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The role of posterior aortopexy in the treatment of left mainstem bronchus compression

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Abstract

OBJECTIVES: We reviewed the role of posterior aortopexy for left mainstem bronchus compression in infants and children.

METHODS: Eighteen children with respiratory symptoms were enrolled between 2005 and 2015 for surgical decompression of the left mainstem bronchus. The children were managed from diagnosis to follow-up by a dedicated tracheal team. Primary outcomes were the complete relief of symptoms or improvement with respect to preoperative clinical status.

RESULTS: The median age was 4 years (0.3–15.4) and the median weight was 13.2 kg (3, 1–40). Symptoms or indications for bronchoscopy included difficult weaning from mechanical ventilation (*n* = 3, 17%), difficult weaning from tracheotomy (*n* = 4, 22%), recurrent pneumonia (*n* = 4, 22%), wheezing (*n* = 3, 17%), atelectasis (*n* = 1, 5.5%), bitonal cough (*n* = 1, 5.5%) and stridor (*n* = 2, 11%). Associated malformations were present in 88.7%. The diagnosis was made by bronchoscopy and computed tomography. Indication for surgery was the presence of pulsations and reduction in the diameter of the left mainstem bronchus compression of more than 70%. Surgery was performed by left posterolateral thoracotomy. Aortopexy was done under bronchoscopic control. No early or late deaths were observed, nor were reoperations necessary. Residual malacia was observed in 8 children (44%). Median follow-up was 4.1 years (0.1–7.1). At last follow-up, 17/18 (94.4%) children showed adequate airway patency.

CONCLUSIONS: The intrathoracic location of the left mainstem bronchus predisposes it to compression. Vascular anomalies represent the most frequent causes. Aortopexy has been advocated as a safe and useful method to relieve the compression, and our results confirmed these findings. Management of these patients is challenging and requires a multidisciplinary team.

Keywords: Coronary heart disease • Vascular compression • Pulmonary artery • Bronchial disease • Bronchial endoscopy

INTRODUCTION

Left mainstem bronchus compression (LMBC) is a well-described cause of respiratory distress in infants and children with symptoms ranging from stridor to Apparent Life Threatening Events (ALTE) [1]. Age at onset and severity of the symptoms most often depend on the degree of airway narrowing [2], but concurrent respiratory infections or immunological disorders may sometimes create obstacles to diagnosis [3]. Several anatomical and structural wall abnormalities are described as factors associated with bronchial collapse, although vascular anomalies are considered to be the most frequent [4]. Among them, the 'forceps', in particular, which consists of the left pulmonary artery, anteriorly, and the descending thoracic aorta, posteriorly (LMBC-PAD), has been well described, and its surgical treatment by aortopexy has been defined as an effective therapy against LMBC [5, 6].

In this article, we describe our experience in the surgical treatment of LMBC-PAD by posterior aortopexy.

MATERIALS AND METHODS

From 2005 to 2015, 76 patients with a median age of 6.4 months (range 0.1–182.8 months) and a median weight of 6.9 kg (range 0.3–

40 kg), suspected to have airway compression, underwent a bronchoscopic examination and surgical decompression of the tracheobronchial tree. Among these, 18 patients (males = 7, females = 11) presented with LMBC. Because they required surgical intervention by posterior aortopexy, they became the subjects of our study.

Patients were diagnosed with LMBC under general anaesthesia and without positive end-expiratory pressure. Well-trained operators conducted the examination using a flexible (Karl Storz®, Germany; Ø 2.5/3.7/5.2 mm) or rigid bronchoscope (Karl Storz®, Germany; Ø 3.5/4/4.5/5/5.5 mm). The endoscopic examination

