# Pseudostratified ciliated metaplasia of the distal esophagus diagnosed at adolescence

An 11-month-old infant with recurrent respiratory infections was diagnosed with isolated congenital left pulmonary agenesis. He presented with gastroesophageal reflux disease at the same time, and was treated with prokinetics and a proton pump inhibitor (PPI) for 1 year.

When the boy was 15 years of age, endoscopy was performed because of recurrent episodes of dysphagia, heartburn, food impaction, and cough. A 3-cm circular nodular aspect of the distal esophagus suggested the presence of Barrett's esophagus. Esophageal biopsies confirmed focal intestinal metaplasia. Long-term PPI relieved the symptoms. Repeated esopha-

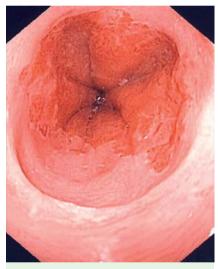


Fig. 1 Endoscopic view of the distal esophagus: circumferential Barrett's esophagus, 3-cm in length.

geal biopsies 1 year later showed no more intestinal metaplasia but fundictype gastric metaplasia. Heartburn and food impaction reappeared after PPI was stopped.

When the boy was aged 17 years, 24-hour pH-metry was normal, whereas endoscopy showed a 3-cm-long circumferential Barrett's esophagus ( Fig. 1).

Histology identified a fundic-type gastric metaplasia with focally ciliated pseudostratified metaplasia (> Fig. 2).

After a 13-month follow-up, the patient remained asymptomatic while receiving PPI.

Ciliated epithelium of the esophagus has been described since 1876 in human embryos [1]. Several authors considered multilayered epithelium in the esophagogastric junction (EGJ) mucosa to be a precursor of Barrett's esophagus in adults [2,3]. This entity was called a squamous metaplasia-like change, with both a similar appearance and immunohistochemical profile as respiratory bronchial epithelium [4]. For these authors, the squamous metaplasia-like change represents a metaplastic change, similar to the pancreatic acinar metaplasia sometimes seen at the EGJ; the mature form of this change may be ciliated pseudostratified epithelium [4,5]. In our case, a congenital rather than acquired origin can also be argued because of the association with pulmonary agenesis. However, although patchy lesions cannot be formally ruled out, the sequence of the biopsies suggest an evolutive lesion, which we consider a potential precursor of Barrett's esophagus. We recommend prolonged PPI treatment and regular endoscopic follow-up of such patients.

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Competing interests: None

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#### References

- 1 Neumann E. Flimmerepithel im Oesophagus menschlicher Embryonen. [in German] Arch Microskop Anat 1876; 12: 570-574
- 2 Glickman JN, Chen YY, Wang HH et al. Phenotypic characteristics of a distinctive multilayered epithelium suggests that it is a precursor in the development of Barrett's esophagus. Am J Surg Pathol 2001; 25: 569 - 578
- 3 Shields HM, Rosenberg SJ, Zwas FR et al. Prospective evaluation of multilayered epithelium in Barrett's esophagus. Am J Gastroenterol 2001; 96: 3268 - 3273
- 4 Takubo K, Vieth M, Honma N et al. Ciliated surface in the esophagogastric junction zone: a precursor of Barrett's mucosa or ciliated pseudostratified metaplasia? Am J Surg Pathol 2005; 29: 211-217
- 5 Wang HH, Zeroogian JM, Spechler SJ et al. Prevalence and significance of pancreatic acinar metaplasia at the gastroesophageal junction. Am J Surg Pathol 1996; 20: 1507 -1510

#### **Bibliography**

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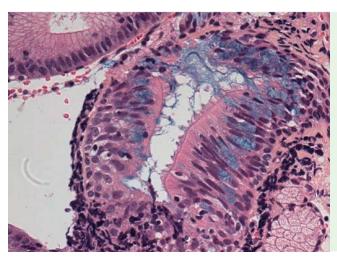


Fig. 2 Histological aspect of a biopsy obtained from the distal esophagus: ciliated and mucosecreting pseudostratified epithelium surrounded by columnar epithelium (hematoxylin and eosin astra blue stain, × 400).