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CASE REPORTS

Retroaortic Innominate Vein With Coarctation of the Aorta: Surgical Repair and Embryology Review

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A newborn girl with a retroaortic innominate vein, coarctation of the aorta, ventricular septal defect, and subaortic stenosis underwent a complete repair at 8 days of age. The ascending aorta was transected and the innominate vein was brought in front of it. We review the anatomy and embryology of this rare entity and describe the surgical technique to avoid recoarctation and innomante vein compression.

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A left retroaortic brachiocephalic (innominate) vein is a rare condition. It has an incidence of 0.2% to 1% among congenital cardiac anomalies [1–3]. Although is was first described by Kerschner [4] more than 100 years ago, the clinical experience with this anomaly is limited with less then 100 cases described worldwide [1–5]. It is frequently associated with right aortic arch or high left aortic arch [1, 6]. We believe that this association is not accidental. Herein we review the embryology to support our theory and provide a basis for surgical correction.

A newborn girl weighing 3.12 kg was noted to have a murmur at birth with weak femoral pulses. Her arm-to-leg blood pressure gradient was 20 mm Hg. Echocardiogram demonstrated an unrestrictive ventricular septal defect (VSD) with posterior malalignment of the outlet septum, subaortic stenosis, restrictive patent foramen ovale (PFO), almost closed ductus, bicuspid aortic valve and good biventricular function. Aortic annulus was 0.5 cm, pulmonary annulus 1.1 cm, tricuspid annulus 1.2 cm, ascending aorta proximal to left carotid artery (LCA) was 0.45 cm. There was a juxtaductal coarctation at the level of the left subclavian artery (LSA). There was 1-cm and 0.31-cm in diameter narrow segment of the arch between the LCA and LSA The patient was started on prostaglandin (PGE1) at 0.05 $\mu g \cdot kg^{-1} \cdot min^{-1}$. She remained stable and underwent elective surgery on March 20, 2002 at the age of 8 days.

The surgery was performed through the midline sternotomy. There was an incidental finding of the retroaortic innominate vein (Fig 1A). The patient was place on cardiopulmonary bypass (CPB) in standard fashion using ascending aortic and bi-caval cannulation. Patient was cooled to 25°C. Descending aorta and arch vessels were mobilized. After cardioplegic arrest, the PDA was ligated and circulatory arrest was established for 25 minutes. During the period of circulatory arrest, ascending aorta was transected and the innominate vein was brought in front of the ascending aorta. The hypoplastic aorta was ligated just beyond the LSA and ductal tissue was resected. The continuity of the ascending aorta was reestablished by a "semilunar" anastomosis and the descending aorta was anastomosed into the ascending aorta end-to-side (Fig 1B). CPB was resumed. A subaortic myectomy was performed and the ventricular septal defect was closed with a Dacron Sauvage patch. PFO was closed with direct suture. Total aortic cross-clamp time was 93 minutes. Total CPB time was 79 minutes. Upon weaning off pump transesophageal echocardiogram (TEE) was performed and demonstrated unobstructed flow in the ascending aorta and through the end-to-side aortic anastomosis. There was flow acceleration in the subaortic area with a mean gradient of 8 mm Hg. Patient developed complete heart block and required sequential pacing for 5 days but regained normal sinus rhythm on postoperative day 5. She was discharged home 10 days after surgery.

Comment

In our patient, there was a significant distance between the origin of the left common carotid and the left subclavian artery, and a high aortic arch. These findings are typical of "pseudocoarctation" of the aorta, which distinguishes itself from a true coarctation by a high aortic arch and the absence of clinical signs or hemodynamic abnormalities, with the trans-stenotic pressure gradient remaining below 25 mm Hg [7, 8]. Embryologically, this anomaly results from a lack of shortening of the left dorsal and ventral aortic roots of the left fourth aortic arch [8]. At the eighth week both anterior cardinal veins are interconnected by the superior and inferior capillary plexuses (Fig 2A). Normally, the aortic arch shortens during the embryological development and occupies the space of the inferior transverse capillary plexus, thus causing its regression, while the rest of venous blood shunts into the superior transverse capillary plexus. This facilitates normal development of the innominate vein