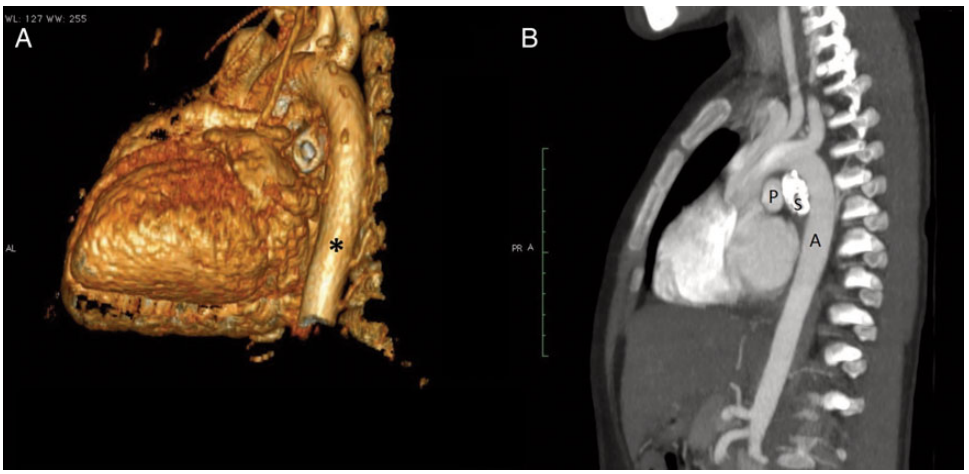

Figure 1: Chest tomograms: (**A**) 3D volume rendering showing the spatial relationship between the thoracic descending aorta (star) and the left mainstem bronchus; (**B**) sagittal plane showing the presence and ovalization of a previously inserted intrabronchial stent (S) compressed between the descending thoracic aorta (A) and the left pulmonary artery (P).

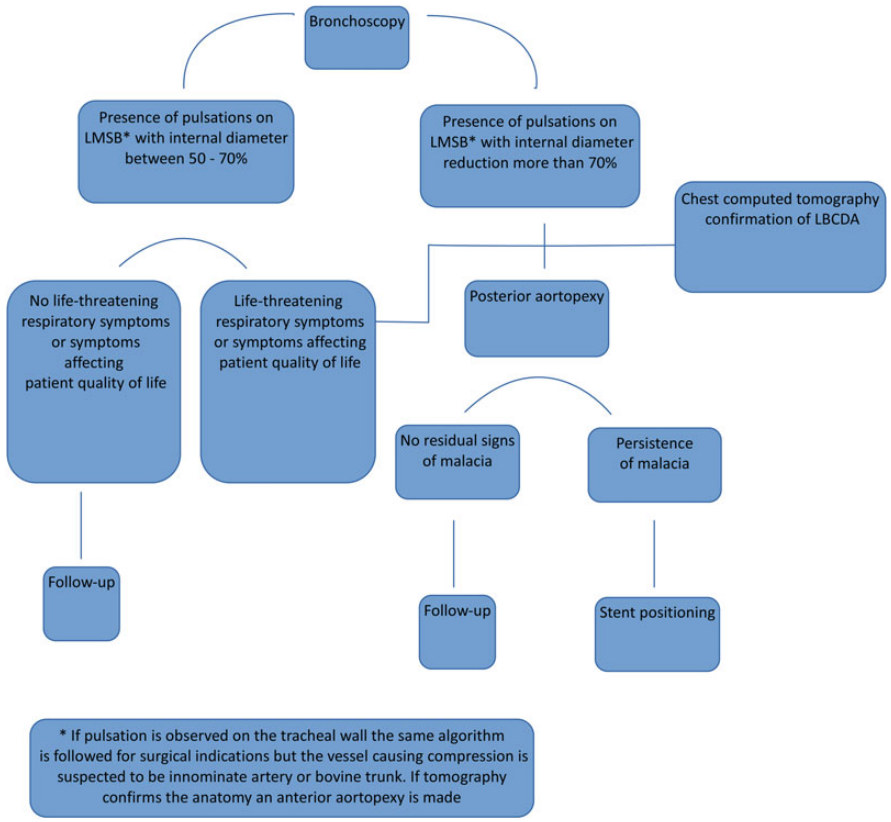

Figure 2: Therapy-guided algorithm.

