Pediatr Cardiol 10:81–89, 1989

Pediatric Cardiology

Pulmonary Vascular Sling: Report of Seven Cases and Review of the Literature

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SUMMARY. **A total of 130 cases of pulmonary vascular sling, including seven new cases in our collection, were studied. The sex distribution was 60% male and 40% female. The age at presentation was in the first year of life in ~90% of cases. Barium-esophagraphy showing anterior indentation was diagnostic in most of the cases. Bronchoscopy and tracheobronchography were useful in detecting associated tracheobronchial anomalies preoperatively. The analysis of 68 autopsied cases revealed associated tracheobronchial anomalies in 40% of the cases. Anomalies of the tracheobronchial tree took three major forms: abnormal distribution of cartilage in the walls of the trachea and major bronchi, intrinsic stenosis, and abnormal branching, the latter being that of *bronchus suis*. Acquired changes secondary in the sling resulted in compression of the major respiratory pathway by the anomalous left pulmonary artery. Major associated cardiovascular anomalies were present in 30% of the cases. These were represented by ventricular septal defect, atrial septal defect, patent ductus arteriosus, tetralogy of Fallot, common ventricle, and coarctation of the aorta.**

KEY WORDS: **Pulmonary vascular sling — Tracheobronchial obstruction — Cardiovascular anomalies**

The term, vascular sling, coined in 1958 by Contro and coworkers [11], describes a congenital anomaly in which the left pulmonary artery originates anomalously from the right pulmonary artery. In the typical vascular sling, the anomalous left pulmonary artery arises from the posterior aspect of the right pulmonary artery, passes over the right main stem bronchus near its origin and turns to the left, coursing between the trachea and esophagus to reach the left pulmonary hilus (Fig. 1). In a rare case, as in our case 6 and that reported by Bamman and associates [3], the pulmonary trunk gave rise to an accessory artery to the upper lobe (Fig. 2).

The first case of pulmonary vascular sling was reported in 1897 by Glaevecke and Doehle [18]. To date, to the best of our knowledge, 123 cases have been reported in the literature [1–4, 8–72]. The anomaly characteristically presents with respiratory obstruction in neonates and young infants, with a mortality rate of 90% in the medically managed cases. Potts and coworkers [51] devised an operative procedure for correction of the anomaly, but, in spite of successful surgical correction, mortality and morbidity are high postoperatively and appear to be related to underlying tracheobronchial abnormalities [57]. The arterial anomaly, nevertheless, in some cases is compatible with asymptomatic long-term survival as reported in six adult cases [13, 22, 30, 37, 64].

This report analyzes the literature of the 123 reported cases of pulmonary vascular sling, in addition to the seven unreported and autopsied cases observed by us. The latter cases are presented in summary form in the Appendix to this report. These cases will be added to the 123 reported cases, to yield 130 cases in our review of the literature (62 cases observed clinically only and 68 with autopsy). The two cases in our collection that were reported by Jue and associates [29] are included in our re-

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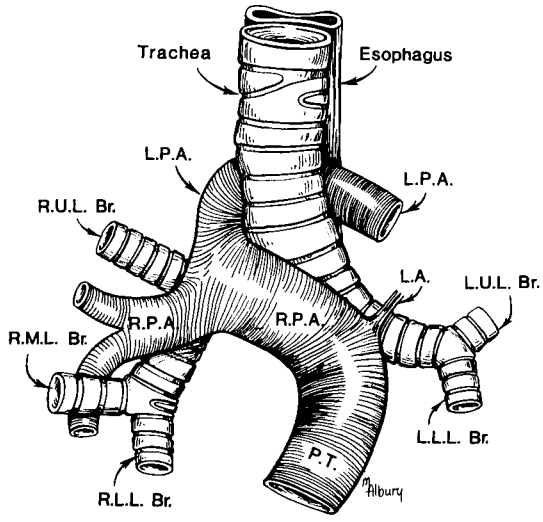

Fig. 1. Classical example of pulmonary arterial sling. The anomalous left pulmonary artery arises from the posterior of the right pulmonary artery, passes over the right main stem bronchus near its origin, turns to the left and courses between the trachea and esophagus to reach the left hilus (courtesy of Dr. Russell V. Lucas, Jr.). *E*, esophagus; *LA*, ligamentum arteriosum; *LPA* and *RPA*, left and right pulmonary arteries, respectively; *LLL Br* and *LUL Br*, left lower and left upper lobe bronchi, respectively; *PT*, pulmonary trunk; and *RLL Br*, *RML Br*, and *RUL Br*, right lower, right middle, and right upper lobe bronchi, respectively.

