

Pediatric spontaneous retropharyngeal emphysema: A case report and comparative analysis of the existing literature



Enfisema retrofaríngeo espontáneo pediátrico: reporte de un caso y análisis comparativo de la literatura existente en población pediátrica

Dear Editor:

Spontaneous retropharyngeal emphysema (SRE) is characterised by the presence of ectopic air at the retropharyngeal level without a clear underlying aetiology. It is a diagnosis of exclusion and considered very infrequent, with few isolated cases reported in the scientific literature. To date, cases in the paediatric population have been exceptional.^{1–5}

We present the case of a boy aged 8 years with an unremarkable medical history assessed in the emergency department for chest pain radiating to the neck with sudden onset while playing football. The patient reported worsening pain when lying down or eating. He reported no recent trauma or foreign body ingestion. The physical examination revealed that the patient was in good health with haemodynamic and respiratory stability. The patient had no fever at any point. There was no evidence of bone abnormalities, although subcutaneous crepitus was detected in the cervical region. The tonsillar pillars were not swollen and the uvula was not deviated. Blood tests showed leucocytosis and neutrophilia without elevation of acute phase reactants. A lateral cervical radiograph was taken, revealing the presence of air in the retropharyngeal space, without an increase in the size of the space (Fig. 1, left). The nasolaryngoscopic exam was normal. A barium swallow was not performed. The patient was hospitalised for 24 hours, receiving intravenous analgesia and fluids, but not supplemental oxygen, and his symptoms resolved. A radiograph performed on an outpatient basis 15 days later evinced near-full resolution of the abnormalities (Fig. 1, right).

Table 1 details the cases of paediatric SRE published to date. It can be seen that the majority occurred in female adolescents and that in many cases there was an identifiable trigger (cough or sports). Two of the patients experienced voice changes. Most had onset with sudden chest or neck pain. The presence of fever or low-grade fever was documented in 3 cases, and leucocytosis and neutrophilia in 1. The nasolaryngoscopic exam only revealed a slight protrusion of the posterior pharyngeal wall in 1 patient, and was normal in all others. Three patients underwent a CT scan. All cases were managed conservatively and had a favourable outcome.

In the case reported by Lee et al.,⁵ a female patient with asthma, the presentation was atypical, with increased work



Figure 1 Left: lateral cervical radiograph evincing the presence of ectopic air in the retropharyngeal space (white arrow). Right: lateral cervical radiograph obtained 15 days later showing near-full resolution of abnormal features.

of breathing, desaturation and pneumomediastinum. We suppose that this patient probably presented with pneumomediastinum secondary to asthma that eventually extended to the cervical region, and therefore we do not consider this a true case of SRE.

The literature on the aetiology, pathogenesis and pathophysiology of SRE is scarce. Macklin et al.⁶ published an article in which they proposed that the aetiology could involve the formation of pulmonary interstitial emphysema secondary to an overdistension of marginal alveoli and decrease in calibre of perialveolar vessels, generating a pressure gradient between both structures that would cause alveolar rupture at the basal level and leaking of air into the interstitial space. The air would spread along the path of least resistance through the perivascular sheaths, chiefly to the mediastinum (as seen in the cases described by Cho et al.³ and Lee et al.⁵) and potentially reaching the cervical region.

As regards the overdistension of marginal alveoli, the coughing physical activity described in the cases included in the series could have contributed to triggering this pathophysiological mechanism. Similarly, this mechanism could be involved in the case of pneumomediastinum associated with asthma described by Lee et al.⁵

In conclusion, SRE seems to be a distinct albeit infrequent entity predominantly found in female adolescents. Its management includes nil per os, intravenous fluids, analgesia and rest, and supplemental oxygen may be an additional option. Although it is a benign disease, it is imperative to remember that it is a diagnosis of exclusion and that all potential causes of ectopic air in the retropharyngeal space need to be ruled out, especially those that require urgent treatment, such as oesophageal perforation. Surgical intervention should be considered in the event of complications like mediastinitis or airway obstruction.

Table 1 Cases of spontaneous retropharyngeal emphysema reported in the paediatric population and its clinical characteristics.

