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Case Report

Spontaneous Closure of a Ductus Arteriosus Aneurysm in a Neonate

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ABSTRACT

We report a case of ductus arteriosus aneurysm (DAA) in a neonate with respiratory distress. Echocardiography and cystographic computed tomography were useful for establishing the diagnosis. After 10 days, the DAA had been filled by thrombus, and the commissure with the aorta had disappeared. Clinical findings due to infarction by thrombus or rupture of a localized dilatation were not recognized. The dyspnea gradually resolved without treatment. DAA can cause sudden death by rupture. However, deciding when to resect the DAA is difficult. Decreasing velocity of blood influx and the presence of thrombus in the DAA may indicate imminent occlusion of the DAA. DAA may be more common than now recognized. Echocardiography should be performed to exclude DAA in neonates with stridor. (Jikeikai Med J 2002 ; 49 : 55-8)

Key words : ductus arteriosus aneurysm, neonate, respiratory distress

Because the ductus arteriosus is a vessel necessary only for fetal circulation, it normally closes spontaneously after birth. Patent ductus arteriosus is a congenital anomaly in which the ductus arteriosus does not close. Ductus arteriosus aneurysm (DAA) is visible as a localized dilatation on thoracic X-ray films when the ductus arteriosus expands. Although DAA is rare[1], follow-up by cardiologists is necessary because death can occur should the aneurysm rupture[2]. However, the natural course of DAA is poorly understood[3,4]. We report a case of spontaneous closure and thrombosis of a DAA.

CASE REPORT

A female neonate was admitted soon after birth because of severe inspiratory stridor. The child had been born at 41 weeks' gestation after a normal pregnancy ; birth weight was 2,970 g. Inspiratory stridor was present even during sleep, and generalized cyanosis developed when she cried. No external congenital anomalies were recognized on initial physical examination. We initially suspected respiratory tract stenosis due to a vascular ring. However, bronchial transmission appeared normal and no localized dilatation was seen on thoracic X-rays films. No abnormal densities were recognized in the pulmonary area.

On cystographic computed tomography the aortic arch was to the left of the trachea, and a double aortic arch was not recognized. The brachiocephalic trunk, the left subclavian artery, and the left common carotid artery arose from the aorta in their normal positions. However, a vessel with a localized dilatation was present leftward of the aortic arch. The localized dilatation was larger than the lumen of the descending aorta with a maximal diameter of approximately 10 mm. No commissure was seen between

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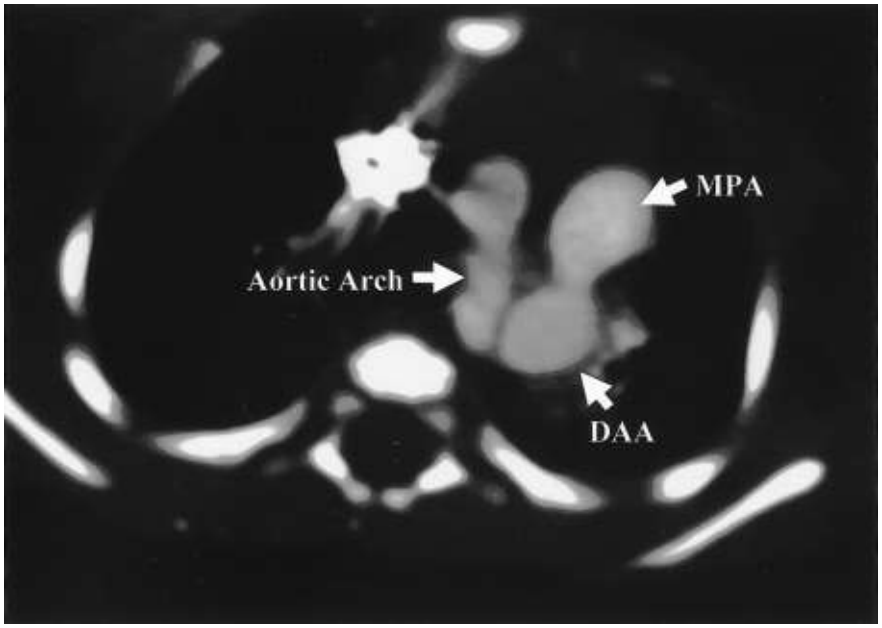

Fig. 1. On cystographic computed tomography the aortic arch was to the left of the trachea, and a vessel with a localized dilatation (DAA) was existed in leftward of the aortic arch. MPA, main pulmonary artery : DAA, ductus arteriosus aneurysm.

