

## DOUBLE AORTIC ARCH IN AN INFANT OF THREE MONTHS

A. B. ROSENBERG, B.Sc. (STELL.), M.B., CH.B. (CAPE TOWN), *Senior Medical Officer, Department of Paediatrics, Somerset Hospital*; AND R. P. HEWITSON, F.R.C.S. (ENG.), F.R.C.S. (EDIN.), *Thoracic Surgeon, Somerset and Red Cross War Memorial Children's Hospitals*

Of the various causes of respiratory difficulty in infancy, one of the less common, but nevertheless remediable, is tracheal obstruction due to abnormalities of the great vessels. The following case illustrates some of the aspects of this possibility that are worth bearing in mind when dealing with any child who wheezes.

## CASE REPORT

A Coloured infant, aged 3 months, was admitted to the Somerset Hospital on 17 September 1965, because of a history of coughing for 5 days and for gastroenteritis.

*Past History*

The birth was a full-term normal delivery to a mother with 4 children, who are all well. The child was born at home, and was given BCG at the regional clinic. Before this present complaint, there had been no complaints and the child took all its feeds well, with normal bowel action.

*General Examination*

The child was clinically anaemic, tachypnoeic, and weighed 8 lb. 11 oz. on admission. The pulse rate was 110/min. and temperature (rectal) 99.4°F. Examination of the respiratory system showed a central trachea, good air entry in both lungs, despite diffusely scattered crepitations and a fair amount of bronchospasm.

All other systems appeared to be normal. Haemoglobin 9.5 G/100 ml., white cell count 11,900/cu.mm. X-ray of the chest at this stage showed partial pneumonic consolidation in both right upper and left lower lobes. There was also patchy consolidation in the right lower lobe. Treatment included a broad-spectrum antibiotic and a bronchodilator.

Investigations done included a Heaf test, PPD and Mantoux test, which were all negative. Culture of the throat swab showed a good growth of *Staphylococcus aureus* sensitive to the tetracycline that was being given. Gastric washings, examined for acid-fast bacilli, were negative, and so was the sweat test.

During the first week there was clinical improvement and this was confirmed radiologically, where the consolidation had largely cleared, leaving some paracardiac opacification on the right side. Weight gain during the ensuing month was slow, and from time to time it was necessary to administer adrenaline intradermally, for severe bronchospasm, audible for quite a distance from the side of the cot.

With the slow clinical improvement, the right hilar area and paracardiac region remained opacified on serial X-rays of the chest. At this stage it was decided to change the antibiotic in an effort to improve the condition. On 2 November 1965 the radiologist reported the right hilar density to be better defined, and to have a lobulated appearance, in keeping with a gland which was compressing the right main bronchus at its origin.

On 20 November 1965, after being picked up for feeding by a nurse, his head became acutely flexed forward,

resulting in the child becoming cyanosed and momentarily apnoeic. A day later, this recurred.

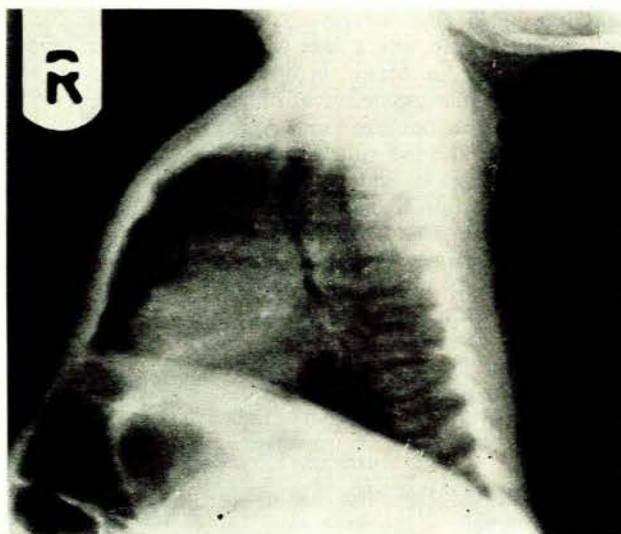


Fig. 1(a)

