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Virtual bronchoscopy for evaluating cervical tumors of the fetus

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ABSTRACT

We report on four cases of fetal cervical tumor, comprising three lymphangiomas and one teratoma, evaluated by ultrasound and magnetic resonance imaging (MRI) between 26 and 37 weeks' gestation. The aim was to investigate the use of virtual bronchoscopy to evaluate fetal airway patency in each case. A three-dimensional (3D) model of the airway was created from overlapping image layers generated by MRI. The files obtained were manipulated using 3D modeling software, allowing the virtual positioning of observation cameras, adjustment of lighting parameters and creation of simulated 3D movies for analysis of a virtual path through the model. In all fetuses, fetal airway patency was clearly demonstrated by virtual bronchoscopy and this was confirmed postnatally. MRI with virtual bronchoscopy could become a useful tool for studying fetal airway patency in cases of cervical tumor. Copyright © 2013 ISUOG. Published by John Wiley & Sons, Ltd.

CASE SERIES

Four fetuses with a cervical tumor were evaluated in our tertiary imaging center (Clínica de Diagnóstico por Imagem) in Rio de Janeiro, Brazil, between September 2010 and March 2011. The cases included three with lymphangiomas and one with teratoma. All cases were diagnosed by ultrasound, and in all cases another ultrasound examination was performed on the day of magnetic resonance imaging (MRI) (Table 1). The ethical issues associated with this work were considered and approval was granted by our research ethics committee (CEPIFF: 0012/10).

The main outcomes of this study were the creation of three-dimensional (3D) virtual models of the airway of fetuses with cervical tumors and the performance of virtual bronchoscopy based on these models. The median gestational age at ultrasound diagnosis of cervical tumor was 24.7 (range, 22–27) weeks. MRI was performed between 26 and 37 weeks. In Case 2, MRI was performed twice, at 26 and 37 weeks.

All ultrasound scans included 3D imaging and were performed transabdominally using a high-resolution probe with harmonic imaging (4–8-MHz transducer, Voluson 730 Pro/Expert system, GE Medical Systems, Zipf, Austria).

MRI examination was performed using a 1.5-T scanner (Siemens, Erlangen, Germany). The protocol used was a T2-weighted sequence in the three planes of the fetal body (half Fourier acquisition single shot turbo spin echo (HASTE) with: repetition time (TR), shortest; echo time (TE), 140 ms; field of view (FOV), 300–200 mm; 256×256 matrix; slice thickness, 4 mm; acquisition time, 17 s; 40 slices)[1]. A T1-weighted sequence was also obtained for the three planes of the fetal body (TR, shortest; TE, 140 ms; FOV, 300–200 mm; 256×256 matrix; slice thickness, 4 mm; acquisition time, 17 s; 40 slices). Additionally, we applied 3D T2-weighted true fast imaging with steady state precession (truefisp) sequence in the sagittal plane (TR, 3.02 ms; TE, 1.34 ms; voxel size, $1.6 \times 1.6 \times 1.6$ mm^3; FA, 70; PAT, 2; acquisition time, 0.26 s). The entire examination time did not exceed 40 min[1,2].

A 3D model of the fetal airway was created from overlapping image layers from the MRI datasets using the software Mimics (Materialize, Leuven, Belgium). Contrast detection was applied to areas of interest in order to delineate the surface of the airway and the resulting 3D model was exported using the standard triangular language (STL) file format, then converted into an OBJ file using the 3D modeling software MAYA (Autodesk, San Rafael, CA,

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Table 1 Pregnancy, tumor and neonatal characteristics of the four cases of fetal cervical tumors in this series