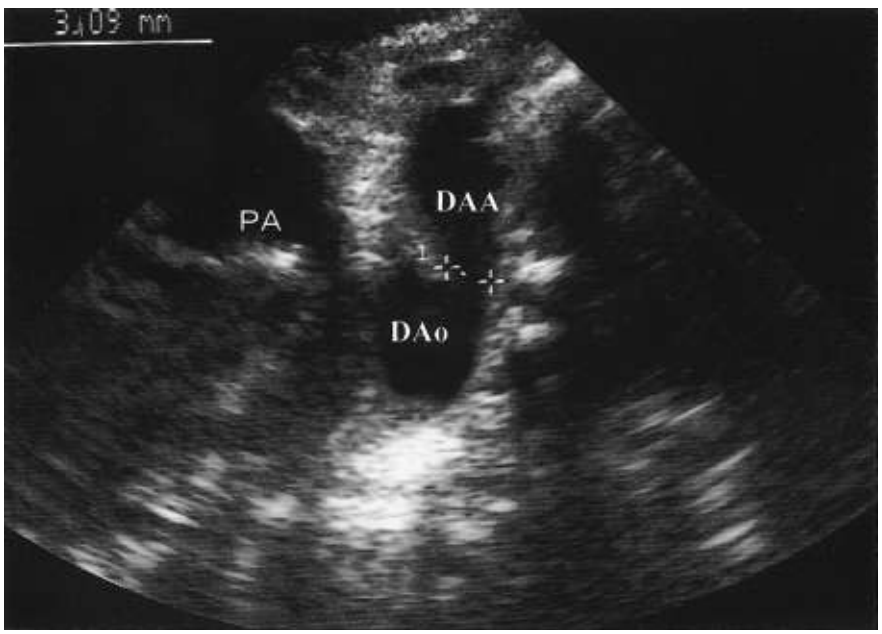

Fig. 2. Cardiac echography on admission. A localized dilatation (DAA) was existed on superior sternal approach. PA, pulmonary artery : DAo, descending aorta.

the pulmonary artery and the localized dilatation (Fig. 1).

The localized dilatation was also recognized in the same location on echocardiography (Fig. 2). In addition, a commissure separated the localized dilatation from the descending aorta but from the pulmonary artery. The velocity of influx flow to the localized dilatation was 1 m per second. No thrombus

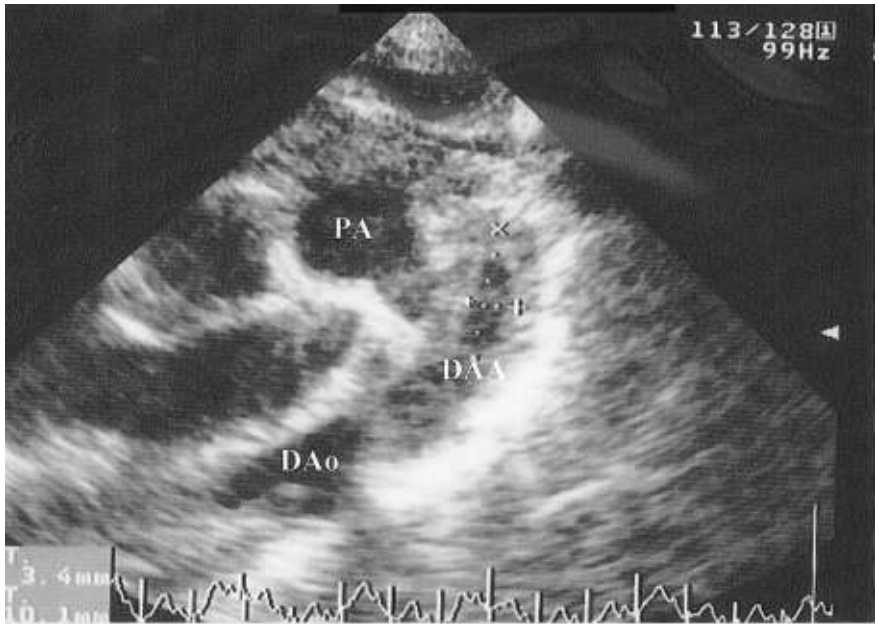

Fig. 3. Cardiac echography on the fourth day of admission. The presence of thrombus within the aneurysm was confirmed.

