Coronary artery disease is a pediatric curiosity; even the well-known aberrant origin from the pulmonary artery is rare. More rare is the condition of arterial calcification leading to death from cardiac ischemia in infants of which some 40 cases have been reported (Hunt and Leys, 1957). Still rarer are cases of coronary aneurysm in children, some 12 being recorded by 1957 (Crocker and others, 1957), and of these only three were in infants. Of these three, one was in a child with multiple cardiac anomalies, but the other two bore a remarkable resemblance both clinically and pathologically to the case described here.

Case Report

Melanie C. was the first child of healthy parents. Immunisation procedures had recently been started but she had suffered no illness whatever till 6.10.61 when aged 6 months. She then became fretful, feverish and vomited. She seemed to resent being touched. Next day she was no better, in spite of chloramphenicol and developed a dusky red blotch rash on her face and body. In spite of the vomiting her appetite remained good, and there was no photophobia.

She was admitted on 7.10.61 as there was no improvement. She had conjunctivitis and an infected throat and the rash was still present, but no purpura had developed. No cardiovascular abnormality was noted, and her pulse (160/min.) and respiration rate (60/min.) were thought to be compatible with her temperature (105°F). Blood culture was negative, blood picture showed moderate neutrophil leucocytosis (79% of 14,000 with pronounced left shift); ESR was 35 (Wintrobe). Chest X-ray was normal.

The clinical diagnosis was septicaemia, so antibiotics were given in series. As response was poor, prednisolone was added in a dose of 30 mg. on the first day, then 20 mg. daily (Weller, 1956). Temperature tended to subside during the first two days on steroid treatment but then recurred; it settled gradually on methicillin and tetracycline but slowly returned during the third week of illness. All treatment was stopped during the fourth week with very little effect on the temperature or general condition. Throughout the whole period the child continued to take feeds well and gained nearly 2 lbs. in weight until 28.10.61 when weight loss began. The pulse and respiration rates remained high but at no time was any heart condition suspected. The white count rose, reaching a peak of 39,000 on 17.10.61, and the left shift persisted with a number of smear cells. The leucocytosis was subsiding by the end of October, by which time mononuclear cells predominated. At intervals the child vomited without good explanation. The abdomen was sometimes distended but the liver was never conspicuously large.

On 15.10.61 a coughing attack led to sudden collapse, with skin white and marbled. Recovery was fairly rapid but there were moist sounds in the lungs for a few hours. By 7.11.61 the child was thought to be reasonably well except for occasional vomiting. However, on 8.11.61 two attacks of severe breathlessness occurred during the night and in the second the child died.

Autopsy

A well nourished female infant. Body weight: 7595 g. and length: 70 cm.

Cardiovascular (Heart 95 g.). A large heart, on the surface of which there were two tortuous surfaces, each corresponding to the course of a coronary artery. (Fig. 1.) There was hypertrophy and dilatation of the left ventricle. No valvular abnormalities. Musculature good. On opening the left coronary artery it was normal for 5 mm., then for the next 40 mm. it was grossly dilated and filled by organising thrombus. The right coronary artery was normal for 2 mm. only and

ANEURYSM OF CORONARY ARTERIES IN AN INFANT

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spreading through the remains of the media and adventitia into the peri-adventitial tissue. The whole lesion was widely infiltrated by polymorpho-nuclear leucocytes and eosinophils and, to a lesser extent, plasma cells. (Fig. 2).

Discussion

These appearances are virtually identical with the two similar cases described by Crocker and others (1957).

It would seem reasonable to conjecture that one coronary artery became occluded at the time of the collapse on 15.10.61 and the other on the day of death. No conclusion could be reached either from the clinical findings or the post-mortem appearances regarding the cause of this illness, but it is interesting that a reasonably large dose of steroids had no certain effect on the course of the illness as one might have expected if the basic abnormality had been that of a 'collagen disease'.

The two similar cases of Crocker and others (1957) were boys dying at 4 months and 8 months of age. Each fell suddenly ill with fever, blotchy rash, conjunctivitis, sore throat, tachycardia and leucocytosis. One had a large heart on X-ray and the E.C.G. pattern suggested myocardial infarction. He also had an eosinophilia. In spite of treatment the illness progressed with cardiac murmur, ectopic beats and progressive cardiac enlargement.

The illness of the second started with irritability, conjunctivitis, pharyngitis and a blotchy rash that waxed and waned for two weeks. There was a neutrophil leucocytosis with left shift which subsided with treatment of an incidental urinary infection. The child went home, only to return after two months in heart failure which had developed rapidly over two days.

Summary

A case of aneurysmal coronary arteries with terminal bilateral occlusion in an infant is described. Two remarkably similar cases have been reported previously.

REFERENCES


CONGENITAL ABSENCE OF PAIN

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The ability to react normally to painful or noxious stimuli serves to protect the body from damage. Interference with pain perception can occur from a variety of lesions at different anatomical levels from peripheral nerve endings (as in progressive sensory radicular neuropathy) to cerebral cortical disorders (as in congenital absence of pain). In the latter condition there is no demonstrable lesion
Aneurysm of Coronary Arteries in an Infant

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