Characteristic	*Case 1*	*Case 2**	*Case 3*	*Case 4*
Pregnancy				
Maternal age (years)	27	31	35	27
GA at US diagnosis (weeks)	27	24	22	26
GA at MRI + VB (weeks)	27	26/37	36	31
GA at delivery (weeks)	36	38	38	39
Tumor				
Tumor type	Lymph.	Tera.	Lymph.	Lymph.
Mean diameter on US† (mm)	63	60/94	64	35
Mean diameter on MRI (mm)	70	62/91	58	50
Invasion	No	No/No	No	No
Polyhydramnios	No	Yes/Yes	No	No
Neonatal				
Birth weight (g)	2850	3210	2910	3150
Apgar scores at 1, 5 min	7, 9	6, 8	7, 9	9, 9
Sex	Female	Male	Female	Female
Tracheostomy	No	No	No	No
EXIT	No	No	No	No
Pregnancy outcome	LB	LB	LB	LB
Postnatal surgery	No	Yes	Yes	No

*MRI performed at both 26 and 37 weeks. †US at time of MRI. EXIT, *ex-utero* intrapartum treatment; GA, gestational age; LB, live birth; Lymph., lymphangioma; MRI, magnetic resonance imaging; Tera., teratoma; US, ultrasound; VB, virtual bronchoscopy.

USA)[2]. The latter program allows the virtual positioning of observation cameras while working with multiple on-screen windows, and lighting parameters can be adjusted to optimize visualization. Finally, a path was plotted through the 3D model in order to create a simulated movie for analysis of the fetal airway: a virtual bronchoscopy[2].

The mean tumor diameters observed by ultrasound and MRI are presented in Table 1. Polyhydramnios was detected only in Case 2. Virtual bronchoscopy was successful in all cases, showing the absence of tumor invasion, and of tracheal distortion and compression (Figures 1–4, Videoclips S1–S4). The mean time required to perform virtual bronchoscopy after MRI was 2 hours.

All infants had normal karyotype and were delivered by Cesarean section after 36 weeks' gestation with good neonatal outcome (Table 1). *Ex-utero* intrapartum treatment (EXIT) procedure and tracheostomy were not necessary in any case. In two cases (Cases 2 and 3), postnatal surgery to remove the tumor was performed. In Cases 1 and 4, treatment with OK-432 was performed, with successful results to date.

DISCUSSION

Although the incidence of fetal cervical tumors is low, ultrasound is effective for identifying such tumors[3]. They tend to be large and can encircle essential structures,

