Spinal Subarachnoid Haemorrhage in Patient with Coarctation of Aorta

**Br. med. J., 1967, 4, 278-279**

Patients with coarctation of the aorta are liable to develop cerebrovascular complications (Wolman and Sheiden, 1927; Tyler and Clark, 1958). Intracranial haemorrhage with or without a demonstrable aneurysm occurs in some 12% of untreated cases (Reifenstein et al., 1947). Spinal complications are much rarer (Weenink and Smilde, 1964). They are believed to be ischaemia resulting from reduced blood flow to the lower portion of the cord, and from vascular abnormalities associated with the collateral circulation. This paper describes the occurrence of paraplegia after spinal subarachnoid haemorrhage in a patient with coarctation.

**Case Report**

A 44-year-old woman was admitted to Sydney Hospital in May 1964 with confusion, severe pain between the shoulder blades, and loss of power in the legs. At the age of 7 she had been known to have a heart murmur, and at 12 she had been diagnosed as having "rheumatic carditis," the illness consisting of slight fever, chest pain, occasional involuntary writhing movements, and sore joints. At 14 she had a systolic blood pressure of 180 mm. Hg, for which she was not treated. During her second pregnancy, at the age of 25, she had an episode of loss of consciousness while in hospital for an antepartum haemorrhage. On regaining consciousness she had no neck stiffness or headache and no residual neurological symptoms. She was told that she had had a "cerebral haemorrhage," and caesarean section was carried out with delivery of a normal infant. At the age of 32 a chest x-ray film taken during a mass survey showed rib notching and an enlarged heart. Clinical examination revealed a blood pressure of 220/90, with femoral pulses greatly diminished in intensity and delayed in comparison with the radial pulses. The second heart sound was increased in intensity, and from the more usual adult picture. Though most reports of familial dysautonomia have appeared in the American literature, there are three reports in British journals (McKusick, 1958; Hutchison and Hamilton, 1962; Keith, 1965) and one in Australia (Burke, 1966), and other cases in the United Kingdom are known.

My thanks are due to Professor Victor A. McKusick for his kind encouragement and permission to publish this case.

**PETER W. BRUNT,* M.RB., M.R.C.P., Division of Medical Genetics, Department of Medicine, Johns Hopkins University, Baltimore, Maryland, U.S.A.**

*In receipt of a Wellcome Trust travel grant.*

**References**


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with hypertrophic changes on the medial aspect of the joint and with shift of the patella laterally.

Neurological examination revealed hypotonia and absent deep tendon reflexes. Cerebellar function appeared intact. Light touch and position sense were normal, but vibration sense was somewhat diminished and pin sensation greatly reduced. Temperature sensation also appeared to be diminished.

Figs. 2-4 show the right knee, the radiological changes in the left knee being similar but less severe. At age 11, soon after the initial symptoms in the right knee, there was considerable destructive change involving principally the lateral aspect of the epiphysis (Fig. 2). A year later (Figs. 3 and 4) soft-tissue calcification and ossification were visible. There was subluxation and irregularity of the femoral condyles. The ossification was compatible with PelliGrini-Sieda fracture, indicating recurrent trauma to the joint.

**Comment**

In attempting near complete ascertainment of cases in the North American continent we (McKusick et al., 1967) have obtained first-hand information on 164 families (200 patients). Among the 60 children examined in detail we have found two further young adults with similar joints—one with a painless, acutely swollen, disorganized knee joint and the other with a painless arthropathy of the shoulder.

Charcot joints occurring before closure of the epiphyses are rather uncommon and present different radiographic findings from the more usual adult picture. Though most reports of familial dysautonomia have appeared in the American literature, there are three reports in British journals (McKusick, 1958; Hutchison and Hamilton, 1962; Keith, 1965) and one in Australia (Burke, 1966), and other cases in the United Kingdom are known.

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blood-clot filling the subarachnoid space with no abnormality of the cord. One abnormal dilated vessel was found and tied off, and there was no further active haemorrhage.

