## Case Report Étude de cas

# Dysphagia Lusoria: extrathoracic surgical management

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OBJECTIVE: To report a case of dysphagia lusoria managed by an extrathoracic approach.

DESIGN: Case report and literature review.

SETTING: A university hospital.

PATIENT: A 39-year-old man, who presented with weight loss and dysphagia. Aortography and computed tomography revealed an aberrant subclavian artery compressing the esophagus against the aortic arch.

INTERVENTION: The right subclavian artery was divided at its origin and reimplanted onto the right carotid artery. The operation was performed through a right supraclavicular incision without opening the chest.

RESULTS: There was no operative morbidity. Six months postoperatively the patient was asymptomatic and had gained weight. There was no radiologic evidence of esophageal compression.

CONCLUSIONS: Based on the results of our case of dysphagia lusoria and the reports of others that have started to appear in the literature, consideration should be given to repairing a symptomatic, nonaneurysmal aberrant right subclavian artery through an extrathoracic approach.

OBJECTIF: Décrire un cas de dysphagia lusoria traité par voie extrathoracique.

CONCEPTION: Étude de cas et recension des écrits.

CONTEXTE: Hôpital universitaire.

PATIENT : Un homme de 39 ans, qui s'est présenté aux prises avec une perte de poids et une dysphagie. Une aortographie et une scanographie ont révélé qu'une artère sous-clavière aberrante comprimait l'œsophage contre la crosse de l'aorte.

INTERVENTION : L'artère sous-clavière a été divisée à son point d'origine et réimplantée sur l'artère carotide droite. On a effectué l'intervention en procédant à une incision supraclaviculaire droite sans ouvrir le thorax.

RÉSULTATS: Il n'y a pas eu de morbidité opératoire. Six mois après l'intervention, le patient ne présentait aucun symptôme et avait repris du poids. Les examens radiologiques n'ont révélé aucune compression de l'œsophage.

CONCLUSION : D'après les résultats de notre cas de dysphagia lusoria et les comptes rendus d'autres cas qui ont commencé à paraître dans les écrits, il faudrait envisager de réparer une artère sous-clavière droite aberrante non anévrysmale symptomatique par voie extrathoracique.

ysphagia lusoria means difficulty in swallowing caused by a "jest of nature." Its original description links the clinical symptoms of variable degrees of dysphagia to the aberrant origin of the right subclavian artery from the left side of the aortic

arch. The artery courses posterior to the esophagus, causing compression of this structure against the aortic arch. Patients may present at any time from infancy to old age, and there is currently no accepted standard treatment. We recently managed such a case.

#### CASE REPORT

A 39-year-old man with chronic schizophrenia reported a 3-year history of worsening dysphagia. His difficulty swallowing progressed from solids to liquids, with "sticking" in the upper

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thoracic region. These findings were substantiated by a 10-kg weight loss. No abnormalities were detected on physical examination. There was no difference in the blood pressure measured in both arms. A barium swallow revealed compression of the upper third of the esophagus by an extrinsic mass (Fig. 1) and was confirmed by endoscopy, which also showed that there was no intrinsic mucosal lesion. Arteriography demonstrated the presence of an aberrant right subclavian artery arising distal to the left subclavian artery (Fig. 2). There was minimal dilation of the origin of this vessel. Computed tomography demonstrated the esophagus coursing between and being compressed by the right subclavian artery and the aortic arch. Additional motility disorders were ruled out with esophageal manometry. The weight loss and the patient's discomfort indicated operative repair of this condition.