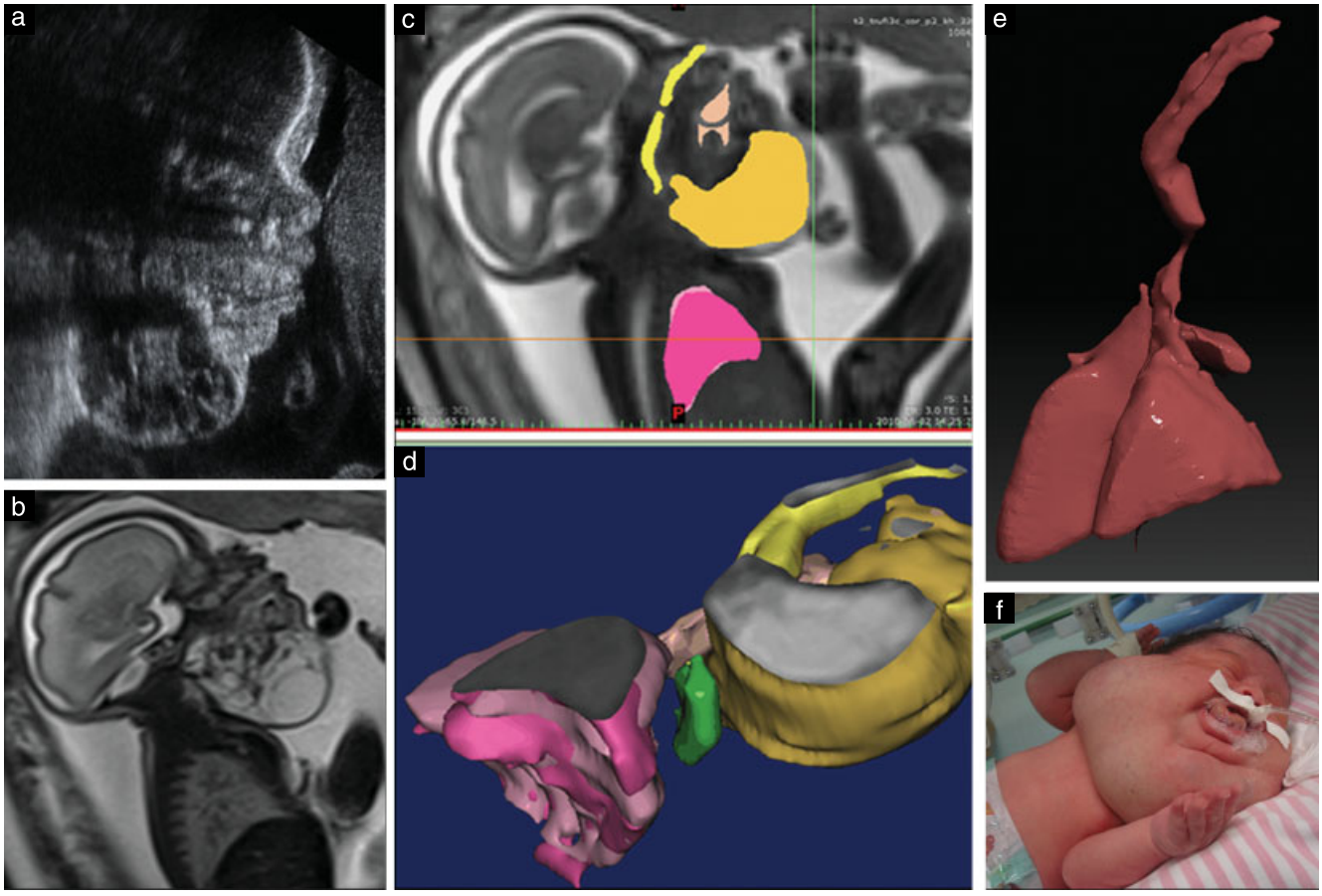

Figure 1 Case 1. Facial profile illustrated by ultrasound (a) and magnetic resonance imaging (MRI) (b), showing cervical lymphangioma at 27 gestational weeks. (c–e) Three-dimensional reconstructions of fetal airway and lungs obtained using MRI datasets. (f) Neonatal image.