Table 1: Characteristics of vascular structures

Thoracic structure	Mean	Min–Max
Aortic arch diameter[a] (mm)	9.8	7.2–19.5
Aortic arch position	Left position: 100%	
Descending thoracic aorta diameter[b] (mm)	10.3	5.3–18.5
Descending thoracic aorta position[c]	Midline position: 4/18 (22.2%)	
Left pulmonary artery diameter[b] (mm)	8.6	5.1–12.6

[a]Calculated before the origin of the left carotid artery.
[b]Calculated at the point where it crosses the left mainstem bronchus.
[c]With respect to the vertebral column.

revealed the presence of a pulsatile compression at the level of the left mainstem bronchus, causing a reduction in LMB diameter and indicating the presence of LMBC-PAD.

Evidence of pulsations excluded treatment with an intrabronchial stent as a first resort, the reason being the risk of ischaemia of the bronchial wall due to double pressure on the wall itself, i.e. the pressure of the stent on the one hand and the pressure of the vessels on the other. We came to this conclusion also based on the appearance of ovalization of stents that had been previously implanted (Fig. 1B). The positioning of a stent was thus limited to cases where malacia persisted following the operation (see Results section and Fig. 2 for details), and the stent was placed safely after spacing the vessels appropriately. We used stainless steel metallic stents (Jostent, Abbott®) since their structure has the advantage of allowing air to flow through the wires, which proves to be useful if the stent is placed over a bronchial orifice.

The pulsating site of compression (at the tracheal level or at the left bronchial level) also guided the diagnosis and surgical approach adopted (anterior versus posterior aortopexy, Fig. 2).

A chest computed tomographic (CT) scan was performed in all children (Fig. 1A and B) to confirm the results of the endoscopic examination. The CT scan (Philips Brilliance i-Dose 64 slice) was performed according to age and weight and adjusted following a low-dose protocol (120 or 80 kV; 120 or 80 mA; collimation, 1.5 or 0.75 mm; gantry rotation, 0.5 s).

For diagnostic assessment, the following medical analyses were conducted for each patient: (i) axial contrast, material-enhanced, single-phase low-dose multiple detector CT scans in both the sagittal and coronal planes as algorithms of the soft tissues (two-dimensional multiplanar reformation), (ii) maximum intensity projection, (iii) volume rendering of the thoracic vessels by 3D-reconstructed images and (iv) surface rendering of the thoracic vessels by virtual tracheobronchoscopy. Further anatomical measurements evaluated were the cross-sectional diameter of the left pulmonary artery and descending thoracic aorta, at the exact point where they crossed the LMB, the position of the aortic arch (to the left or to the right) and the position of the descending thoracic aorta compared with that of the vertebral column (refer to Table 1 for details). A preoperative chest radiograph and transthoracic echocardiographic scans completed the diagnostic assessment.

All clinical cases were discussed by a multidisciplinary team that included cardiac surgeons, airway endoscopists, radiologists and paediatric cardiologists. The study was approved by the Ethics Committee of the Gabriele Monasterio Foundation of Massa and of the Meyer Hospital of Florence.

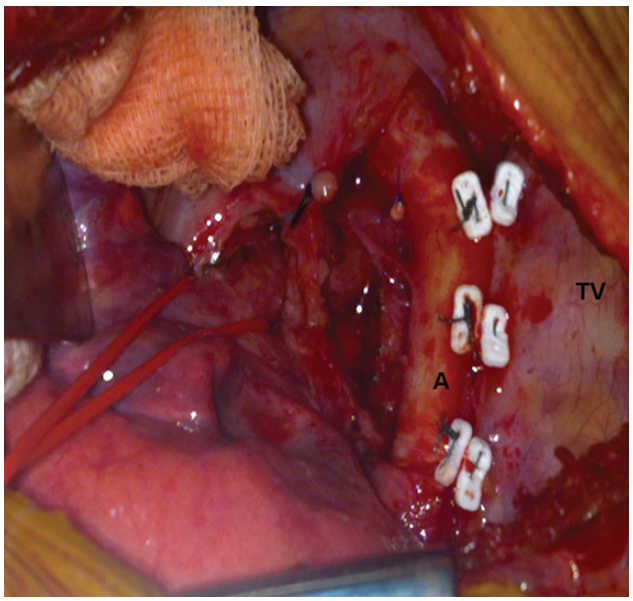

Figure 3: Left thoracotomy, intraoperative view: The image shows the results of the technique [anchorage of the descending thoracic aorta (A) to the thoracic vertebrae (TV)].

