EXIT procedure (ex utero intrapartum treatment). *Acta Med Port* 2007; 20: 221-227.
2. Smith NM, Chambers SE, Billson VR, Laing I, West CP, Bell JE. Oral teratoma (epignathus) with intracranial extension: a report of two cases. *Prenat Diagn* 1993; 13: 945-952.
3. Calderon S, Kaplan I, Gornish M. Epignathus: Case report of long-term survival with no surgery. *Int J Oral Maxillofac Surg* 1991; 20: 322-324.
4. Thurkow AL, Visser GH, Oosterhuis JW, de Vries JA. Ultrasound observations of a malignant cervical teratoma of the fetus in a case of polyhydramnios: case history and review. *Eur J Obstet Gynecol Reprod Biol* 1983; 14: 375-384.
5. Baumann FR, Nerlich A. Metastazing cervical teratoma of the fetus. *Pediatr Pathol* 1993; 13: 21-27.
6. Shoenfeld A, Ovadia J, Edelstein T, Liban E. Malignant cervical teratoma of the fetus. *Acta Obstet Gynecol Scand* 1982; 61: 7-12.
7. World Health Organization Classification of Tumors. Pathology and genetics of head and neck tumors. In: Barnes L, Eveson JW, Reichart P, Sidransky D. (eds.). IARC Press, Lyon 2005.

8. Pediatric Pathology. Berry CL. (ed.). Springer-Verlag, Berlin-Heidelberg- New York 1981.
9. Herman TE, Siegel MJ. Cervical teratoma. *J Perinatol* 2008; 28: 649-651.
10. Tonni G, De Felice C, Centini G, Ginanneschi C. Cervical and oral teratoma in the fetus: a systematic review of etiology, pathology, diagnosis, treatment and prognosis. *Arch Gynecol Obstet* 2010; 282: 355-361.
11. Hirose S, Sydorak RM, Tsao K, et al. Spectrum of intrapartum management strategies for giant fetal cervical teratoma. *J Pediatr Surg* 2003; 38: 446-450.
12. Werner H, Dos Santos JR, Fontes R, et al. Additive manufacturing models of fetuses built from three-dimensional ultrasound, magnetic resonance imaging and computed tomography scan data. *Ultrasound Obstet Gynecol* 2010; 36: 355-361.
13. Werner H, Lopes dos Santos JR, Fontes R, et al. Virtual bronchoscopy for evaluating cervical tumors of the fetus. *Ultrasound Obstet Gynecol* 2013; 41: 90-94.
14. Robiony M, Salvo I, Costa F, et al. Virtual reality surgical planning for maxillofacial distraction osteogenesis: the role of reverse engineering rapid prototyping and cooperative work. *J Oral Maxillofac Surg* 2007; 65: 1198-1208.
15. Werner H, Rolo LC, Araujo Júnior E, Lopes Dos Santos JR. Manufacturing models of fetal malformations built from 3-dimensional ultrasound, magnetic resonance imaging, and computed tomography scan data. *Ultrasound Q* 2014; 30: 69-75.