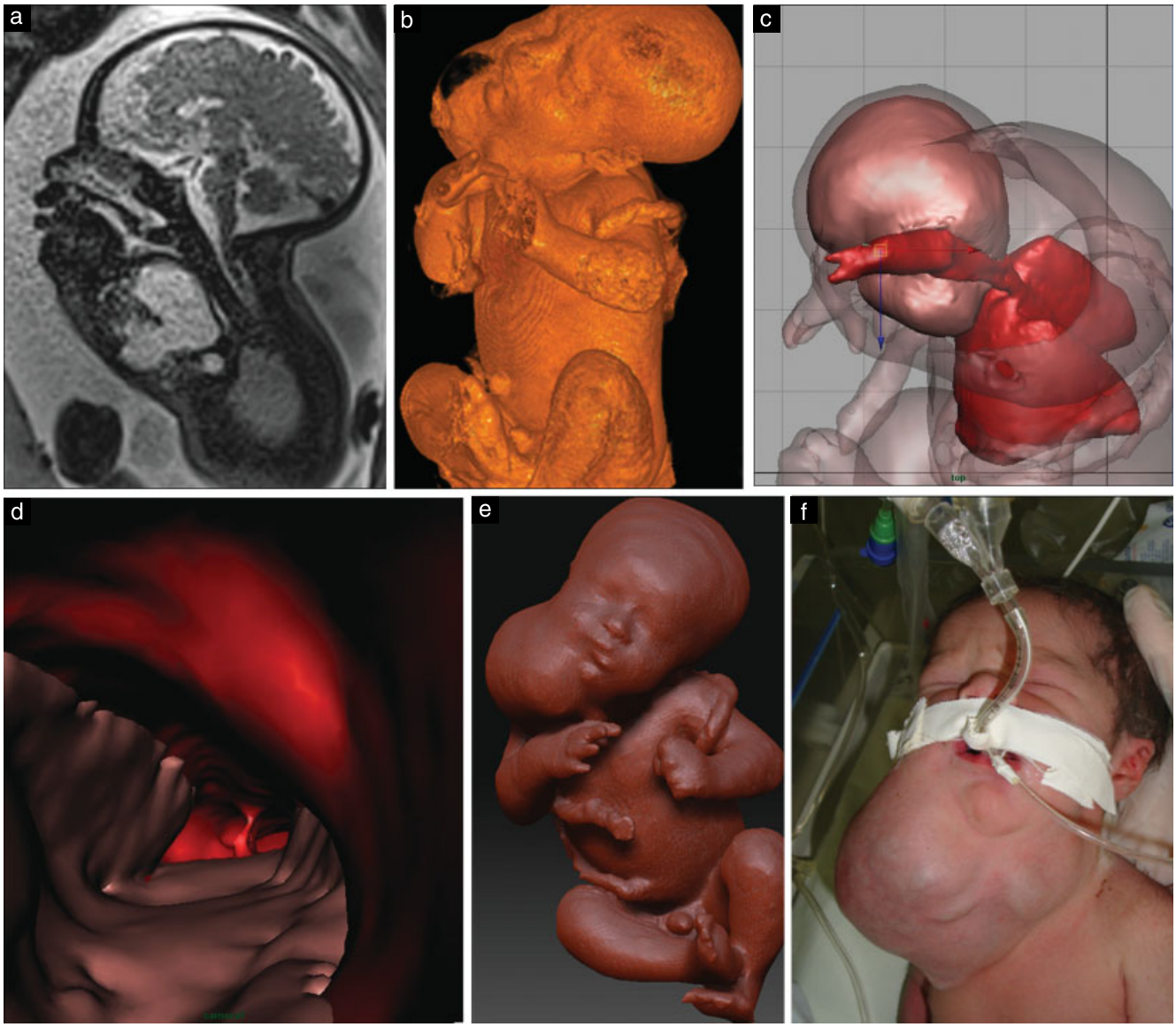

Figure 2 Case 2. (a) Magnetic resonance imaging (sagittal view) showing cervical teratoma at 37 gestational weeks, with three-dimensional (3D) reconstruction (b). (c) 3D view of airway path and (d) still image from virtual bronchoscopy, demonstrating patency of the airway. (e) 3D fetal model obtained by MRI. (f) Neonatal image.

such as the esophagus, thyroid and trachea, making it impossible for a neonate to breathe after birth. Estimation of the degree of tracheal compression or distortion allows multidisciplinary planning for delivery and neonatal resuscitation[4,5].

Three of the four cases in this series had cervical lymphangioma. Lymphangioma is a benign congenital malformation of the lymphatic system that has the potential to infiltrate surrounding structures. It constitutes approximately 5–6% of all benign lesions in childhood and adolescence[5], and occurs most frequently in the head, neck or axilla[6]. The prognosis depends on the presence of other associated features, such as skin edema, hydrops and polyhydramnios, abnormal karyotype, location and extent of the lesion[5].

The remaining case had a cervical teratoma, a rare congenital tumor that tends to be large and is usually solid/cystic. It occurs with an incidence of 1 in 20 000–40 000 live births, accounting for about 6% of all fetal teratomas. Airway obstruction in the newborn as determined by tracheal compression or occlusion is reported to be the reason for an 80–100% mortality rate in the neonatal period[4,5].

