Case Report

Expandable Airway Stents: Success in the Extrinsic Bronchial Compression Approach After Reconstruction of the Aortic Arch

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Introduction

The interruption of the aortic arch is a rare congenital cardiopathy, whose surgical correction generally requires reconstruction of the aortic arch. This intervention can cause distortion of the aortopulmonary space and results in bronchial compression with a consequent pulmonary atelectasis and dependence on ventilatory assistance. The most commonly used therapeutic options for bronchial decompression are not always enough to make pulmonary expansion possible. Our study presents a case of the implant of an endoluminal stent in an infant who, in the post-operative stage of the correction of the interruption of the aortic arch, developed complete atelectasis of the left lung secondary to bronchial compression, with no response after posterior aortopexy.

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Our study presents the case of a 3-year-old, preschool, female patient with a diagnosis of the interruption of the aortic arch and a major interventricular communication, submitted to the reconstruction of the aortic arch at 13 days of life, with end-to-side anastomosis of the descending aorta with the ascending aorta, due to the large distance from the stumps and difference in the diameter of the aortic extremities.

In post-op, the patient presented complete and persistent atelectasis of the left lung (Figure 1A) and was kept on mechanical ventilation with high-level parameters, with no possibility of extubation.

Bronchoscopy suggested an extrinsic compression of the proximal portion of the left bronchus, a hypothesis confirmed by angiotomography, which demonstrated an external vascular compression of the main left bronchus between the left atrium and the descending aorta (Figure 2A).

Despite the tracheostomy performed at two months of age and positive pressure ventilation, the child continued with a collapsed left lung and frequent episodes of hypoxia. At three months of age, she was submitted to aortopexy, which promoted an adequate distancing of the aorta from the bronchus, but this did not result in lung aeration, demonstrating damage to the bronchial structure. A resection of the altered tracheal segment was contraindicated due to technical difficulties and the severity of the ventilatory condition.

At four months, after a multidisciplinary discussion, an expandable mechanical stent (Cordis Palmaz® Blue™ 5x15 mm) was implanted in the left bronchus by the hemodynamic service team (Video 1). Hence, progressive aeration of the affected lung (Figure 1B) led to a decrease in the ventilatory parameters. The child evolved with refractory heart failure due to the drug treatment, and was thus submitted to surgical correction of the ventricular septal defect and an implant of a stent in the anastomosis region of the aorta (Video 1), which proved to be stenotic, with good results.

During bronchoscopy follow-up, an excessive proliferation of the granulation tissue was observed (Figure 3A), causing a progressive obstruction of the bronchial light, with no response to the three attempts of balloon dilatation and consequent total obstruction after three months of stent implant.

As the child presented a good respiratory pattern, using oxygen inhaled through the tracheostomy, she was released at eight months of life, with the proposal of the evaluation of the implant of a new bronchial stent (Dynamic Renal® 5x15 mm).

At approximately one year of life, five months after bronchoscopic confirmation of the total obstruction of the stent, a chest X-ray, performed within an infectious context, showed left lung aeration (Figure 1C).

At one year and six months, the child was admitted to an intensive care unit (ICU) with a severe respiratory failure. The chest X-ray showed a diaphragmatic hernia on the right side and complete aeration of the left lung (Figure 1D), confirmed through chest computed tomography (CT) (Figure 2B) and bronchoscopy (Figure 3B, video 2). After having performed surgery to correct the Morgani hernia, the child was released in good condition, with planning to be gradually weaned off of the tracheostomy.

Discussion

Bronchial compression is a potentially severe complication described in both the pre-operative and post-operative stages of congenital cardiopathies with an obstruction of the aortic arch, such as the coarctation of the aorta, the interruption of the aortic arch, and left heart syndrome. The altered relation between the blood vessels of the arch and the tracheobronchial tree found in these pathologies causes...
**Figure 1** – A) complete atelectasia of the left lung in the post-operative stage immediately after correction of the aortic arch; B) left lung aeration shortly after the endoluminal stent implant in the left bronchus; C) late aeration of the left lung after the spontaneous resolution of the obstruction of the stent; D) right diaphragmatic hernia, left lung aerated.

**Figure 2** – A) extrinsic compression of the main left bronchus (arrow: left lung without aeration; star: aorta compressing the bronchus); B) left lung aeration even in the face of a right diaphragmatic hernia.
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a distortion and straightening of the aortopulmonary space, and can lead to an extrinsic bronchial compression.\textsuperscript{1,2} In this case, this alteration occurred in the geometry and position of the aortic arch after the surgical correction of the interruption.