Surgical technique

Aortopexy was performed under general anaesthesia and endotracheal intubation. Intraoperative bronchoscopy was performed with a rigid bronchoscope. Left thoracotomy in the fourth intercostal space was made starting 1 cm anteriorly from the mid-axillary line. The latissimus dorsi and the intercostal muscles were divided. The left lung was pulled back, the left superior intercostal vein, if present, was ligated and interrupted, and the mediastinal pleura was opened. The aorta was mobilized from the distal aortic arch to the diaphragm as well as the left pulmonary artery. The ligamentum arteriosum or the ductus arteriosus was divided. At this time, the patient was hyperoxygenated (FiO_2 100% for 30 s) and intraoperative bronchoscopy was performed. The 3/0 Ethibond stitches (Ethicon Endo-Surgery, Inc. Johnson and Johnson, USA) were positioned in the medial layer of the descending thoracic aorta and on the side opposite to the periosteum of the thoracic vertebrae (or their surrounding tissue; Figs 3 and 4).

The stitches were fixed definitively after the pulsations ceased and an appropriate internal diameter of the left main bronchus

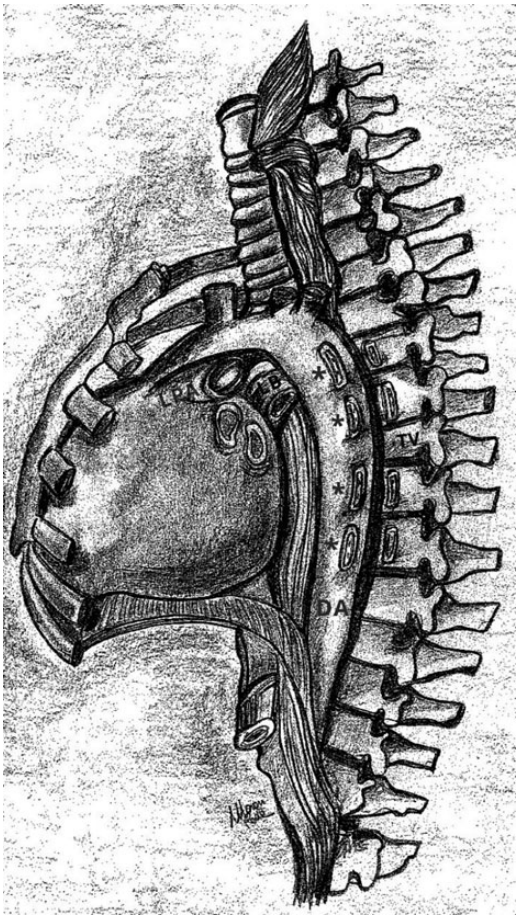

Figure 4: The drawing shows the principle of the surgical technique: The descending thoracic aorta (DA) is pulled back to the thoracic vertebrae (TV) and fixed on them by interrupted stitches (stars). This manoeuvre removes the compression on the left bronchus (LB) and gives more space to the left bronchus between the aorta (DA) and left pulmonary artery (LPA).

was achieved. A drainage tube was left *in situ*, the left lung was re-expanded and the chest was closed. Two children underwent section of an aberrant right subclavian artery and aortopexy at the same time. The vessel was divided only after careful monitoring of the radial pressure, and the vascular stump was closed by a running suture. At the end of the procedure, the bronchoscope was replaced with the endotracheal tube.

