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Taussig-Bing Anomaly in a Premature Neonate with Isotretinoin Embryopathy

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Double outlet right ventricle (CCSV) is a congenital heart malformation characterized by large arteries coming out of the morphologically right ventricle. The concomitance of CCSV and subpulmoner ventricular septal defect (VSD) is named the Taussig-Bing anomaly, which is one rarely seen congenital heart malformation.[1]

Isotretinoin is a human teratogen that is known as retinoids. It may trigger anomalies in craniofacial, cardiological, and central nervous systems in utero exposure named isotretinoin embryopathy. Being exposed to isotretinoin between 2 and 5 weeks following conception appears to be critical; however, being exposed to isotretinoin at any period of pregnancy may pose a high teratogenic risk.[2]

The birth of an infant, 2550 g, occurred with a cesarean section due to fetal distress at gestational week 33. Because the mother had taken isotretinoin, 40 mg/day for cystic acne during the first month of her pregnancy, on the 11th day, further evaluation for undefined cardiac anomalies was referred.

Slight facial asymmetry, ptosis on the right eye, bilateral anotia with stenosis of the external ear canal, posterior helical pits, a narrow sloping forehead, micrognathia, depressed nasal bridge, and ocular hypertelorism were observed in the physical examination of the baby. As a result of auscultation, a grade 3/6 systolic murmur was revealed over the cardiac apex. We detected 2 large arteries coming out of the right ventricle on an echocardiogram. The aorta arises from the right ventricle anteriorly, and rightward of the pulmonary artery arcus was at left and in an elongated appearance. As arcus calibration decreased in the proximal and distal arcus localization, proximal arcus, and distal arcus were measured as 4.0 and 4.1 mm, respectively. The pulmonary artery was mildly larger than normal, branches were confluent, and peripheral vasoconstriction was not detected.

The patient was diagnosed with Taussig-Bing anomaly, subpulmonary ventricular septal defect, D-malposed great vessel artery relation, and patent ductus arteriosus, patent foramen ovale on echocardiography. Karyotype showed as a normal female in the chromosome studies. Inotrope and diuretic medications were used in the treatment of the newborn. Due to her poor neurologic outcome and prognosis of her illness, palliative procedures were not applied. As a result of congestive heart disease, the patient died on the 59th day.

Taussig-Bing anomaly is seen in 9 out of a hundred thousand births. The etiology is not known precisely. One study showed malformation in 21 fetuses out of 154 pregnant women exposed to isotretinoin during the fetal period, and 8 of these fetuses had conotruncal heart malformations and aortic arch anomalies.[3] Coberly[4] presented CCSV as an autopsy finding of the heart in a term baby with isotretinoin embryopathy.

Unluckily, isotretinoin (Accutane®) is reported to cause major congenital disabilities, the rate of which is approximately 30%. If a pregnant woman takes 0.5-1.5 mg/kg isotretinoin as a daily dosage per maternal body weight, this is considered teratogenic. To use isotretinoin, informed consent and a new dated pregnancy test with a negative result should be obtained

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from the patient, which accompanies contraception use in the course of treatment as a prerequisite. The findings related to our patient's history give extra support to the casual relationship that is proposed between Taussig-Bing anomaly and in utero isotretinoin exposure.[5]

In this article, we report a newborn having Taussig-Bing malformation and anotia resulting from exposure to isotretinoin during the first trimester, and assert that using isotretinoin (acne treatment) during pregnancy should be avoided.

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