#### Operative details

The patient was positioned supine on the operating table with a sandbag behind his shoulders, extending his neck. Under general endotracheal anesthesia, the chest and the neck were prepared. A 10-cm transverse incision was made 1 cm superior to the right clavicle, extending almost to the midline. The clavicular head of the sternocleidomastoid muscle was divided. The common carotid artery was identified and exposed for 3 to 4 cm. The aberrant subclavian vessel was then located in the neck and its course was followed to the aortic arch (Fig. 3). We were able to encircle the right subclavian artery as it arose from the aortic arch by retracting the esophagus anteriorly. The origin of the artery could be easily palpated, and there was no evidence of aneurysm formation. The patient was given 5000 units of heparin intravenously,

and a vascular clamp was applied to the right subclavian artery at its origin from the aortic arch. This artery was divided and oversewn with a running 5-0 Prolene suture. On removal of the clamp, the arterial stump retracted to the left of the esophagus. The distal segment of the right subclavian artery was sufficiently long to allow its implantation, end to side, into the right common carotid artery (Fig. 4). At the end of the procedure the patient had a palpable right radial pulse and equal blood pres-

sure in both arms. All major branches of the subclavian artery were preserved. At follow-up 6 months later, the patient had regained weight and denied any symptoms of dysphagia. Follow-up barium swallow demonstrated disappearance of the extrinsic mass and no hold-up of barium.

#### DISCUSSION

The most common anomaly of the aortic arch is an aberrant right subcla-

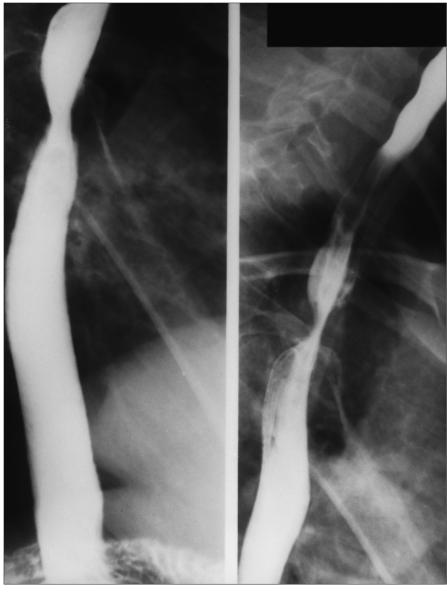


FIG. 1. Barium swallow demonstrates smooth extrinsic compression of esophagus.

vian artery, reported to occur in 0.5% to 1.8% of the population.<sup>2</sup> The right subclavian artery may arise as the last branch of the aortic arch distal to the right and left common carotid arteries and the left subclavian artery. The embryologic origin of the anomaly can be explained by the hypothetical double aortic arch of Edwards.<sup>3</sup> It is hypothesized that the first portion of the aberrant subclavian artery is formed from the persistent right distal aortic arch. This leads to the aberrant course of the subclavian artery behind the esophagus and the aortic arch.

This anomaly is entirely asymptomatic in many patients, and there is no indication for repair of these lesions.

Occasionally extrinsic compression of the esophagus may be caused as the right subclavian artery courses posterior to the aortic arch, leading to dysphagia. It is unclear why some patients present with dysphagia in infancy whereas others do not have symptoms until middle or old age. A variety of other complications of the anatomic variant have been described, including aortoesophageal fistulae,<sup>4</sup> aneurysm formation<sup>5,6</sup> with rupture,<sup>7</sup> arteriovenous fistula formation<sup>8</sup> and respiratory compromise.<sup>9</sup>

This disorder was first described by Bayford<sup>1</sup> in 1794. The first report of repair was that of simple ligation by Gross in 1946.<sup>10</sup> In infancy, the

EB 52
6/1
16/1

Phu
W-E
W-C

FIG. 2. Angiogram shows aberrant origin of right subclavian artery from left side of aortic arch.

most common approach is through a left thoracotomy where the right subclavian artery is divided and usually not reimplanted. In adults, the treatment is more controversial. A variety of approaches have been tried. Both right<sup>4,11</sup> and left<sup>12</sup> thoracotomies have been described as well as median sternotomy13 and combined approaches.14 Many of these approaches provide inadequate exposure and have a high associated complication rate. Concern for ischemia of the arm prompted Bailey, Hirose and Alba15 to reimplant the subclavian artery onto the aortic arch. Orvald, Sheerer and Judge<sup>16</sup> described an extrathoracic approach to the repair of dysphagia lusoria, and this was simplified further by Valentine, Carter and Clagett<sup>17</sup> and Kieffer, Bahnini and Koskas.<sup>18</sup> We concur with their recommendations that an extrathoracic approach through a single neck incision can be used to repair the nonaneurysmal anomaly. This approach may be facilitated by the unusually high position of the subclavian artery in patients with dysphagia lusoria. This exposure in our patient allowed division of the subclavian artery to the left of the esophagus and reimplantation to the carotid artery. In our opinion no additional exposure at the time of operation would have been gained by median sternotomy.