RESULTS

The pathological symptoms of LMBC or the indications for bronchoscopic examination of the patients in the study ($n = 18$) ranged from tracheotomy ($n = 4$, 22%) and recurrent pneumonia (22%) to difficult weaning from mechanical ventilation ($n = 3$, 17%) and wheezing (17%). Only 2 patients showed stridor ($n = 2$, 11%), 1 had atelectasis ($n = 1$, 5.5%) and 1 had bitonal cough (5.5%). Sixteen out of 18 children (88.7%) had associated malformations (Table 2). Of these, 5 (22%) had already undergone an anterior aortopexy, 5 (22%) had been exposed to a tracheostomy, and 6 (27.2%) had previously received an intrabronchial stent (Fig. 1B).

Posterior aortopexy was performed on all patients ($n = 18$). At operation, the patients presented a median age of 4 years (range 0.3–15.4 years) and a median weight of 13.2 kg (range 3.1–40 kg). Median ventilation time was 5 days (1–30 days), and median length of hospital stay was 45 days (12–270 days). No early or late deaths were observed and none of the patients required reoperation.

Two children (11%) concurrently underwent sectioning of the aberrant right subclavian artery and one child (5.5%) underwent sectioning of the ligamentum arteriosum. Residual severe malacia following posterior aortopexy was observed in 8 children (44%), 7 of whom required placement of a stent (6 in the left mainstem bronchus and 1 at the level of the distal trachea), and the last one required a tracheostomy. Two children already had a stent prior to

Table 2: Associated malformations and previous operations

Associated malformations	Previous operations
Cardiac	
Ventricular septal defect + hypoplastic aortic arch	Ventricular septal defect closure + aortic arch enlargement
Ventricular septal defect + aortic coarctation	Ventricular septal defect closure + aortic coarctation repair
Right aortic arch + aortic coarctation	Aortic coarctation repair
Type I truncus arteriosus	Truncus arteriosus complete correction
Complete atrioventricular septal defect	Pulmonary artery banding (associate anterior aortopexy)
Complex univentricular heart malformation	Norwood stage I
Complex univentricular heart malformation	Bidirectional cavopulmonary anastomosis
Abdominal	
Oesophageal atresia/tracheo-oesophageal fistula	Oesophageal atresia/tracheo-oesophageal fistula complete correction
Oesophageal atresia/tracheo-oesophageal fistula	Oesophageal atresia/tracheo-oesophageal fistula complete correction
Oesophageal atresia/tracheo-oesophageal fistula	Oesophageal atresia/tracheo-oesophageal fistula complete correction (associate anterior aortopexy)
Gastro-oesophageal reflux	Niessen fundoplication
Gastro-oesophageal reflux	Niessen fundoplication (associate anterior aortopexy)
Gastro-oesophageal reflux in tetralogy of Fallot	Niessen fundoplication
Respiratory	
Congenital lobar emphysema of left superior lung lobe	Left lung superior lobectomy
Tracheal compression	Anterior aortopexy
Tracheal compression	Anterior aortopexy

the operation. The stent was removed in 2 children after 6 and 25 months, respectively.

The tracheostomy tube was left *in situ* for 18 months. Postoperative bronchoscopic evaluations were customized for each patient according to clinical status, but in general, the patients were followed up according to our protocol, as previously described [7].

A final endoscopic examination (median follow-up 2.9 years, range 0.1–4.6 years) to measure adequate airway patency was conducted in 17 children (94.4%). Thirteen patients (out of 18, 72.2%) no longer required an intrabronchial stent. The remaining patients showed residual mild tracheomalacia that, however, did not affect the airstream. An echocardiogram demonstrated no gradient across the entire aortic course.

DISCUSSION

Vascular compression of the left mainstem bronchus is considered one of the most frequent causes of respiratory distress in infants and children. If one considers that the left mainstem bronchus is smaller than the right bronchus and notes its spatial location in the mediastinum, it is possible to observe its relationship with the surrounding structures along the path from the carina to the hilum [8, 9]. Previous surgery, additional respiratory infections and reactive airway disorders, occurring during growth, may worsen the symptoms related to LMBC [3], making diagnosis challenging.