The knowledge of a possible compression of the airways is of utmost importance, as its incidence can reach 27.2\% in the pre-operative and 33.3\% in the post-operative stages of cardiopathies that require the reconstruction of the aortic arch, according to findings from Jhang et al.\textsuperscript{1}

The respiratory symptoms are generally unclear and may be absent in many cases.\textsuperscript{2,3} In a study conducted by An et al., only 50\% of the patients with airway compression found in CT images presented symptoms.\textsuperscript{2} The bronchoscopy enabled the direct viewing of the compression of the tracheobronchial tree, while the CT enabled the evaluation of the spatial region between the airway and cardiovascular structures, and should therefore be considered, especially in patients with persistent atelectasis and failure to wean off of mechanical ventilation.\textsuperscript{3-5}

The early diagnosis of this medical condition is essential, since a prolonged bronchial compression can cause significant functional involvement and tracheobronchomalacia, which can persist even after the release of extrinsic compression\textsuperscript{2,5}, as observed in the case described above.

If on the one hand the diagnosis of airway compression can be easily performed, the therapeutic handling can be challenging, especially in cases of severe residual malacia. The choice between a surgical option, such as posterior aortopexy, the remodeling of the aortic arch, and transverse extension of the aortic arch using an autograft of the pulmonary artery, as compared to a conservative conduct,\textsuperscript{1,6} should be based on the severity of the obstruction and the clinical condition of the patient.\textsuperscript{2,5} The tracheostomy with prolonged mechanical ventilation has been proposed when one considers the tracheomalacia to be self-limited, with progressive improvement during childhood.\textsuperscript{5} In this case report, even if one had removed the bronchial compression, posterior aortopexy did not result in lung aeration, illustrating damage to the bronchial structure.

In recent years, attention has been geared toward placing the endoluminal stent in airways in severe cases of bronchomalacia that did not respond to the implemented therapy. Barnes et al. described the successful use of the stent in a case of severe bronchomalacia after stage I of the Norwood procedure, which was not reverted after posterior aortopexy.\textsuperscript{7} Arcieri et al. also reported the use of the left bronchial stent in six patients with severe residual bronchomalacia after posterior aortopexy; however, these authors only analyzed the results of aortopexy.\textsuperscript{4}

In the study conducted by Serio et al., the stent implant restored the spontaneous respiration in 20 patients with severe residual malacia after surgery.\textsuperscript{8} Based on these descriptions and faced with the complexity of the case and absence of new surgical proposals, we chose the endobronchial stent implant.

Although the stent implant in airways is a tempting conduct, as it can quickly restore the permeability of the bronchus and enable pulmonary aeration, it is not free of complications, such as migration, fracture, obstruction caused by mucus and granulation tissue, the rupture of the airways, and infection.\textsuperscript{9,10} The formation of the granulation tissue is a constant, since the airway stent did not become epithelated like the endovascular stent.\textsuperscript{5} In the case described herein, we detected the proliferation of serial dilatations with a balloon catheter, but with a partial and spontaneous resolution after nearly five months, most likely after a decrease in the reactional inflammatory process.

Therefore, a recommendation must first undergo a detailed analysis, since the stents are easy to implant, but difficult to remove, especially metallic stents, which should be considered permanent.\textsuperscript{3}

Complication related to the use of the stent indicate the need for protocols to maintain the permeability of the lumen\textsuperscript{9} and for regular follow-up by bronchoscopy and/or CT in order to achieve an early identification of the complications.\textsuperscript{9,10}
Conclusions

Bronchial compression after the reconstruction of the aortic arch is a severe complication, which one may not be able to solve with available surgical interventions. In these cases, the stent implant in the location where the bronchial light is tapered can represent a successful alternative, though reserved for children in a critical condition, whose airway decompression surgery did not result in affective lung aeration.

Author Contributions

Conception and design of the research: Guimarães AFM; acquisition of data: Silva CM, Guimarães AFM, Wang R, Alves CMS, Oliveira EC; analysis and interpretation of the data: Wang R, Alves CMS, Oliveira EC; writing of the manuscript: Silva CM, Guimarães AFM; critical revision of the manuscript for intellectual content: Guimarães AFM, Oliveira EC.

References


*Supplemental Materials

For supplementary figure, please click here.
For supplementary tables, please click here.
See the Supplemental Video 1, please click here.
See the Supplemental Video 2, please click here.

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