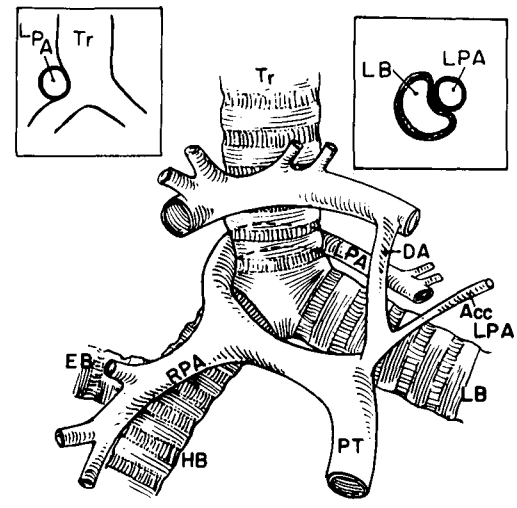

Fig. 2. Atypical pulmonary vascular sling in case 6. An accessory left pulmonary artery passes to the upper lobe of the left lung, while the remaining pulmonary arterial pattern is that of classical sling. Ductus leads to aorta. Features of tracheal and bronchial compression not appreciated in the main diagram are illustrated by the **inserts.** The **top left insert** shows compression and indentation of the right side of the terminal trachea and the origin of the right main stem bronchus by the anomalous left pulmonary artery. The **top right insert** shows a cross section of the left main bronchus and the compression on its posterior aspect caused by the left pulmonary artery. *Acc LPA*, accessory left pulmonary artery; *DA*, ductus arteriosus; *EB*, and *HG*, eparterial and intermediate bronchi, respectively, of right lung; *LB*, left main stem bronchus; *LPA*, left pulmonary artery; *PT*, pulmonary trunk; and *RPA*, right pulmonary artery; (**top left insert**) tracheal bifurcation viewed from in front with the indentation of the angle between the trachea and right bronchi caused by the anomalous left pulmonary artery, and (**top right insert**) a cross section of the left main bronchus with indentation of its posterior wall by the anomalous left pulmonary artery as seen in cross section.

view of the literature, but are not among the seven cases summarized in the Appendix. Our analysis includes (a) clinical features, (b) tracheobronchial abnormalities, (c) radiologic features, and (d) congenital cardiac anomalies associated with pulmonary vascular sling.

In analyzing the 130 cases, the details of the clinical features were extracted from case reports of both survivors and nonsurvivors. Identification of the cardiovascular anomalies resulted from review of cardiac catheterization data and autopsy data. The tracheobronchial abnormalities were identified from reports of autopsies.

Review of Literature

Clinical Features

The ratio of male to female patients was 3 : 2. Of the 130 patients surveyed, 90% were symptomatic; 90% of these were symptomatic in the first year of life, and nearly half of these were symptomatic in the newborn period. When present, symptoms were usually those of respiratory obstruction characterized by stridor and wheezing. The obstruction was maximal during expiration, and there was no associated dysphagia. This is in contrast to the respiratory symptoms caused by vascular rings in which inspiratory obstruction predominates and may be associated with dysphagia. Only 14 (12%) of the reported cases were asymptomatic, comprising six adults, five of whom presented with a paratracheal mass on thoracic roentgenography (in case 6, the condition was first identified at autopsy), and eight children with pulmonary vascular sling identified during catheterization for other forms of congenital cardiac disease or at workup for other conditions.

Tracheobronchial Anomalies

Particular attention was directed toward both congenital anomalies and acquired abnormalities of the tracheobronchial system.

Table 1. Tracheobronchial anomalies in 68 autopsied cases of pulmonary vascular sling

Anomaly	No. of cases[a]	%	References[b]
Hypoplastic distal trachea	35	51	3 (5 cases), 4 (3 cases), 8, 10 (3 cases), 11, 16, 17, 18, 21, 27 (2 cases), 29, 33–35, 47, 54, 58, 66, 67, G (3 cases)
With complete cartilagenous rings, i.e., absent pars membranacea	26	38	3 (4 cases), 4 (1 case), 10 (3 cases), 16, 17, 18, 21, 27 (2 cases), 29, 33–35, 47, 54, 58, 66, 67, G (3 cases)
With normal cartilagenous rings, i.e., pars membranacea present	9	13	3, 4 (2 cases), 8, 11, 46, 57, 65, 71
Stenosis of main stem bronchus	5	5	10 (2 cases), 18, 36, G (1 case)
Right epiarterial bronchus	8	12	3, 16, 27, 29, 36, 65, 67, G (1 case)
Left upper bronchus	1	1	27
Tracheoesophageal fistula	1	1	67

[a] Some cases showed more than one anomaly; the numbers refer to the number of times that each anomaly was observed, not to the number of patients with associated cardiac anomalies.
[b] G refers to case or cases in our series (Appendix) exhibiting particular feature.

