Nonaneurysmal rupture 30 years after patch aortoplasty for coarctation

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Aneurysm formation after patch aortoplasty for the repair of coarctation of the aorta is a widely known complication.¹ These patients should be routinely followed up, and surgical treatment is required in case of progressive growth of the aneurysm.² However, there has not been any report of a case of rupture of the aorta in the absence of an aneurysm.

Clinical Summary

A 36-year-old man with sudden transient chest pain and massive pleural effusion was hospitalized by the primary physician. Left-sided hemothorax was suspected, but the cause of the hemorrhage was not detected then because no apparent aneurysm or rupture was noted on chest computed tomography (CT). Inasmuch as the patient was in hemodynamically stable condition, he was referred to our hospital the day after the left-sided hemothorax was detected.

His medical history included 1-stage surgical repair for complex coarctation, patch closure of a ventricular septal defect, ligation of a patent ductus arteriosus, and Dacron patch aortoplasty of preductal type coarctation of the aorta performed in 1975 when he was 6 years old.

CT images obtained on arrival demonstrated a pinpoint extravasation at the descending aorta (Figure 1, A), where the Dacron patch for the aortoplasty was located. Emergency surgical treatment was readied, but hemodynamic collapse occurred on the patient's arrival in the operating room and he died.

CT images with 3-dimensional reconstruction showed later an extravasation at the site of the aortoplasty patch (Figure 1, B). The diameters of the aortic arch and descending aorta were within normal limits (22-26 mm) and there was no aneurysm.

Autopsy findings revealed a 2-mm diameter perforation at the aortic isthmus (Figure 2, A). Histopathologically, the aortic wall around the ruptured site was rather thin (0.5-1 mm) compared with other sites (1.5-2 mm) and consisted of collagen fibers (Figure 2, B).

Figure 1. Enhanced images of the chest CT scan. **A**, An extravasation from the descending aorta was recognized only in a single slice (arrow). **B**, CT with 3-dimensional reconstruction demonstrating extravasation (arrow) at the side of the aortoplasty patch (arrowheads).

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Discussion

The incidence of aneurysm formation after Dacron patch aortoplasty is frequent, ranging from 4% to 51% in the long term.³ Therefore, a lifelong follow-up by radiographic examination is recommended for patients subjected to patch aortoplasty in childhood,^{3,4} and if progressive growth of an aneurysm is recognized, surgical treatment is required.²⁻⁴ However, a rupture in the absence of an aneurysm was not predictable by radiographic imaging.

The aortic wall surrounding the perforation consisted of collagen fibers and lacked the components of the normal aortic wall. This finding is a common factor identified among patients

Figure 2. Postmortem specimen. **A**, Gross examination of the isthmus revealed the pinpoint perforation (*arrow*) located 3 mm from the edge of the aortoplasty patch (*arrowheads*) (left, cranial side). **B**, Histopathologic examination of the ruptured site (*arrow*) demonstrated absence of the elastic fibers and smooth muscle layer between the normal aortic wall (*single asterisk*) and the patch (*double asterisks*). (Elastic-van Gieson stain; original magnifications 6 $\times$.)

with peripatch aneurysm.³ The residual tissue of the coarctation may have led to a disruption of the suture line and was probably one of the causes of the sudden perforated rupture. It was difficult to determine the period when the aortic wall around the perforation became disrupted, perioperatively or gradually in the long term. In either case, it was not clear why histologic disruption of the previous aortic wall remained intact for 30 years without formation of an aneurysm. The participation of adhesional surrounding mediastinal tissue may be assumed.

In conclusion, it seems probable that, even in the absence of an aneurysm, a life-threatening risk may persist for patients subjected to patch aortoplasty for coarctation of the aorta. Second intervention may be warranted to exclude the risk of sudden death for the patients, even without aneurysm, after patch aortoplasty.

References

1. Jenkins P, Ward C. Coarctation of the aorta: natural history and outcome after surgical treatment. *Q J Med.* 1999;92:365-71.
2. Hauser M, Pollock J, Wilson N. Sudden death 10 years after patch aortoplasty for coarctation. *Cardiol Young.* 1998;8:383-5.
3. Parks WJ, Ngo TD, Plauth WH Jr, Bank ER, Sheppard SK, Pettigrew RI, et al. Incidence of aneurysm formation after Dacron patch aortoplasty repair for coarctation of the aorta: long-term results and assessment utilizing magnetic resonance angiography with three-dimensional surface rendering. *J Am Coll Cardiol.* 1995;26:266-71.
4. Roth M, Lemke P, Schonburg M, Klovekorn WP, Bauer EP. Aneurysm formation after patch aortoplasty repair (Vosschulte): reoperation in adults with and without hypothermic circulatory arrest. *Ann Thorac Surg.* 2002;74:2047-50.