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## Retrospective chart review of 44 fetuses with cervicofacial tumors in the sonographic assessment



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#### ABSTRACT

Objectives: The aim of this retrospective study was to review and analyze ultrasonography examinations and follow-up of fetuses with cervicofacial tumors to develop bases for counseling specialist involved in perinatal treatment.

Methods: The study consisted of case series with chart review of 44 fetuses with cervicofacial tumors diagnosed in utero by ultrasonography. The study was carried in Department of Diagnosis and Prevention of Congenital Malformations, Medical University of Lodz in years 1998–2013. The analysis of the fetuses with cervicofacial tumors included assessment of fetal sonographic features, neonatal survival and in utero as well as perinatal treatments. The obtained data were analyzed by the standard statistical tests and the Pearson's Chi square test, statistical significance at p = 0.05.

Results: Cervicofacial tumors were detected at mean  $19\pm7$  weeks of gestation. Eighty-two percent of the fetuses were males. Lymphatic malformations followed by teratomas were the most common fetal tumors in the cervicofacial region. In most cases, fetuses with cervicofacial tumors had other abnormalities. Mortality rate in our case series was 43%. In utero treatment was introduced in 6 fetuses. In 4 neonates prenatal sonographic assessment revealed upper airway patency and EXIT procedure (ex-utero intrapartum treatment) was introduced.

Conclusion: Prenatal sonographic detection of cervicofacial tumor, in case of lymphatic malformations possibly as early as in the first trimester, in case of craniofacial teratomas, cervical teratomas, hemangiomas and thyroid tumors possibly as early as in the second trimester, and in case of epignathi possibly in the third trimester, permits planning further course of pregnancy as well as EXIT procedure before delivery.

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#### 1. Introduction

Cervicofacial tumors form a specific group of fetal neoplastic pathologies in the face as well as neck and nuchal areas. They form few pathological categories of tumors but their common presence may affect functions of upper respiratory and alimentary systems [1].

Methods like MRI (magnetic resonance imaging) and fetoscopy may be helpful in diagnosing cervicofacial tumors in

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fetuses but prenatal ultrasound examination is the most commonly performed and non-invasive diagnostic procedure in these cases [2,3].

Two-dimensional ultrasonography performed by an experienced diagnostician is considered to be a very accurate tool in the assessment of prenatal fetal cervicofacial anomalies [4]. 3D ultrasonography is of great value in visualizing the surfaces of the head and neck and presenting the results to parents or other physicians involved in the care of neonates and infants [5].

It is very demanding to offer counseling for specialists involved in perinatal treatment and plan proper management of the gestation in the cases of cervicofacial tumors diagnosed by prenatal ultrasound examination.

The aim of the study was thus to assess cases of fetuses with cervicofacial tumors, their sonographic features, concomitant

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abnormalities diagnosed by ultrasonography, gestation age at the time when tumors were detected and follow-up based on retrospective analysis.

#### 2. Material and methods

#### 2.1. Ethical considerations

Department's review board approved the methods of the present study. Authors have complied with the World Medical Association Declaration of Helsinki regarding ethical conduct of research involving human subjects. The study was conducted under the assumption that research findings would be kept anonymous.

The examinations were performed during the 1998–2013 period on the Philips/ATL HDI 5000 equipment and on GE Voluson machines in a single referral center for fetal disorders. The study included all the fetuses diagnosed prenatally with cervicofacial tumors in Department of Diagnosis and Prevention of Congenital Malformations that is both a tertiary teaching center and a referral center serving many millions of people of central part as well as other regions of Poland. All pregnant women were hospitalized in the Department with suspicion of cervicofacial tumors in their fetuses stated previously during commonly performed ambulatory gestational diagnostic sonographic examinations. Age of the pregnant woman, her past obstetric history, sex of the fetus and its age at the time of tumor detection was recorded in every case. Ultrasound features of tumors, including their locations, and initial diagnoses were assessed. Findings of the initial prenatal diagnosis were compared with the results of the examinations performed after the deliveries or terminations of pregnancies. Some fetuses underwent cytogenetic diagnostics. In addition, we included data concerning other fetal abnormalities diagnosed in prenatal ultrasound examinations as well as details of the methods of delivery or termination of pregnancy and procedures performed both in utero and during labor. The data were analyzed by the standard statistical tests and the Pearson's Chi square test, statistical significance at p = 0.05.