Autor(es)	Relevant history	Sociodemographic characteristics	Trigger	Clinical presentation	Clinical or laboratory abnormalities	NLC	CT	Treatment	Outcome
Marín Garrido et al. (2003) ⁴	None	Male. 13 years	Sports	Sudden chest and neck pain. Voice changes	Low-grade fever (37.5°C). BT results not reported	Mild protrusion of posterior laryngeal wall	Yes	Rest. Intravenous antibiotherapy	Favourable (radiological resolution in 48 h)
Lee et al. (2005) ⁵	Asthma	Female. 12 years	None	Respiratory distress. Chest pain. Voice changes	Desaturation, tachypnoea, fever (38.2°C)	Normal	No	Rest. Oxygen. Antibiotics. Bronchodilators	Favourable (resolution of symptoms in 7 days)
Haro et al. (2014) ²	None	Female. 14 years	Coughs	Sudden cervical pain	Fever (39°C). Normal BTs	Normal	No	Rest. Analgesia. Supplemental oxygen	Favourable
Cho et al. (2016) ³	None	Female. 14 years	Coughs	Odynophagia	Afebrile. BT results not reported	Normal	Yes	Rest. Nil per os. Supplemental oxygen. Intravenous antibiotherapy	Favourable (3 day hospitalization)
Chi et al. (2018) ¹	None	Female. 13 years	None	Cervical pain	Afebrile. Leucocytosis. Neutrophilia	Normal	Yes	Analgesia. Monitoring. Nil per os	Favourable (resolution of symptoms in 7 days)
Arredondo Montero et al. (2022)	None	Male. 8 years	Sports	Sudden cervical pain	Afebrile. Leucocytosis. Neutrophilia	Normal	No	Analgesia. Monitoring. Nil per os	Favourable (24-h hospitalization, radiological resolution in 15 days)

BT, blood test; CT, computed tomography; NLC, nasolaryngoscopy.

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Recurrent *Clostridium difficile* infection treated with bezlotoxumab*



Infeción recurrente por *Clostridium difficile* tratada con bezlotoxumab

Dear Editor:

Infection by *Clostridioides* (formerly *Clostridium*) *difficile* is a disease mediated by toxins. This pathogen is a strictly anaerobic gram-positive rod. Its main virulence factors are toxins A and B, encoded by the *tcdA* and *tcdB* gene, respectively.¹ Infection by *C. difficile* is the leading cause of nosocomial diarrhoea associated to the use of antibiotics and its incidence is increasing in the community, which constitutes a significant public health problem due to the associated morbidity and costs.²

The European Society of Clinical Microbiology and Infectious Diseases defines infection by *C. difficile* as a compatible clinical picture such as diarrhoea, ileus and toxic megacolon in combination with either microbiologic evidence of free toxins in stool or the presence of toxigenic *C. difficile* in stool without reasonable evidence for an alternative aetiology. Recurrent *C. difficile* infection is defined as recurrence of symptoms within 8 weeks after the onset of a previous episode, and severe infection as an episode result-

ing in need for intensive care unit admission, colectomy or death.³

The usual treatment of *C. difficile* is metronidazole or oral vancomycin, for both mild and severe cases, and rifaximin is a widely accepted alternative, in addition to faecal microbiota transplantation in recurrent cases.³ Strategies have been developed to reduce the likelihood of recurrence, including bezlotoxumab, an antitoxin B human monoclonal antibody.⁴

Bezlotoxumab was approved for use in humans based on the outcomes of the MODIFY I and II international multicentric, double-blind randomised placebo-controlled trials.⁵ These studies demonstrated that the use of bezlotoxumab (10 mg/kg as a single dose) was associated with a greater reduction in the recurrence of *C. difficile* infection and a similar safety profile in adults compared to placebo.

The MODIFY trials were conducted in the adult population (>18 years) treated with standard antibiotherapy (metronidazole, oral vancomycin or fidaxomicin) and showed that its effect was greater in individuals with at least 1 risk factor.⁶ The risk factors for recurrence were: age 65 years or greater, history of infection by *C. difficile* in the past 6 months, compromised immunity, severe *C. difficile* infection or isolation of a strain associated with poor outcomes.⁶ In addition, it showed that bezlotoxumab reduced the recurrence of infection by *C. difficile* for a period of 12 weeks.⁴ Bezlotoxumab has yet to be approved for use in the paediatric population.

We present the case of a female adolescent aged 12 years with non-Hodgkin lymphoma at the prepyloric level causing pyloric stenosis that precluded enteral nutrition (complete liquid diet). The patient had 3 episodes of *C. difficile* infection over a 3-month period. Both the initial infection and the

* Previous presentation: case presented at the XIV National Congress of Paediatric Haematology and Oncology.