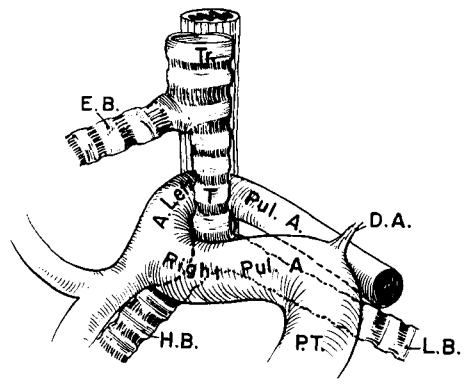

Fig. 3. Pulmonary vascular sling associated with *bronchus suis*. The right upper lobe bronchus arises independently from the branching of the trachea, *Tr*. The branching of the trachea involves the origin of the left main stem bronchus (*LB*) and the intermediate or hyparterial bronchus (HB). In pulmonary vascular sling, the left pulmonary artery passes over the intermediate bronchus inferior to the independently arising right upper lobe bronchus. *DA*, ligamentum arteriosum; *EB*, eparterial bronchus; and *PT*, pulmonary trunk. Illustration modified from Jue and associates [29].

Analysis of 68 autopsied cases forms the basis for the recognition of associated anomalies of the tracheobronchial tree, as well as of secondary acquired complications of the sling (Table 1).

Anomalies of the tracheobronchial tree took three major forms: (a) abnormal branching, (b) abnormal distribution of cartilage in the walls of the trachea and major bronchi, and (c) intrinsic stenosis.

Abnormal branching seen in four of the autopsied cases was that called *bronchus suis*. (Three additional cases were observed in the living series.) In this condition, the right upper bronchus arises independently from the trachea, leaving the intermediate bronchus of the right lung and the left main bronchus as the terminal branches of the trachea. In each of the cases with *bronchus suis*, that portion of the trachea distal to the origin of the right upper bronchus was hypoplastic. In two cases, the anomalous branching complicated interpretation of the vascular anomaly during thoracotomy [29]. Characteristically, the vascular sling passes over the intermediate bronchus inferior to the independently arising right upper bronchus (Fig. 3). Six cases of abnormal lobulation of the lungs were reported, consisting of either unilobulation or incomplete lobulation. One case of hypoplastic right lung was seen.

In instances of abnormal distribution of cartilage, there was either (a) encirclement of the trachea by cartilage instead of the normal presence of the membranous segment or (b) absence of cartilage in the terminal part of the trachea.

Among the 68 autopsied cases analyzed, encirclement of the trachea by cartilage was seen in 26 (38%) of cases. In three of these cases, the abnormality extended into the major bronchi. Cartilaginous encirclement of the trachea usually, but not always, is a cause of intrinsic tracheal stenosis [4, 10].

Absence of cartilage in the distal segment of the trachea and similar involvement of the origin of both main stem bronchi was reported in one case by Park and coworkers [47]. This condition predisposes the patient to expiratory collapse of the airway resulting in air trapping and further compromise of the respiratory system. Intrinsic tracheal stenosis was observed in nine cases, none showing complete cartilaginous encirclement.

Acquired Abnormalities of Airway

Acquired changes secondary to the sling took the form of compression of the major respiratory path-

Table 2. Radiologic features in 89 reported cases of pulmonary vascular sling

Features	No. of cases	%	References[a]
Unilateral obstructive emphysema involving right lung	25	28	19, 20 (2 cases), 29, 31, 36, 41, 42, 43, 51, 52, 55, 57, 60, 62, 67 (3 cases), 70, G (3 cases)
Unilateral obstructive emphysema involving left lung	9	11	2, 10 (2 cases), 36, 42, 57, 61, 68, 70
Bilateral emphysema	7	8	4, 9, 42, 47, 61, 67, 71
Mediastinal mass	12	12	3, 5, 10, 22, 28, 32–34, 36, 50, 57, 61
Opaque right lung	2	2	70
Decreased perfusion left lung	1	1	40
Barium swallow normal, i.e., no indentation on esophagram	14	16	10, 12, 27, 31 (2 cases), 36, 40, 51, 61, 62, 67, 69, 70, 71, G (1 case)

[a] G refers to case or cases in our series (Appendix) exhibiting particular feature.

