Anomalous origin of single coronary artery with multiple heart malformations

S. Zachariah and R. Reif

From the Department of Pathology, Asaf Harofe Government Hospital, Tel-Aviv University Medical School, Zerifin, Israel

A newborn female infant is reported who had a single coronary artery arising from the innominate artery. In addition, a truncus arteriosus, mitral atresia, and single ventricle were found. Only one case with exactly the same malformation has apparently been published so far.

Single coronary arteries having their origins from the aortic or pulmonary sinuses have frequently been described. Origins of the single coronary artery from sites other than these are extremely rare, and to the best of our knowledge only three cases have been described to date (Mayer, 1827; Forster, 1847; Keeling, 1970). The purpose of this report is to put on record another case of rare anomalous origin of a single coronary artery arising from the innominate artery, in association with multiple heart malformations.

Case report

This 3-day-old female infant was born in Israel to parents of Moroccan extraction on 22 April 1972, with a birthweight of 1.67 kg. The mother’s pregnancy was complicated by threatened abortion and the infant was born prematurely. The delivery was normal. The mother had one living son and suffered a previous abortion (details of which are not available). The infant was apparently normal at birth and was rated 10 on the Apgar score. Because of her premature birth and low birthweight, she was transferred to a special premature care centre. On admission there the infant was found to be in respiratory distress with tachypnoea and cyanosis of lips and nail beds. She was placed in an incubator and resuscitative therapy with oxygen, intravenous fluids, antibiotics, and other supportive measures instituted. She failed to respond to any of these measures and died on the morning of 25 April 1972.

Necropsy

The infant weighed 1.52 kg and measured 44.5 cm in length. There was cyanosis of the face, lips, and nail beds. No external malformations were present. The pericardial reflexions were normal. There was cardiac enlargement involving the right atrium and the ventricle.
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With the rapid advance in diagnostic procedures that are instituted preoperatively, the surgeon today need not enter into totally unknown territory. A good knowledge of possible variations in anatomy is, however, indispensable in planning surgical procedures. Our case here demonstrates another variation in the spectrum of coronary artery anomalies, this type having been described only once before by Keeling (1970), and the type of malformation found in our case resembles exactly that found in his patient. In the case described by Mayer (1827) the coronary originated from the right carotid artery. In the case of Forster (1847) the single coronary emerged from the inferior aspect of the aortic arch.

Attempts have been made to classify the anomaly of the single coronary artery. Smith (1950) proposed three groups based on whether the artery was distributed according to one or both of the coronary arteries or was atypical. Our case falls into the third of these groups. Ogden and Goodyear (1970) devised an elaborate classification from a study of 95 cases according to basic distribution patterns. They based their classification on the number of major divisions of the single coronary artery and the subsequent course of these major branches. Our case, however, falls outside their classification. It might be interesting to note that in both cases with anomalous origin of the coronary artery the pregnancy was complicated by threatened abortion. The birth weight in Keeling’s case was 2·15 kg; in our case it was 1·67 kg.

Discussion

References


Requests for reprints to Dr. R. Reif, Department of Pathology, Asaf Harofe Government Hospital, Zerifin, Israel.
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