#### 3. Results

#### 3.1. General data concerning mothers and their obstetric histories

Mean age of the studied mothers was 28 years, SD 4.7 years, minimum 20 years, and maximum 36 years. Twenty-six women (59%) were primigravidas at low-risk pregnancies. Eighteen women (41%) were considered at high-risk pregnancies due to the previous medical history, previous miscarriages or stillbirths.

#### 3.2. Sonographic assessment

In three cases, an ultrasound examination revealed twin pregnancies with other fetus with no abnormalities (7%).

Thirty-two fetuses were males (73%) and eight fetuses (18%) were females. In four cases (9%) it was impossible to determine the

fetal sex based on the ultrasound examination. In those last cases, the babies were stillborn and postnatal examinations confirmed male stillbirths.

Thirty-one fetuses had normotrophic built for gestational age (70% – 22 fetuses with lymphatic malformation, 3 with epignathus, 3 with craniofacial teratoma, 1 with cervical teratoma, 1 with hemangioma and 1 with thyroid tumor). One fetus was hypertrophic (2% – 1 fetus with lymphatic malformation) and twelve were hypotrophic (28% – 10 fetuses with lymphatic malformation, 1 with epignathus and 1 with cervical teratoma). In 30 pregnancies the volume of the amniotic fluid was regular (68% – 26 fetuses with lymphatic malformation, 2 with craniofacial teratoma, 1 with hemangioma and 1 with thyroid tumor); in three cases (7% – 2 fetuses with lymphatic malformation and 1 with epignathus) there was oligohydramnios and in eleven fetuses (25% – 5 fetuses with lymphatic malformation, 3 with epignathus, 1 with craniofacial teratoma and 2 with cervical teratoma) – polyhydramnios.

The sonographic examinations in the Department of Diagnosis and Prevention of Congenital Malformations in all the cases confirmed diagnoses of fetal cervicofacial tumors stated previously during ambulatory gestational diagnostic sonographic examinations.

Prenatal ultrasound suggested lymphatic malformation in 33 fetuses (75%), oropharyngeal teratoma–epignathus in four (9%), craniofacial teratoma in three (7%), cervical teratoma in two (5%), hemangioma in one (2%), and thyroid tumor in one (2%).

Tumor was hypoechogenic in 33 cases of lymphatic malformations and in one case of cervical teratoma (a thick-walled hypoechogenic tumor). A hyperechogenic tumor was observed in all cases of epignathi, one craniofacial teratoma and one cervical teratoma. Hemangioma was a well-isolated tumor with very rich vascularization. Tumor was partly hyperechogenic and partly hypoechogenic in the thyroid tumor, and two craniofacial teratomas.

There was a statistically significant correlation between type of the tumor and its appearance in the ultrasound examination (hypoechogenic, hyperechogenic, partly hyperechogenic and partly hypoechogenic, tumor with increased vascularization) p < 0.05 (Table 1).

The findings of the prenatal sonographic diagnoses were confirmed by the results of the examinations performed after delivery or termination of pregnancy (clinical examination of the neonate or pathological examination of the dead fetus) in all cases.

In 10 cases with lymphatic malformations, non-immunological hydrops fetalis, hydrothorax ascites or all three conditions (23%) were recorded. Short long bones were present in 5 fetuses (11%), bilateral clubfeet in 1 fetus (2%), kidney anomalies in 5 fetuses (11%). Lymphatic malformations were found in these cases. One fetus with cervical teratoma presented herniation of posterior cranial fossa, as well as cerebellum and rhombencephalon anomalies (2%). In fetal echocardiographic assessment, cardiovascular anomalies including ventricular septal aneurysm of the aortic arch, common atrioventricular canal, functional tricuspid insufficiency and cardiomegaly were present in 17 fetuses (39% – all fetuses with lymphatic malformations).

**Table 1**Differential diagnoses by ultrasound in fetal 44 cervicofacial tumors.

Cervicofacial tumors	Number of cases (%)	Ultrasound features
Lymphatic malformation	33 (75%)	Hypoechogenic tumors in the cervical region.
Epignathus	4 (9%)	Hyperechogenic tumor within oral cavity and pharynx.
Craniofacial teratoma &	3 (7%)	Hyperechogenic tumors sometimes with hypoechogenic elements in different areas of the face or column
Cervical teratoma	2 (5%)	
Hemangioma	1 (2%)	Well-isolated tumor with very rich vascularization
Thyroid tumor	1 (2%)	Cervical partly hyperechogenic, partly hypoechogenic, well isolated tumor in the region of thyroid gland.

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