Case Report

UNUSUAL ORIGIN OF A SINGLE CORONARY ARTERY
A CASE REPORT

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A single coronary artery usually originates from one of the aortic sinuses, above the aortic sinuses or the pulmonary artery. Cases with a single coronary artery arising outside these usual sites are extremely rare. Autopsy examination of a male infant revealed a single coronary artery originating from the innominate artery, persistent truncus arteriosus and many other congenital anomalies. Five cases with unusual origin of a single coronary artery were found in the literature. All of them also had persistent truncus arteriosus and other severe anomalies.

Anomalies of the coronary arteries include abnormalities of number, origin and distribution. A single coronary artery is a relatively uncommon anomaly and is defined as that originating from an arterial trunk by one ostium and supplying the entire heart. It occurs as an isolated anomaly, but approximately one third of the cases reported accompanied other congenital malformations of the cardiovascular system. Cases of its unusual origin except from the pulmonary artery were reported 5 in the English and German literature.

This report describes a case of a single coronary artery originating from the innominate artery.

CASE REPORT

A male infant who was clinically diagnosed as harelip, cleft palate and congenital heart disease died on the thirty-sixth day of life.

Autopsy examination revealed a poorly nourished male infant with multiple congenital anomalies as listed on Table I. The cause of death was attributed to congestive heart failure due to congenital malformations of the cardiovascular system.

Fig. 1. Ventricular septal defect (thick arrow) and orifices of the pulmonary arteries (thin arrows) from the truncus arteriosus.

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Key Word:
Single coronary artery

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Examination of the heart. The weight after formalin fixation was 40 grams. The apex was rounded and composed of the left ventricular wall. The epicardium showed no abnormal findings. Both atria were dilated. The wall of the right ventricle measuring 9 mm, in thickness was thicker than that of the left ventricle. The myocardium was reddish brown in color. The endocardium was smooth and glistening. There were atrial septal defect (so-called ostium secundum defect) and ventricular septal defect (membranous type) (Fig. 1).

Single arterial trunk with tricuspid semilunar cusps arose from both ventricles superior to the ventricular septal defect. The right and left pulmonary arteries arose close together from the dorsal wall of the truncus arteriosus. The orifice of the right pulmonary artery located directly beneath that of the left pulmonary artery (Fig. 1). Ductus arteriosus was not found. No dimple suggesting the remnant of the coronary artery was present in the persistent truncus arteriosus.

A single coronary artery arose from the innominate artery and descended along the right posterolateral aspect of the truncus to the atrioventricular groove to divide into two main branches (Figs. 2 and 3). One of them ran along the atrioventricular groove to right as a normal right coronary artery, and it terminated as a posterior descending branch. The other branch courses behind the truncus to left and merged between the truncus and the left auricle. Then, it distributed similar to a normal left coronary artery (Fig. 3).

Histological examination revealed neither myocardial infarct nor fibrosis.

DISCUSSION

In 1950 Smith found 43 cases with a single coronary artery in his review of the literature, two of which he discarded, and added two his own cases. He classified those into three types according to the distribution of the single coronary artery. The first type includes those cases with the single coronary artery which follows the course of only the normal right or left coronary artery. The second type represents those cases in which the single coronary artery arising by one ostium divides so that branches are present in the distribution of both the right and left coronary arteries. The third type includes those cases in which the distribution of the single coronary artery is so atypical that it can not be compared
TABLE I  ANATOMICAL DIAGNOSIS

1. Multiple congenital malformations
   1) Persistent truncus arteriosus
   2) Single coronary artery arising from the innominate artery
   3) Ventricular septal defect
   4) Atrial septal defect
   5) Harelip
   6) Cleft palate
   7) Pterygium colli (webbed neck)
   8) Lowering of the hairline on the forehead
   9) Hypoplasia of the auricles
  10) Undescended testes
  11) Duplication of the bilateral renal pelves and the left ureter
  12) Meckel's diverticulum
2. Miscellaneous
   1) Congestion of the organs
   2) Uric acid infarction of both kidneys

 TABLE II

<table>
<thead>
<tr>
<th>Author</th>
<th>Site of origin</th>
<th>Other cardiovascular anomalies</th>
</tr>
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<tbody>
<tr>
<td>1) Mayer² (1827)</td>
<td>Right carotid artery</td>
<td>Cor biloculare</td>
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<tr>
<td></td>
<td></td>
<td>Persistent truncus arteriosus</td>
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<tr>
<td></td>
<td></td>
<td>Anomalous origin of aortic arch branches</td>
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<tr>
<td>2) Forester³ (1847)</td>
<td>Inferior aspect of aortic arch</td>
<td>Cor biloculare</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Persistent truncus arteriosus</td>
</tr>
<tr>
<td>3) Clark⁴ (1848)</td>
<td>Lower part of subclavian artery</td>
<td>Cor triloculare</td>
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<tr>
<td></td>
<td></td>
<td>Persistent truncus arteriosus</td>
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<tr>
<td></td>
<td></td>
<td>Patent foramen ovale</td>
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<tr>
<td>4) Grosse⁵ (1919)</td>
<td>Innominate artery</td>
<td>Cor triloculare</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Persistent truncus arteriosus</td>
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<tr>
<td></td>
<td></td>
<td>Atresia of mitral ostium</td>
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<tr>
<td>5) Keeling¹ (1970)</td>
<td>Innominate artery</td>
<td>Cor triloculare</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Persistent truncus arteriosus</td>
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</tbody>
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with the right or left coronary artery. Longenecker et al. reviewed 25 cases since 1950 and added type 4, which included cases with insufficient data. In 1970 Ogden and Goodyer collected 142 cases of a single coronary artery found in the literature and their own case survey, and classified them into five basal types on the basis of the anatomical distribution patterns. A single coronary artery usually originates from one of the aortic sinuses, above the aortic sinuses or pulmonary artery. Cases with a single coronary artery arising outside these usual sites have been rarely reported. We could find five such cases in the English and German literature (Table II). Sites of origin were the innominate artery, lower part of the subclavian artery, the right carotid artery and inferior aspect of the aortic arch. All of them accompanied truncus arteriosus and other severe congenital cardiac anomalies.

A single coronary artery as an isolated anomaly does not preclude the longevity and the oldest reported case was 84 years of age. It might, however, predispose the myocardial infarction in the presence of acquired coronary artery disease. Anomalous origin and distribution of the coronary arteries present problems in the cardiovascular surgery, since the major branches may be accidentally cut in the operation when the surgeon is unaware of these variations.

Collet and Edwards classified persistent truncus arteriosus into four major types on the basis of the embryologic development of the pulmonary arteries from the sixth aortic arches. Our case corresponds to the type 2 (subtype 1) of their classification. It was already mentioned that the ostium of the left pulmonary artery was often almost directly above that of the right pulmonary artery in this type of persistent truncus arteriosus.

**Summary**

A single coronary artery originating outside the aorta or pulmonary artery is extremely rare. A case is described that had persistent truncus arteriosus, atrial and ventricular septal defects, a single coronary artery arising from the innominate artery and many other congenital malformations.

**REFERENCES**