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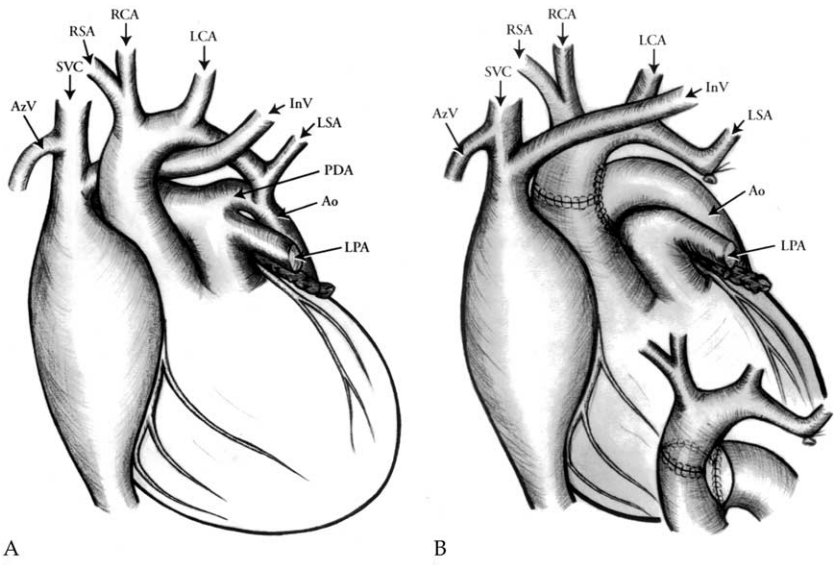

*Fig 1. Intraoperative view of the anomaly. (A) Anatomy. Retroaortic innominate vein enters the superior vena cava (*SVC*) below the azygos vein (*AzV*). Increased distance between the left carotid artery (*LCA*) and left subclavian artery (*LSA*). (B) Surgical repair. Ascending aorta transected, the innominate vein brought in front of the aorta. Descending aorta anastomosed into the ascending aorta end-to-side. (*Ao *= aorta;* InV *= innominate [left brachiocephalic] vein;* LPA *= left pulmonary artery;* PDA *= patent ductus arteriosus;* RCA *= right carotid artery;* RSA *= right subclavian artery.)*

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(Fig 2B). In case of the right aortic arch or high aortic arch, the inferior capillary plexus may persist and form a retroaortic innominate vein (Fig 2C). It is likely that in most cases, the ascending aorta still compresses the orifice of the inferior transverse capillary plexus causing the venous blood to shunt through the left anterior cardinal vein. The latter results in persistence of the left superior vena cava. In fact, there was a distinct narrowing of the retroaortic innominate vein at the site of its entry to the SVC due to ascending aortic compression. The above theory would explain frequent association of the retroaortic innominate vein with tetralogy of Fallot and right aortic arch [1].

As expected, there was a long narrow area between left carotid and left subclavian artery in our patient. An extended end-to-end repair of this type of coarctation associated with high aortic arch would invariably result in shortening of the aortic arch and compression of the retroaortic innominate vein. Thus, it seemed mandatory to bring the innominate vein in front of the ascending aorta to prevent such compression. The concern was, however, that transection and anastomosing of the ascending aorta might result in narrowing at the site of the anastomosis later in life. To avoid this complication, we performed a "semilunar" anastomosis in the shape of an

