Editorial

Putting on airs again: new insights and questions about spontaneous pneumomediastinum recurrence

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Spontaneous pneumomediastinum is a benign condition that is problematic because it is frequently mistaken for esophageal perforation (1). Reported recurrence rates are typically around 1%. Due to the low incidence of spontaneous pneumomediastinum combined with the apparent rarity of recurrence, the clinical aspects of recurrence are essentially unknown. In a recent issue of the *Journal of Thoracic Disease*, Kumeda *et al.* report a retrospective case series with a striking 17% rate of recurrent spontaneous pneumomediastinum. Their results provide interesting new insights and raise important questions about the disease (2).

Kumeda *et al.* conducted a single-institution retrospective review to assess clinical factors and characteristics associated with spontaneous pneumomediastinum recurrence. Using their institutional database, they found 30 cases of spontaneous pneumomediastinum between January 2012 and December 2021. They used phone follow up to identify 5 cases of recurrent spontaneous pneumomediastinum. Most demographic parameters, including young age and male predominance (87% male) correlated well with observations of prior studies (1-3). The presenting signs and symptoms, including chest pain (70%), dyspnea (23%), and neck or throat pain (50%) were also similar to prior reports (1).

Spontaneous pneumomediastinum patients often have a history of Valsalva events such as cough, emesis, and strenuous activity. However, some patients have no such apparent "trigger factor". Kumeda *et al.* observed similar trigger factors, including sports activity and vomiting. They also observed that nearly half of the patients had no known inciting event. Taken together with the demographics and presentation, these results are generally consistent with prior studies (1,4,5).

While many of the findings of Kumeda *et al.* are consistent with prior studies, the high rate of recurrence is not. A recurrence rate of nearly 17% is a striking finding. This disparity in recurrence rates between prior studies and the results of Kumeda *et al.* raises several interesting questions. Is it possible the high rate is related to the small sample size? Or is the recurrence rate of spontaneous pneumomediastinum much higher than previously recognized? Perhaps the phone follow-up identified recurrences that would not have been captured by chart review alone? Alternatively, there may be a component of recall bias by the patients. Further studies are needed to clarify these questions. A prospective study would likely be required to best address such questions. However, the low incidence of spontaneous pneumomediastinum and low rate of recurrence pose challenges to such studies. A multi-institutional study would likely be required to adequately address these questions.

Spontaneous pneumomediastinum is primarily a disease of youth. The mean patient age in a review of nineteen case series was 23 years (1). In the current study, patients ranged in age from 12 to 26 years (median age 16 years). All the recurrences found by Kumeda *et al.* occurred in patients aged 16–18 years. Is this finding simply a reflection of the relatively high proportion of pediatric

cases in the study? Or should consider the possibility of differences in recurrence rates in pediatric versus adult patients? In other words, is the pathophysiology of pediatric and adult spontaneous pneumomediastinum different? In a large pediatric case series of spontaneous pneumomediastinum containing 87 patients, Wong *et al.* showed differences in pathophysiology within the pediatric population (6). All patients under age 6 had secondary spontaneous pneumomediastinum (e.g., from bronchial asthma), whereas only 40% of patients aged >6 years had secondary spontaneous pneumomediastinum. With these considerations in mind, we must ask ourselves if there are also differences in the pediatric and adult spontaneous pneumomediastinum patient populations. Could such differences explain the higher incidence of recurrence in the current study population?

Several other interesting observations on spontaneous pneumomediastinum recurrence were made by the authors. For example, none of the recurrences had a known "trigger activity" such as cough. As noted earlier, the absence of a trigger factor is not in and of itself unusual. However, it does seem surprising that none of the recurrences were associated with a trigger factor. Furthermore, all recurrences occurred within one year of the initial episode and all recurrences were reportedly similar, or less severe, than the index episode. Finally, one of the most notable findings was that all the recurrences were in patients who had pre-existing lung disease (e.g., asthma). This intuitively makes sense. However, this apparent association has not been reported until now. While these results may be prone to recall bias and other errors, it is tempting to speculate that they may be hinting at the underlying pathophysiology of recurrent spontaneous pneumomediastinum. Whether these associations are borne out in larger studies remains to be seen.

Kumeda *et al.*'s results emphasize of the importance of improving our understanding of spontaneous pneumomediastinum. Their findings also highlight interesting issues regarding management of spontaneous pneumomediastinum. For example, patients presenting with spontaneous pneumomediastinum should be counseled on the possibility of recurrence and to notify future providers of their history. In particular, patients with risk factors for recurrence, including asthma, should be counseled on the risk of recurrence. By the same token, we should be inquiring about past episodes in all patients presenting with signs, symptoms and imaging suggestive of spontaneous pneumomediastinum. Documentation of spontaneous pneumomediastinum as a diagnosis in the electronic medical record is logical, but in our experience this documentation is inconsistent at best. Improved documentation could benefit patients who present with recurrence by providing the important history for clinicians during diagnosis and management. The many questions raised by Kumeda *et al.*'s findings are an indication of how much we have to learn about the diagnosis and management of spontaneous pneumomediastinum. Due to the rarity of spontaneous pneumomediastinum, and the limitations of retrospective analysis, many questions will be difficult to answer without prospective multi-institutional studies.

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