Recently, the ability to diagnose fetal tumors in the prenatal period has improved greatly because of technical advances in imaging[7]. Early diagnosis and determination of the pathological type of tumors may affect prognosis, making it possible for infants with fetal tumors to be saved. Ultrasound is the method of choice for fetal evaluation because of its relatively low cost, real-time capability and non-invasive nature. Cervical tumors are usually diagnosed in the second and third trimesters of gestation, being identified as multiseptate, thin-walled cystic masses in the fetal neck. MRI is a valuable complementary method to ultrasound for the diagnosis of fetal malformations[8]. Technical developments in this

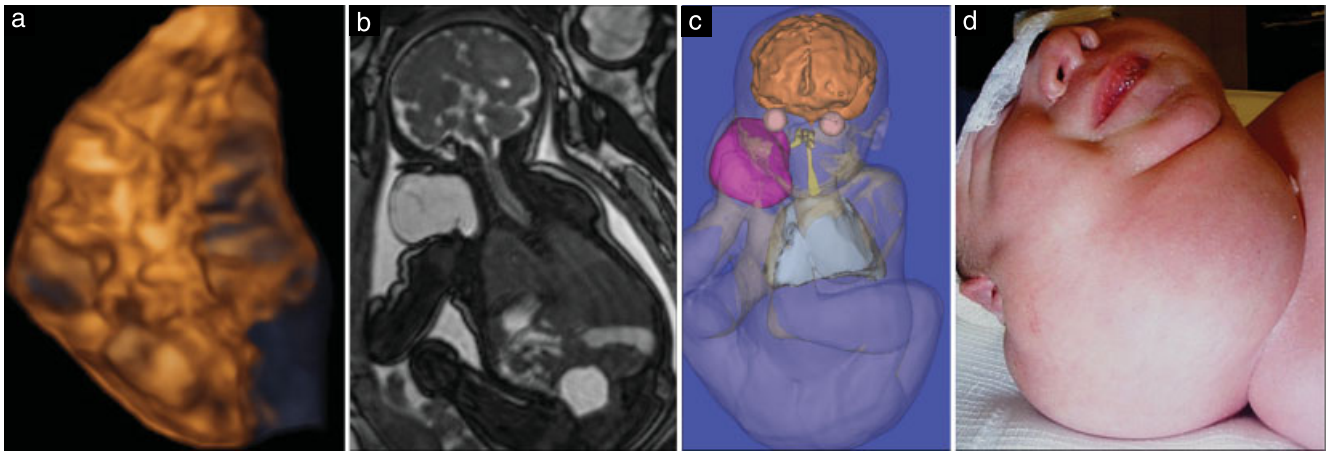

Figure 3 Case 3. (a) Three-dimensional (3D) rendered ultrasound image showing lymphangioma at 36 gestational weeks and (b) coronal T2-weighted magnetic resonance image (MRI) of the fetus. (c) 3D reconstruction obtained using MRI datasets showing tumor close to unobstructed airway path. (d) Neonatal image.