The aberrant right subclavian artery may arise from a dilated or aneurysmal aorta and subclavian segment, which is referred to as a diverticulum of Kummeral.<sup>19</sup> In this case a direct approach with repair of the aneurysm may be indicated.<sup>18</sup> Other anatomic abnormalities may exist with the aberrant subclavian artery, including other arterial anomalies, a non-recurrent recurrent laryngeal nerve and an aberrant thoracic duct.

To date no studies have been done outlining the need for reimplantation

of the subclavian artery; however, we are supported by a number of authors in our recommendation to reimplant the artery to prevent ischemia of the dominant arm and possible subclavian steal syndrome.<sup>1120</sup>

#### Conclusions

For patients with symptomatic dysphagia lusoria related to an aberrant origin of the right subclavian artery, operation is indicated to relieve dysphagia and restore swallowing. Treatment must be individualized to accommodate other abnormalities. In the most common form, our favoured approach is a right supraclavicular incision with division of the nonaneurysmal aberrant subclavian artery to the left of the esophagus and reimplantation directly onto the carotid artery. This avoids potential major

complications associated with thoracotomy or sternotomy. The cervical incision provides adequate exposure of the right subclavian artery and right internal carotid artery. The aortic arch and origin of the right subclavian artery were easily palpable in our case, and retraction of the esophagus anteriorly provided sufficient mobility to encircle, divide and safely oversew the arterial stump of the subclavian artery. When there is no contraindication, such as aneurysm formation, the cervical approach provides excellent exposure for the correction of dysphagia lusoria.

Based on our experience and review of the literature we suggest that the best treatment for dysphagia lusoria is division of the aberrant subclavian artery, through a right cervical incision, with direct reimplantation onto the carotid artery.

#### References

- 1. Bayford D: An account of a singular case of obstructed deglutition. *Mem Med Soc Lond* 1794; 2: 369–374
- 2. Richardson JV, Dotty DB, Rossi NP et al: Operation for aortic anomalies. *Ann Thorac Surg* 1981; 31: 426–432
- 3. Edwards JE: Congenital malformations of the heart and great vessels. Section H. Malformations of the thoracic aorta. In Gould SE (ed): *Pathology of the Heart*, Charles C Thomas Publisher, Springfield, Ill, 1960: 391–462
- 4. Guzzetta PC, Newman KD, Ceithaml E: Successful management of aberrant subclavian artery–esophageal fistula in an infant. *Ann Thorac Surg* 1989; 47: 308–309
- 5. Esposito RA, Khalil I, Galloway AC

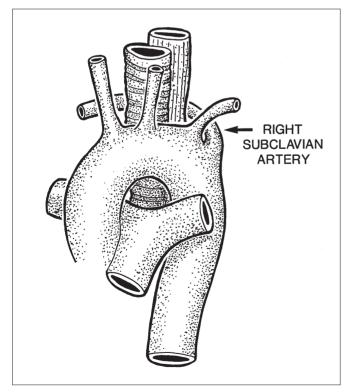


FIG. 3. Preoperative anatomy. Right subclavian artery arises from left side of aortic arch and courses posterior to esophagus, compressing it against aortic arch.

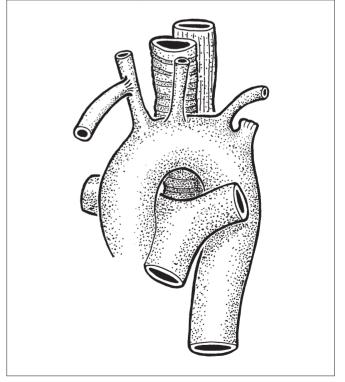


FIG. 4. Postoperative correction of dysphagia lusoria. Note oversewn stump of subclavian artery and its reimplantation into right common carotid artery.