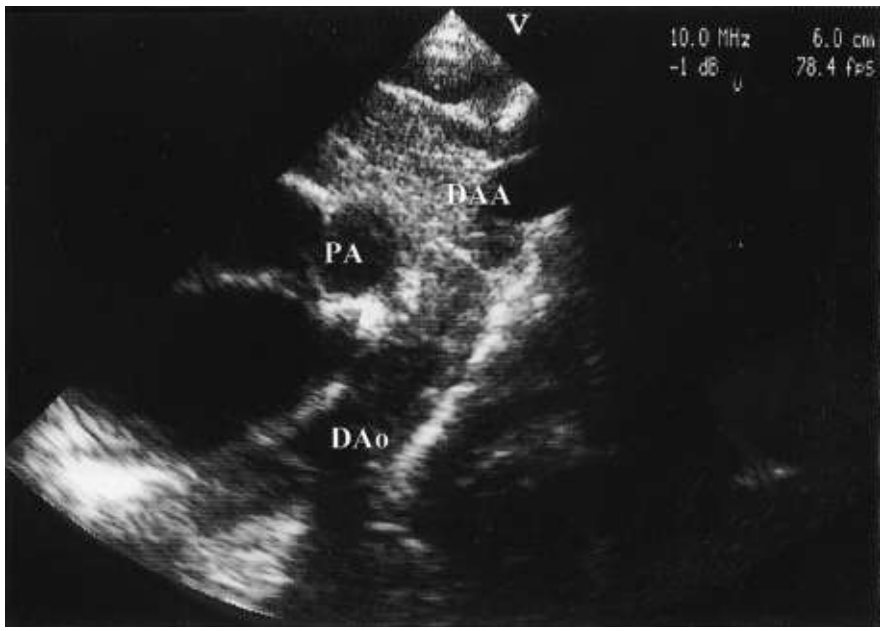

Fig. 4. Cardiac echography on the tenth day of admission. The DAA had become completely filled with thrombus.

was present in the localized dilatation.

DAA was diagnosed on the basis of imaging studies. The DAA was believed to have caused inspiratory stridor by pressing upon the trachea. Because dilation was not seen to increase on follow-up echocardiography, emergency resection of the DAA was not considered necessary. The presence of thrombus within the aneurysm was confirmed on the fourth day of admission, when the inflow velocity to the aneurysm had decreased (Fig. 3). The DAA had become completely filled with thrombus by the 10th day of admission, when the velocity of blood inflow

became zero (Fig. 4). The inspiratory stridor also resolved at this stage. The DAA would assume regression like normal ductus arteriosus. Neither Respiratory distress nor thrombus was present on examination 6 months later.

DISCUSSION

DAA is rarely reported in Japan. In most reports from outside Japan, DAA is discovered incidentally on fetal echocardiography or during evaluation of neonatal dyspnea. To our knowledge, morbidity due to DAA has not been reported. Some infants with DAA have associated syndromes, such as Marfan, Smith-Lemli-Opitz, and 21 trisomy and 13 trisomy[5,6]. However, our patient had no congenital anomalies except DAA. Pathologic findings of surgically resected DAA include a weak vascular wall because of reduced intimal cushion formation and abnormal deposition of elastin[2]. This weakening of the vessel wall progresses to a localized dilatation when the pulmonary artery side of the ductus arteriosus becomes occluded.

Indications for DAA resection have not been established, but patients have died after rupture of DAA[1,7]. Therefore, the DAA should immediately be resected when the dilatation is seen to progress on echocardiography[2]. When thrombus reaches the descending aorta or the pulmonary artery, surgery is also indicated[8]. However, because a DAA may rupture when still small, deciding when to proceed with resection is difficult. On the other hand, as thrombus formation gradually increases within a DAA, blood-inflow velocity and blood volume, which place stress on the vessel wall, decrease and may reduce the risk of DAA rupture. In fact, we are aware of no patients in whom a DAA with extensive thrombus ruptured.

Inspiratory stridor in this patient was likely due to compression of the trachea. Although respiratory distress can remain after surgery for vascular rings[9,10], respiratory distress in DAA is transient. Perhaps the DAA presses upon the trachea in the fetus, but does not disturb growth of the trachea because localized dilatations appear in the antenatal period after the tracheal cartilage has formed. In one study, DAA was recognized with ultrasound at 30 weeks' gestation in 3 (1.5%) of 200 fetuses examined but produced no symptoms after birth[2]. In fact, DAA may be more common than now recognized. Echocardiography should be performed to exclude DAA in neonates with stridor.

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