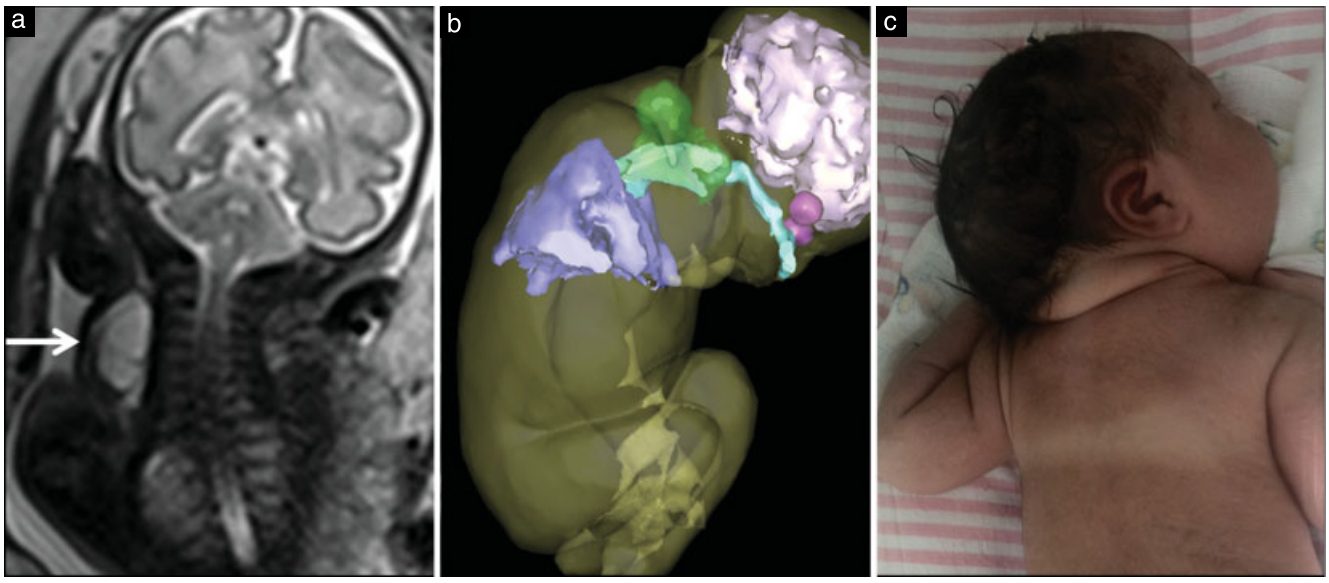

Figure 4 Case 4. (a) Coronal T2-weighted magnetic resonance image at 31 gestational weeks showing lymphangioma (arrow). (b) 3D airway path reconstruction. (c) Neonatal image.

imaging modality have greatly improved fetal imaging, with increasingly rapid sequencing and reduction of fetal motion artifacts[1,7].

The initial stage of management of a prenatally detected cervical tumor involves careful monitoring of the mother and fetus, with the development of a surgical plan[4,5]. Ideally, the planning of treatment will be undertaken by a multidisciplinary team of medical professionals, including a radiologist, a perinatologist and a pediatric surgeon. In some cases, surgery is performed immediately after the infant is delivered by Cesarean section, by an EXIT procedure. This gives the surgeon time to perform multiple procedures to secure the infant's airway while preserving the blood flow and gas exchange between the fetus and the placenta[5,9].

The term 'virtual bronchoscopy' refers to the generation and evaluation of 3D representations of the bronchial tree and surrounding structures derived from imaging sources other than a bronchoscope[10,11]. We have previously described virtual bronchoscopy based on 3D reconstructions of the fetal airway created using MRI datasets, which can provide better-defined images than can conventional imaging techniques[2]. Several software options are available that can generate accurate and realistic renderings of the bronchial tree, which can be used to provide confirmation of the findings on conventional MRI[12]. In this series, we have demonstrated the successful application of the technique in assessing patency of the airway in fetuses with cervical tumors.

In the cases included in this study, ultrasound was used to monitor amniotic fluid volume, tumor size and overall fetal wellbeing. MRI provided useful information about both the anatomy and extent of the tumors, and helped in the evaluation of adjacent structures. The data from MRI examinations were also used in the study of fetal airway patency by means of virtual bronchoscopy. These

examples show that MRI with virtual bronchoscopy in the third trimester of pregnancy can be an important tool for planning the mode of delivery and for studying fetal airway patency in fetuses with a cervical mass. In the four cases described here, the outcome was anticipated to be favorable because virtual bronchoscopy demonstrated an unobstructed airway. Further studies are required to demonstrate whether virtual bronchoscopy is useful in optimizing outcome when the fetal airway is compromised.

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SUPPORTING INFORMATION ON THE INTERNET

The following supporting information may be found in the online version of this article:

Videoclips S1–S4 Virtual bronchoscopy in Case 1 (Videoclip S1), Case 2 (Videoclip S2), Case 3 (Videoclip S3) and Case 4 (Videoclip S4). In Videoclip S4a, the virtual camera can be observed entering the airway of the fetus.