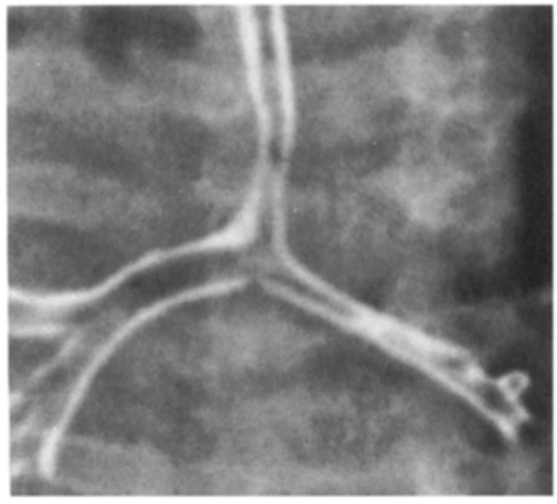

Fig. 4. Bronchogram obtained from case 7 demonstrating long-segment tracheal stenosis with marked hypoplasia of the lower trachea with involvement of the left main bronchus caused by complete tracheal rings.

way by the anomalous left pulmonary artery. When the bronchial branching was normal, compression by this vessel resulted in deformity at the origin of the right main stem bronchus and of the right side of the terminal segment of the trachea; however, compression of the trachea just above the carina or of the left main stem bronchus at its origin was seen, although less frequently. The deformities arising from the direct pressure of the aberrant left pulmonary artery may be of minor significance as in our cases 2 and 4.

Radiographic Features

A summary of the reported radiologic findings is given in Table 2. The percentages given concern those cases in which appropriate studies had been done. In many of the reported cases, findings on thoracic roentgenography done primarily for evaluation of respiratory obstruction yielded the first suggestion of vascular sling. The most common radiologic feature was obstructive emphysema involving the right lung (28%). Mediastinal shift to the left was also found in 60% of these cases. Left-sided emphysema was seen in 10% of cases and bilateral emphysema was identified in 6%. In some cases, the right upper lobe bronchus arose independently from the trachea (*bronchus suis*). In this state, the aberrant left pulmonary artery courses inferior to the right upper bronchus. In five such cases, roentgenography had been done. In these, the upper lobe of the right lung showed hyperexpansion, while there was a tendency for atelectasis of the middle and lower lobes.

The aberrant left pulmonary artery causes a characteristic anterior indentation of the barium-filled esophagus at the level of the tracheal bifurcation, a phenomenon first described by Wittenborg and coworkers [71]. In 66 cases, a barium esophagram gave such results. However, a barium study was interpreted as normal in 20 (23%) of the cases. It has been stressed that only the vascular sling is the vascular anomaly causing anterior indentation of the esophagus [29]. Recently, Castaneda-Zuniga and coworkers [7] reported a case in which a systemic collateral artery caused an anterior indentation in the esophagus at a level higher than that seen with the vascular sling.

Selective pulmonary arteriography has been reported as showing the vascular sling in 56 (93%) of 60 cases. In four cases with complex associated cardiovascular and tracheobronchial anomalies, however, selective pulmonary angiograms could not be interpreted satisfactorily regarding the question of vascular sling.

Bronchoscopy and tracheobronchography were performed in 54 cases to detect additional developmental anomalies in an effort to predict the postoperative respiratory course (Fig. 4). Fifty cases (93%) were reported as showing intrinsic tracheal and/or bronchial narrowing.

Recently, three cases of vascular sling have been demonstrated by computerized tomography with contrast medium [55]. In each of these, the