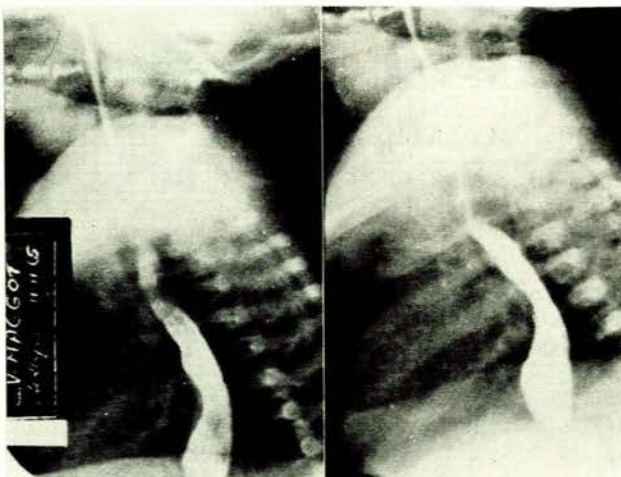


Fig. 1(b)

Fig. 1. Lateral X-ray views of chest (a) showing trachea indented posteriorly and (b) oesophagus outlined with barium similarly indented.

A barium swallow was ordered in an effort to establish the cause of these episodes. It showed, as was suspected, a slight narrowing of the oesophagus, in the region of the aortic knuckle, but there was no impediment to the free passage of the barium. The oesophagus above and below this was normal in calibre and appearance, and there was no displacement.

Bronchoscopy was then performed under general anaesthesia. This showed the lower trachea to be pushed forward and indented by a pulsatile swelling. On oesophago-



scopy, there were suggestions of pulsation, both anteriorly and posteriorly. A diagnosis of a double aortic arch was then made and the child was prepared for thoracotomy.

#### *Course and Management*

At operation on 10 December 1965, a left lateral intercostal thoracotomy was performed. The descending thoracic aorta was found to be on the left with a large right-sided arch crossing the midline behind the oesophagus. The left subclavian artery was identified alongside the oesophagus. There was a long vessel forming an anterior or left arch, about 4 mm. in diameter; the anterior connection with the ascending aorta was buried under the thymus, but the posterior end joined the posterior arch at the level of the left subclavian. This vessel was divided with no change in the pulses and the ends retracted readily. The connective tissue on the left of the trachea and oesophagus was also loosened. The chest was closed in the usual manner with one underwater-seal drain.

Postoperatively the patient made a good recovery. The consolidation in the right upper zone gradually diminished. No further cyanotic spells were noted and the infant gained weight rapidly while in hospital. The drainage tube was removed on the 2nd postoperative day and he was discharged on 28 December 1965.

#### DISCUSSION

A complete vascular ring formed by the aorta and its branches is not a common occurrence. In 1962, it was stated that less than 100 had been reported in the literature.<sup>1</sup> Various combinations are possible; usually there are 2 arches of which the left (or anterior) is the smaller; but a ductus or ligamen'um arteriosum may be the factor completing a ring and thus producing obstruction.

Symptoms usually occur early—within the first few months of life. There is evidence of progressive respiratory obstruction, with inspiratory stridor and costal recession. The neck may be kept extended in an effort to breathe. The obstruction is not relieved by bronchodilatation. Respiratory infections are common and may occur repeatedly. There may be increased respiratory difficulty during feeding, as the oesophagus is enclosed as well as the trachea, and distension of the former adds to the airway obstruction.

The condition should be borne in mind and the diagnosis usually can be made by contrast studies. The simplest is with contrast medium in the oesophagus, when the indentation is readily noted. Bronchoscopy and bronchography may be performed and if readily available, cineangiocardiology should demonstrate the anatomical arrangement. This last is useful if there is any question of associated stenosis of one of the main vessels.

A left thoracotomy gives adequate access to the vast majority of these cases when the constricting ring can be divided without compromising the blood supply to vital organs.

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#### REFERENCE

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