CASE REPORTS

A CASE OF SINGLE CORONARY ARTERY WITH STEREOGRAPHIC DEMONSTRATION OF THE ARTERIAL DISTRIBUTION

BY

P. J. D. SNOW

From the University Department of Cardiology, Manchester Royal Infirmary

A single coronary artery occurring as an isolated congenital defect is rare. It is unfortunate that some of the previous descriptions do not permit an accurate anatomical classification which would clarify the embryology of the anomaly. It is the purpose of this report, therefore, in recording a further case, to demonstrate the distribution of the vessel by stereoscopic radiographs, a method not previously used.

Case Report

A sixty-two-year-old male was admitted under the care of Professor Bramwell, with a peripheral neuropathy and congestive heart failure of six months' duration. The neuropathy and cardiac failure were thought to be related, but no diagnosis was established. Despite all treatment, the condition steadily deteriorated, death occurring six weeks after admission. At necropsy the heart weighed 600 g., both ventricles being hypertrophied. Only one coronary orifice could be found. This was situated in the right coronary sinus and had a lumen of 4 mm. It divided, 1 cm. from its origin, into two large branches passing anteriorly and posteriorly and one very small branch passing laterally towards the right ventricle which was thought to be a conus artery (Schlesinger *et al.*, 1949). In order to establish the presence or absence of an abnormal collateral circulation, cannulae were passed down the main branches separately, but could not be passed into the smallest division: radiopaque medium was then injected using Schlesinger's (1938) technique. Mass injected into one branch issued from the other when the respective pressures were raised and lowered, indicating an anastomosis of abnormal size between these vessels. Stereoscopic radiographs of the entire heart were taken, after which the heart was unrolled and again radiographed. Stereodiagrams made from the original radiographs are shown in Fig. 1 together with a normal coronary tree for comparison.

Study of these radiographs showed that the posterior division corresponded to the right artery and, in addition to the right ventricle, supplied a considerable part of the posterior wall of the left, representing to a striking degree the preponderant right coronary pattern of Schlesinger (1940). The anterior division passed downward through the upper part of the septum, between the pulmonary artery and aorta, to reach the interventricular groove: here it divided into upper and lower branches corresponding to circumflex and anterior descending branches. No occlusions were visible on the radiographs and dissection of the vessels revealed only moderate atheroma. The abnormal anastomosis was attributed to ventricular hypertrophy.

Histological examination of the myocardium revealed that the enlargement was due to extensive amyloid deposits. These were also present in the peripheral nerves, tongue, and spleen. Primary amyloidosis affecting the peripheral nerves and myocardium and severe enough to cause congestive failure is very uncommon and will be the subject of a separate report.

Discussion

Smith (1950) defined a single coronary artery as one arising from an arterial trunk by a single ostium and supplying the whole heart regardless of distribution. He was able to collect 45 such cases, including two of his own. Thirteen of these were known to have other congenital cardiac defects.

The origin of the condition is of some embryological interest, and both Smith (1950) and Krumbhaar and Ehrich (1938) recognized three types: (1) those in which a single artery after supplying one

A

B

FIG. 1.—Stereodiagrams showing the coronary tree of the case described (A) and of a normal heart (B). Many observers are able to view stereographic pairs without a stereoscope. The page should be held at about fifteen inches from the eyes and the visual accommodation relaxed as if to view an object at infinity, so that the visual axes are parallel.

If the page is now brought slowly into focus, each eye will view its own member of the stereographic pair: three images are perceived, of which the centre one is due to the fused pair. This will appear three dimensional.

ventricle, continues on to supply the other, (2) those in which a common trunk divides so that the distribution of both left and right arteries are represented, and (3) those in which the distribution was so atypical that it was comparable with neither left nor right coronary artery.

Although Hyrtl (1882) accepted only cases of the first type as examples of absent coronary

artery, it does not follow, as he suggests, that the rest are merely anomalies of origin. The present case belongs to group (2), in which it is uncertain if the apparently missing artery is really absent, or merely has an anomalous origin. It has been suggested that a branch of the solitary vessel may take on the course and distribution of the absent artery thus simulating a misplaced origin (Sanes, 1937; Speer, 1938; and Roberts, 1947). Both types may be represented in this group and a more detailed anatomical classification is likely to assist in the distinction. Clear depiction of the coronary tree in each case is of obvious importance, yet no suitable method for demonstrating or recording the anomaly has previously been used. Verbal descriptions, in an attempt to avoid ambiguity, become lengthy and involved, and two-dimensional diagrams are not always satisfactory. A more graphic method is obviously desirable and it is suggested that stereoscopic radiography of the injected heart is the most accurate.

As an isolated congenital defect, a single coronary artery does not significantly affect the chance of survival. In Smith's series, of 31 patients with no known associated defects, only 2 did not reach adult life, dying of bronchopneumonia at three days and three months old. Of the remaining cases in which the ages are known, 10 died before the end of the fourth decade and 19 before the end of the sixth; 8 others survived over the age of sixty. When associated with other congenital defects, however, only one out of thirteen reached adult life, the defect in this case being a bicuspid aortic valve. The majority of these cases possess a grossly atypical arterial distribution and belong to group (3). Acquired cardiac lesions were found in 9 of the 30 adult cases, including 4 with coronary occlusion and infarction. Roberts and Loubé (1947) believed the effects of occlusion to be severe since available anastomoses had already been utilized to compensate for the defective arterial distribution. A more likely explanation is the lack of a second coronary artery to provide a collateral circulation.

Summary

A case of single coronary artery occurring as an isolated congenital defect is demonstrated by radiopaque injection and stereoscopic radiography. The developmental types of single coronary artery and the effect of the anomaly on survival are discussed.

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