Postoperatively the pain in the back disappeared, but she could move only her toes and regained no other active movement in the legs. When last seen in 1966 her blood pressure was over 300/120, and there were signs of inadequate blood supply to the legs. She was otherwise well, using a wheel-chair for ambulation.

**COMMENT**

The aneurysmal dilatations of collateral vessels in coarctation of the aorta are well documented in the literature (Boyd and Werblow, 1937; Cleland et al., 1956). Histologically the walls of these dilated vessels are deficient in muscle and elastic tissue, and in some places they are composed solely of intima and adventitia (Cleland et al., 1956). These changes may affect the spinal arteries as well as other collaterals. Wyburn-Mason (1943) recorded a total of seven cases in which enlarged tortuous spinal arteries with or without local aneurysm formation were found in cases of coarctation. Weenink and Smilde (1964) found a dilated anterior spinal artery at necropsy in a patient who presented with paraplegia of sudden onset.

The combination of spinal subarachnoid haemorrhage and coarctation of the aorta has, to my knowledge, been reported only once before (Wyburn-Mason, 1943). This patient had temporary weakness of one leg after the haemorrhage. Henson and Croft (1956), in their review of spinal subarachnoid haemorrhage, found permanent paraplegia directly due to the haemorrhage to be a rare occurrence.

**True Heartburn**


Heartburn, while sounding like a pathological description, is a descriptive term common in dyspeptic symptomatology—a symptom that most people experience and one that is not normally regarded with any great apprehension, nor does it carry a connotation of cardiac pathology. However, exceptions occur and the following case is one of them.

**Case Report**

The patient, a bachelor aged 45 living with his mother, for many years had been a seaman, but during the previous two years had been working as a coffee roaster. On 7 January 1967 while sitting at home with his mother and feeling depressed as a result of delusions of persecution, he placed a poker in the fire and watched it heating until it was red. When he thought that the maximum heat had been generated on the tip of the poker he placed it over his heart and pushed hard. With little or no sensation of pain he observed that the poker passed quite easily into his chest; he felt faint and fell on the floor. His mother summoned the neighbours for assistance, and, as it was seen that, though pale, he was still alive, the ambulance was summoned. He was admitted to the Western Infirmary in the early hours of 8 January.

Observation from a distance showed that the handle of the poker was projecting from the fourth left interspace, about 8 cm. from the midline (Fig. 1). Oscillation of the poker indicated that the heart was heating and the pulse rate was 120 per minute. His blood pressure was 80/50 mm. Hg and he was pale, perspiring, but not in any respiratory embarrassment. On closer observation it was seen that there was a ring of charring around the entrance wound, and that there was little or no haemorrhage. A lateral x-ray film taken with a portable machine confirmed what was already obvious, that the larger portion of the poker was in the left thoracic cavity.

Fig. 2 shows how the poker had passed through the pericardial cavity, the tip of the poker resting about 2 cm. from the posterior thoracic wall. There was no evidence of injury to the lung, but both entrance and exit wounds in the pericardium showed evidence of charring, and when the bridge of pericardium was divided it was seen that an area of the left ventricle, 2 by 4 cm., was whitened and clearly had been burnt as the hot poker passed over it. There was no active bleeding, and the poker was withdrawn without causing any further damage. Cardiac action seemed normal, and, as the patient’s blood pressure had risen to normal levels after the transfusion of 2 pints (1,140 ml.) of blood, it was decided that no further action should be taken about the burning of the myocardium. As

Paraplegia with loss of pain and temperature and retention of touch and joint position sense is characteristic of the anterior spinal artery syndrome (Peterman et al., 1958; Henneaux, 1960). The mechanism of its production in the above case must remain in doubt. The anterior spinal artery may have been compressed by clot. There may have been arterial spasm (Paulson, 1963), or, most likely, a portion of the cord previously supplied by the ruptured vessel became ischaemic.

My thanks are due to Associate Professor Solomon Posen and Mr. C. R. Latham for allowing me to see the patient and for their continuing interest.

A. B. WATSON, M.B., B.S.,
Resident Medical Officer, Royal Newcastle Hospital,
Newcastle, New South Wales, Australia.

**REFERENCES**