- et al: Surgical treatment of aneurysms of aberrant subclavian artery based on a case report and review of the literature. *J Cardiovasc Surg (Torino)* 1988; 95: 888–891
- 6. Hardy JD: Aneurysm of aberrant right subclavian artery and Odine's curse. *J Cardiovasc Surg (Torino)* 1989; 97: 319–320
- 7. Kullnig P: Case of the season. Semin Radiol 1989; 24: 75–76
- 8. Wong CK, Lau CP, Leung WH: An arteriovenous fistula from an aberrant right subclavian artery to the superior caval vein in a congenitally myxoedematous adult. *Int J Cardiol* 1989; 25: 126–129
- 9. Jebara VA, Arnaud-Crozat E, Angel F et al: Aberrant right subclavian artery aneurysm: report of a case and review of the literature. [review] *Ann Vasc Surg* 1989; 3: 68–73

- 10. Gross RE: Surgical treatment of dysphagia lusoria in the adult. *Ann Surg* 1946; 124: 532–534
- 11. Hallman GL, Cooley DA: Congenital aortic vascular ring. *Arch Surg* 1964; 88: 666–675
- 12. Lichter I: The treatment of dysphagia lusoria in the adult. *Br J Surg* 1963; 50: 793–796
- Shumacker HB, Isch JH, Finneran JC: Unusual case of dysphagia due to anomalous right subclavian artery. *J Thorac Cardiovasc Surg* 1971; 61: 304–308
- 14. Siderys H: A new operation for symptomatic aberrant right subclavian artery in adult (dysphagia lusoria). *J Thorac Cardiovase Surg* 1969; 57: 269–272
- 15. Bailey CP, Hirose T, Alba J: Re-establishment of the continuity of the anomalous right subclavian artery af-

- ter operation for dysphagia lusoria. *Angiology* 1965; 16: 509–513
- Orvald TO, Sheerer R, Judge JR: A single cervical approach to aberrant right subclavian artery. Surgery 1972; 71: 227–230
- 17. Valentine RJ, Carter DJ, Clagett GP: A modified extrathoracic approach to the treatment of dysphagia lusoria. *J Vasc Surg* 1987; 5: 498–500
- 18. Kieffer E, Bahnini A, Koskas F: Aberrant subclavian artery: surgical treatment in thirty-three adult patients. *J Vasc Surg* 1994; 19: 100–111
- 19. Salomonowitz E, Edwards JE, Hunter DW et al: The three types of aortic diverticula. *AJR* 1984; 142: 673–679
- Stoney WS, Alford WC, Burrus GR et al: Aberrant right subclavian artery aneurysm. *Ann Thorac Surg* 1975; 19: 460–467

### Concours de dissertation en éthique médicale Logie Date limite : le 1<sup>er</sup> juin 1996

Le Journal de l'Association médicale canadienne (JAMC) parraine de nouveau le Concours de dissertation en éthique médicale Logie ouvert aux étudiants de premier cycle en médecine des universités canadiennes. Cette année, les prix sont de 1500 \$ pour le lauréat, de 1000 \$ pour la deuxième place et de 750 \$ pour la troisième. Le JAMC se réserve cependant le droit de suspendre certains prix ou la totalité de ceux-ci si la qualité des textes est jugée insuffisante. Le jury est formé d'un groupe de rédacteurs de l'équipe scientifique et de celle des informations générales du JAMC qui choisiront les lauréats en fonction du contenu, du style de rédaction et de la présentation des manuscrits. Les dissertations doivent être dactylographiées à double interligne et compter au maximum 2500 mots, y compris les références. Les citations et les références doivent être conformes aux «Exigences uniformes pour les manuscrits présentés aux revues biomédicales» (voir JAMC 1995; 152 : 1466–1473). Les auteurs choisis devront remettre leur dissertation sur une disquette. Les dissertations choisies seront remaniées quant à la longueur et à la clarté, et conformément au style de la revue. Les auteurs recevront une copie remaniée avant la publication. Veuillez faire parvenir vos textes à l'attention du Rédacteur aux informations générales, JAMC, CP 8650, Ottawa ON K1G 0G8.



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