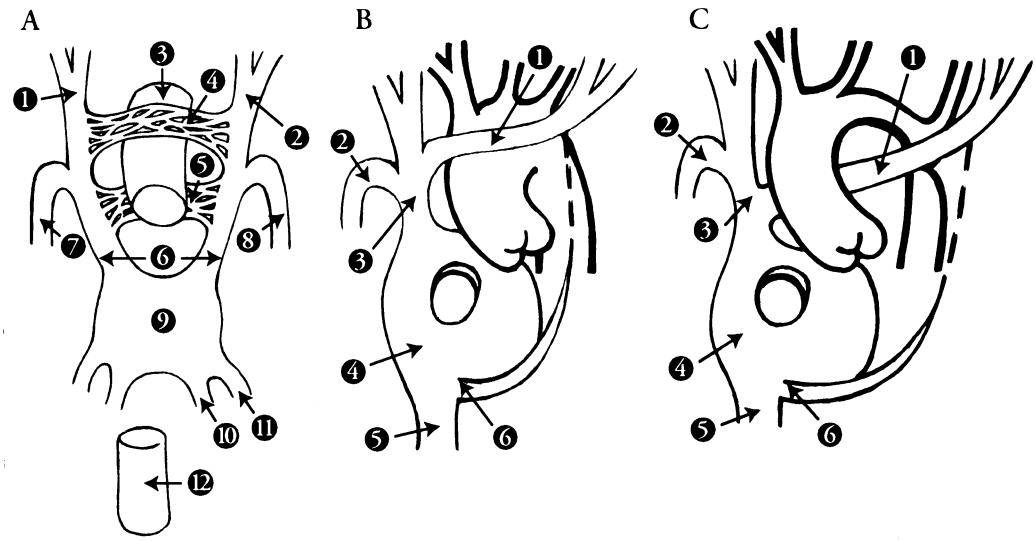

Fig 2. Diagrams illustrating the development of the innominate vein. (A) In the sixth week of gestation: 1 *= right anterior cardinal vein;* 2 *= left anterior cardinal vein;* 3 *= primitive aorta;* 4 *= superior transverse capillary plexus;* 5 *= inferior transverse capillary plexus;* 6 *= common cardinal veins;* 7 *= right posterior cardinal vein;* 8 *= left posterior cardinal vein;* 9 *= sinus venosus;* 10 *= vitelline vein;* 11 *= umbilical vein;* 12 *= developing inferior vena cava. (B) Normal anatomy:* 1 *= innominate vein;* 2 *= azygos vein;* 3 *= superior vena cava;* 4 *= right atrium;* 5 *= inferior vena cava;* 6 *= coronary sinus. (C) Retroaortic innominate vein:* 1 *= innominate vein;* 2 *= azygos vein;* 3 *= superior vena cava;* 4 *= right atrium;* 5 *= inferior vena cava;* 6 *= coronary sinus.*

incomplete ring and restored the continuity of the aorta with the end-to-side anastomosis.

In summary, the described technique allowed us to avoid compression of the retroaortic innominate vein and create a widely open aortic anastomosis. Embryology gives a useful clue to understanding of the surgical anatomy of this rare malformation.

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CASE REPORTS

Development of Brain Metastasis 5 Years Before the Appearance of the Primary Lung Cancer: "Messenger Metachronous Metastasis"

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We report a patient with a brain metastasis that presented 5 years before the primary adenocarcinoma of the lung from which it originated. The metastasis and the primary tumor were removed. To confirm their common origin, we used comparative genomic hybridization. We have named this type of metastasis "messenger metachronous metastasis." The patient remains well 79 months after the brain metastasectomy and 18 months after the lung surgery.

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Bronchogenic carcinoma is the most common malignant tumor diagnosed in the United States, and the development of a brain metastasis during the natural history of non–small cell lung cancer is a common and devastating event. The median survival from the diagnosis of brain metastasis to death is approximately 1 month without treatment and 14 to 24 months in combined brain–lung resection cases [1,2]. The prognosis of a metachronous metastasis is better according to some investigators [3], but others have found no significant difference in survival between patients with synchronous and metachronous lesions [1]. We present an unusual case of brain metastasis from non–small cell lung cancer, which we term "messenger metachronous metastasis," and which appeared 5 years before the discovery of the primary lesion. We treated both lesions aggressively, and the patient remains well.

In 1995, a 51-year-old woman was evaluated for a 1-month history of decreased motor strength in the left lower extremity and for personality changes. Neurologic examination revealed left hemiparesis, and computed tomography (CT) demonstrated a 2-cm tumor in the right parietal area. After surgical removal of the tumor, pathologic evaluation suggested an adenocarcinoma metastasis from the lung; however, meticulous examination by chest CT and bronchoscopy did not disclose a primary lesion. The patient did not receive postoperative whole-brain irradiation. Over the next 4 years, the patient underwent chest CT every 6 months, which revealed no abnormality. At 5 years, after a delay in scanning because of patient neglect, a mass appeared in the hilum of the left upper lobe (cT3 to T4N0). The 8-cm central lung tumor was treated with 25×2 Gy neoadjuvant irradiation. The preoperative staging revealed a 5-mm metastasis in the right cerebellum without any symptoms, but this area was not accessible to surgical removal; accordingly, the patient received 10×3 Gy whole-brain and 5×2 Gy tumor bed irradiation. Because of the good physical status of the patient, we opted to perform left upper lobectomy. The histology demonstrated a T2N0 adenocarcinoma of the lung.

To confirm the common origin of the brain tumor and the lung adenocarcinoma, we used comparative genomic hybridization. After genomic DNA isolation from the paraffin-embedded tissues, we performed amplification and labeling steps as described previously [5]. For comparative genomic hybridization, we used human cDNA microarrays containing 800 random cDNA fragments (amplified and spotted in house) in duplicate. Relative DNA losses and gains were determined by normalizing intensity values to intensities after hybridization with labeled probes obtained from normal lymphoid tissue. We found and located several sequence gains and losses, which were common in both malignant tumors (Table 1). In three cases, brain-specific losses were also demonstrated (Table 1). From these results, we conclude that the two tumors are derived from the same clone. Eighteen months after the lung surgery, the patient was doing well. She had had no neurologic or chest symptoms, and chest radiography did not show any recurrence. Brain magnetic resonance revealed that the brain metastasis had completely disappeared.

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