There are several causes described in the literature that may predispose the left bronchus to compression. Dilatation of the pulmonary arteries, such as that which occurs in the absent pulmonary valve syndrome, was described by Rabinovitch *et al.* [10], while Hahna *et al.* [11] reported a case of respiratory distress in which bronchial compression was due to a dilated ductus arteriosus. Left bronchial compression has also been well documented as a possible complication following surgery, as in the case of oesophageal atresia with tracheo-oesophageal fistula [12]. In children with these conditions, the tracheobronchial wall is exposed to aggressive gastric acid, a situation that causes malacia and the tendency of the airway wall to collapse. The presence of cicatricial adherences and modifications in the relationship among the spatial organs following surgery completes the scenario predisposing the trachea and bronchi to compression by the arterial vessels [5, 6, 10, 11, 13]. Primary bronchomalacia and immunomediated disorders have also been associated with LMBC [3], but whether bronchomalacia is an intrinsic bronchial wall abnormality or occurs due to vascular compression remains to be determined.

It is difficult to diagnose tracheobronchial compression in children, and evaluation of the state of the paediatric airways may require different imaging techniques [5, 14, 15]. Currently, bronchoscopy is considered the gold standard for the diagnosis of tracheobronchial disorders. It allows dynamic visualization of the tracheobronchial tree and of extrinsic pulsatile compression. It can also be used intraoperatively, giving direct feedback to the surgeon during the operation [2, 16]. However, because precise understanding of the anatomy is necessary, CT scans and magnetic resonance imaging remain the most reliable techniques to delineate the airways from the cardiovascular structures [14, 15].

Surgical treatment of airway compression was first reported by Gross and Nehauser [17]. Although several techniques have been described to relieve airway compression [18–20], aortopexy yields the best results in cases of severe, localized tracheobronchomalacia [16, 21].

Several indications for surgical treatment of LMBC are found in the literature [5, 6, 10, 11, 13]; the presence of anomalous vessels that compress the left mainstem bronchus is the most frequent indication of LMBC. An anterior midline location of the descending thoracic aorta is also not infrequent [5, 6] and, in fact, was observed in 22% of cases in our study sample. All of the children in our sample ($n = 4$) with this pattern had previously had operations involving the aortic arch and thoracic aorta (2 had been operated on for aortic coarctation and hypoplastic aortic arch, respectively, and 2 had previously had an anterior aortopexy). In contrast, despite the fact that 38% of our cases were shown to be associated with congenital heart disease, none of them showed dilatation of the cardiac chambers.

Thus, once we established by CT scan that the descending thoracic aorta was the main determinant of LMBC, we chose posterior aortopexy instead of anterior aortopexy, which is used when the compression is due to the innominate artery or the bovine trunk, mobilizing the entire length of the aorta and anchoring it to the vertebral column and surrounding structures. During the manoeuvre, we distributed the traction on a long segment of the thoracic aorta to obtain a harmonious shape and to avoid shrinkage (Fig. 3) [21–23].

Given that LMBC-PAD is an infrequent subgroup of tracheobronchial compression, we consider our results satisfying and in line with previously published reports, particularly those concerning children requiring previous tracheotomy and mechanical ventilation [5, 6, 11, 14]. Furthermore, to the best of our knowledge, our report represents the largest series analysing surgery of the LMBC-PAD.

We are fully aware that our follow-up was too short to give a meaningful analysis and that the children in our study sample comprised a group of very complicated patients, in which the associated pathologies made the interpretation of the results difficult [2]. This is particularly clear if we observe some of the variables, such as mean ventilation time and length of hospital stay, that could have been affected by the associated pathologies (nearly 90% of our patients had associated cardiovascular, abdominal or pulmonary anomalies).

In conclusion, the multidisciplinary approach and the decision to use bronchoscopy to diagnose tracheobronchial disorders proved to be fundamental to the success of this type of surgical programme [24]. Extended posterior aortopexy can be considered a safe and effective treatment for localized LMBC between the descending thoracic aorta and the left pulmonary artery. In fact, evidence of pulsations, as observed in our study sample, excluded treatment with an intrabronchial stent, which would have added an additional force on the tracheobronchial wall, leading to ischaemia.

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