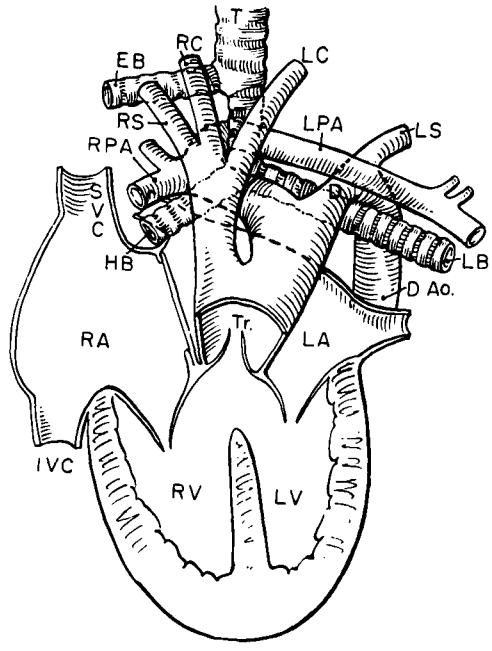

Fig. 5. Case 4. This case had complex cardiovascular anomalies, as well as anomalous branching of the trachea (*bronchus suis*) associated with pulmonary artery sling. The cardiovascular anomalies consisted of persistent truncus arteriosus and interruption of the aortic arch, Type B. *D*, ductus arteriosus; *D Ao*, descending aorta; *EB* and *HB*, eparterial and intermediate bronchi, respectively, of right lung; *IVC*, inferior vena cava; *LA*, left atrium; *LB*, left main stem bronchus; *LC*, left common carotid artery; *LPA*, anomalous left pulmonary artery; *LS*, left subclavian artery; *LV*, left ventricle; *RA*, right atrium; *RC*, right common carotid artery; *RPA*, right pulmonary artery; *RS*, right subclavian artery; *RV*, right ventricle; *SVC*, superior vena cava; *T*, trachea; and *TR*, truncus arteriosus.

condition was confirmed by pulmonary arteriography. The studies reported indicate that simple barium esophagraphy has a high specificity for the diagnosis of vascular sling—this, plus the fact that computerized tomography complemented by cardiac ultrasound [72] seems highly reliable and should be a basis for avoiding pulmonary arteriography in most cases.

Associated Cardiovascular Anomalies

To determine whether associated cardiovascular anomalies were present, use was made of the records of each of the 68 autopsied cases and 25 living patients in which catheterization data were reported. Anomalies classified as major were defined as those that caused abnormal hemodynamics and therefore could exaggerate symptoms other than those of the sling. Minor anomalies were those that did not cause abnormal hemodynamics. In nine cases (10%), minor anomalies were found as isolated associated conditions. In seven cases, minor anomalies were found in cases having major anomalies as well. Among the 93 cases analyzed, major cardiovascular anomalies were present in 30% (Fig. 5 and Table 3).

Table 3. Cardiovascular anomalies associated with 93 cases of pulmonary vascular sling

Cardiac anomalies	No. of times condition observed	References[a]
Major		
Ventricular septal defect	12	8, 17, 24, 31, 32, 34, 36 (2 cases), 57, 65, G (1 case)
Atrial septal defect[b]	9	3, 20, 27 (2 cases), 58, 66, 67, 71, G (1 case)
Patent ductus arteriosus[c] (patients ≥4 months of age)	4	20, 46, 48, 57
Tetralogy of Fallot	3	3 (2 cases), 27
Coarctation of aorta	2	31, 71
Common ventricle	2	11, 31
Pulmonary atresia	1	G (1 case)
Persistent truncus arteriosus with interruption of aortic arch (Type B)	1	G (1 case)
Raghib's syndrome [53]	1	G (1 case)
Total	35	
Minor[d]		
Persistent left superior vena cava, solitary	8	10 (2 cases), 20, 27, 29, 59, 65, 71
Quadricuspid pulmonary valve	1	G (1 case)
Total	9	

[a] G refers to case or cases in our series (Appendix) exhibiting particular feature.
[b] In seven cases, atrial septal defect was associated with other cardiac anomalies.
[c] In six cases, patent ductus arteriosus occurred as an isolated finding, but was considered small, while, in ten cases, it was associated with other cardiac anomalies.
[d] In seven cases, persistent left superior vena cava was found in association with the major anomalies listed. References to these cases are 11, 17, 27, 31, 36, G (1 case).

Comment

Pulmonary vascular sling remains a rare anomaly, with only 123 cases reported in the literature since

the first description in 1897 [18]. Nevertheless, it is an established entity responsible for respiratory obstruction in some neonates and young children. The most common clinical presentation has been that of respiratory obstruction, more prominent in the expiratory phase. Of the autopsied cases, 51% had intrinsic obstructive lesions of the tracheobronchial tree distal and/or proximal to the anatomical site of compression by the sling.

In our initial review of this condition [29], we concluded that death in early infancy would occur unless the airway obstruction caused by the anomalous left pulmonary artery could be corrected surgically. With the marked increase in reported cases in recent years, a better perspective may be gained concerning the variability in the degree of airway obstruction and natural history of this condition.

