Just when you thought you'd seen everything

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We read with some interest the report submitted by Ditah and colleagues¹ in this issue of the *Journal*, which reviews a case of hemoptysis in a 19-year-old woman. The story sounded familiar, and even after skimming over the title, the answer seemed to be as well rehearsed as the latest production of *Phantom of the Opera* (now 30 years into its iconic run). Hemoptysis in a 19-year-old? Carcinoid, mucoepidermoid tumor, broncholith, lymphoma, foreign body, arteriovenous malformation, sequestration, trauma, pulmonary embolism, sarcoma, adenoma, trauma, and—reaching way out into left field—even Eisenmenger syndrome or perhaps Castleman disease are the usual suspects. Then, of course, there is always the misdirection play in which epistaxis is the actual root cause.

As we continued reading, however, all of our certainty dissipated. The comprehensive differential diagnosis of hemoptysis, of which we were so proud and that seemed so exhaustive to us, unfortunately did not include intrapulmonary teratoma. Ditah and colleagues¹ had identified a vascularized right middle lobe tumor. Embolization was initially performed, and then on subsequent resection a mature teratoma was found.

Intrapulmonary teratomas have been described before, but one being heralded by hemoptysis is novel and enlightening. Trichoptysis (expectoration of hair) is the usual Final Jeopardy answer and is pathognomonic for teratoma. Hemoptysis is rarely mentioned in the existing literature.

There are a few other noteworthy points. It has been our institutional practice to perform tumor embolization for systemically vascularized thoracic lesions before resection to decrease intraoperative blood loss and facilitate a safer resection. Some argue about the utility of such a practice, but we have seldom regretted being *too* well prepared for an operation.

We might differ from Ditah and colleagues,¹ however, on one operative point. The authors initially describe a video-assisted thoracoscopic surgery approach, followed by conversion to eventual thoracotomy. We perhaps might have gone straight to the thoracotomy in this case, although

Siva Raja, MD, PhD (left), and Sudish C. Murthy, MD, PhD (right)

Central Message

This report on the case of an intrapulmonary teratoma describes an uncommon presentation (hemoptysis) of a rare tumor. It highlights the management of systemically vascularized intrapulmonary lesions.

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we did appreciate the less invasive attempt. Our experience suggests it is seldom how you get there that is a problem (especially in a 19-year-old), but rather what you do when you are there! Also in our experience, we have used a video-assisted thoracoscopic surgery exploration in these cases to rule out pleural metastasis and generally survey the hemithorax, but have used an open approach for safety reasons. Embolization is often only partially effective, and, paradoxically, an open procedure and easy sponge stick access is more comforting to us for these difficult cases.

In this era of large, randomized, controlled trials, studies of rare diseases and lesions are frequently relegated to scattered case reports and small case series in throwaway journals often not in the English language. Over time, however, even if on the order of decades, these curious, yet spurious, reports eventually amount to an accumulated experience that translates into important information and valuable insight.

Common presenting symptoms in the setting of a common clinical picture should not distract the clinician from entertaining rare clinical entities. Those who think that they know it all don't know enough, and those who recognize that they don't know enough never stop learning.

Reference

1. Ditah C, Templin T, Mandal R, Pinchot JW, Macke RA. Isolated intrapulmonary teratoma. *J Thorac Cardiovasc Surg*. 2016;152:e129-31.