In their detailed pathological studies of the tracheobronchial tree in this condition, Cohen and Landing [10] emphasized that the airway obstruction could be caused by extrinsic compression secondary to the anomalous left pulmonary artery or by intrinsic obstruction related to complete tracheal rings.

Intrinsic airway obstruction caused by congenital anomalies of the tracheobronchial tree take the form of tracheomalacia, bronchomalacia, and tracheostenosis. Berdon and associates [4] have separated tracheal stenosis in to two types. In the first type, long-segment tracheal stenosis is associated with complete tracheal rings, the "ring-sling complex," which has a very grave prognosis. In the second type, a sufficient amount of the *pars membranacea* is present so as to allow tracheal growth, but obstructive symptoms persist. These authors have also emphasized the importance of carefully performed bronchography and bronchograms to delineate the nature and severity of the obstruction as illustrated in case 7 of our series.

Based upon the presence or absence of major airway obstruction with or without associated congenital cardiac malformations, a comprehensive classification has been proposed [28]:

I. Vascular sling with absence of tracheobronchial tree obstruction
   A. Isolated
   B. Associated with congenital heart disease
II. Vascular sling with associated tracheobronchial tree obstruction
   A. Extrinsic compression
      1. Isolated
      2. Associated with congenital heart disease
   B. Intrinsic obstruction
      1. Isolated
      2. Associated with congenital heart disease
   C. Extrinsic compression and intrinsic obstruction
      1. Isolated
      2. Associated with congenital heart disease

Vascular sling without tracheobronchial tree obstruction would include the adult cases [13, 22, 30, 37, 64] in which the malformation was incidently discovered during evaluation for an abnormal thoracic roentgenogram or by an esophagram performed for a gastrointestinal disorder. Rarely have cases in the pediatric age groups been recognized without respiratory obstructive symptoms. Occasionally, these cases are uncovered during evaluation of a gastrointestinal anomaly [42, 56] or of congenital heart disease [20, 49]. Tracheobronchial tree obstruction caused by extrinsic compression by the vascular sling forms the largest category. The best results of surgical repair of the vascular sling can be expected from this category [4]. In those cases with associated congenital heart disease, however, the success of repair of the vascular sling may be related to the complexity of the associated cardiac malformation. Isolated intrinsic obstruction is the rarest group with or without associated congenital heart disease [10, 46–48]. While the majority of cases in this category do very poorly, a rare case has been described whereby obstructive symptoms due to tracheal stenosis have improved over time [48]. Cases with intrinsic obstruction and external compression of the airway with or without associated congenital cardiac disease have been reported with a high mortality rate treated either surgically or medically [10, 27, 29, 36, 41, 57, 58].

The classification presented may also provide a framework from which to determine the mode of management. Cases with no tracheobronchial tree obstruction should be treated medically as recommended by Phelan and Venables [48] and by Gumbiner and coworkers [20]. Patients with symptomatic airway obstruction should be carefully evaluated by bronchograms and bronchography to determine the cause and severity of the tracheobronchial tree obstruction. Those with isolated extrinsic airway compression should be surgically treated by division and anastomosis of the anomalous left pulmonary artery to a position anterior and to the left of the trachea [4]. The greatest surgical success would be expected in this category. Surgical division of the ligamentum arteriosum has also been recommended, although the exact role of this structure in accentuating the external compression is unclear. Symptomatic relief of the airway obstruction has been reported following division of the ligamentum arteriosum alone [43, 48].

Surgical treatment of patients with obstructive

symptoms secondary to intrinsic airway obstruction will depend upon the feasibility of correcting the intrinsic airway anomaly. Whereas complete tracheal ring is potentially correctable [26], extension of the process of hypoplasia into either main stem bronchus would make airway repair impossible according to current techniques. Cases with intrinsic obstruction of a mild degree should be followed closely since the symptoms may remain minimal in some instances and even regress in others [20, 48]. In those patients with both extrinsic compression and intrinsic obstruction, surgical repair will depend upon the feasibility of correcting the intrinsic airway obstruction.

In those cases with associated congenital cardiac malformations the repair of the vascular sling must be incorporated in the overall plan to correct the specific cardiac malformation and may require staging of the operative procedures with the initial approach directed at the vascular sling, while the cardiac malformation would be corrected at a later stage.

Acknowledgment. This study was supported by the United Hospital Foundation, St. Paul, Minnesota.

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