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Case Report

Multiple congenital esophageal strictures masquerading as gastroesophageal reflux disease in an infant: a case report

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ABSTRACT

Congenital esophageal stenosis (CES) is a rare congenital disorder, generally presenting as vague complaints of dysphagia, vomiting and frequent respiratory complaints with symptoms usually appearing with introduction of complimentary feeding. We present the case of an eight-month-old male infant, presenting with regurgitation of feeds, recurrent episodes of cough and poor weight gain. He was diagnosed as a case of isolated multiple membranous esophageal strictures and underwent serial endoscopic esophageal dilatations which subsequently allowed him to swallow solid food and achieve good weight gain.

Keywords: Congenital esophageal stenosis, Esophageal stricture, Esophageal dilatation, Infant, Dysphagia

INTRODUCTION

Congenital esophageal stenosis (CES) is a clinical condition denoted by fixed functional intrinsic narrowing of the esophagus, occurring since birth.[1] Its incidence is approximately 1 in 25000–50000 live births and has been seen in association with other congenital malformations also.[2] It might present with dysphagia, vomiting or respiratory complaints, usually with introduction of complimentary feeding. The treatment may vary from endoscopic dilatation of the stricture to surgical resection, depending upon the type of stricture.

CASE REPORT

We present a case of an 8-month-old developmentally normal male infant, presenting with regurgitation of feeds, drooling, recurrent episodes of cough and poor weight gain. Born normally at term to a non-consanguineously married Hindu couple, his birth weight was 3.2 kilograms (kg). He was referred from peripheral health center with above complaints at 8-months of age. At presentation his weight was 5 kg. He was initially asymptomatic, however his symptoms increased with age and further worsened when complimentary feeding was started at 6 months of age, forcing the parents to almost exclusively milk feed the baby. There was history of regurgitation of un-curdled milk soon after intake. He was being treated as a case of gastroesophageal reflux disease with proton pump inhibitors outside. Initial radiographs did not show any abnormality. Suspecting a structural cause in esophagus, an upper gastrointestinal endoscopy was done, which showed multiple strictures in the middle esophagus with extremely narrow lumen, not permitting the scope to pass (Figure 1). Fluoroscopic examination confirmed multiple strictures (Figure 2). These were membranous strictures. Histological examination of esophageal mucosal biopsy was unremarkable. He had low hemoglobin levels (7.2 g/dl), rest all routine investigations were within normal limits. His screening for other congenital anomalies including abdominal ultrasonography and ECO examination was normal. He was also started on oral iron

supplements. He underwent six endoscopic dilatations (every four weekly) using Savary-Gillard dilators, that subsequently allowed him to swallow solid foods and achieve good weight gain (11 kg at 15 months of age).

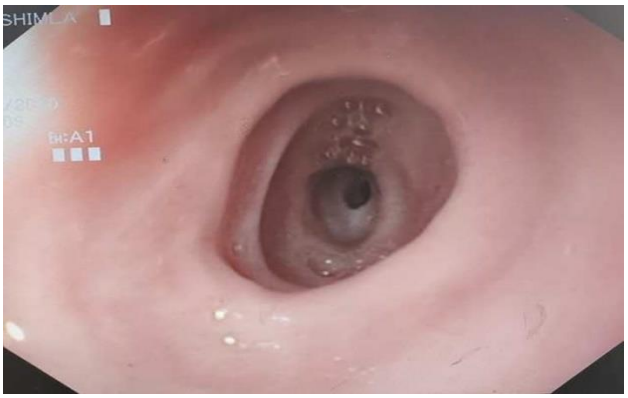

Figure 1: Multiple esophageal strictures seen on Upper gastrointestinal endoscopy.

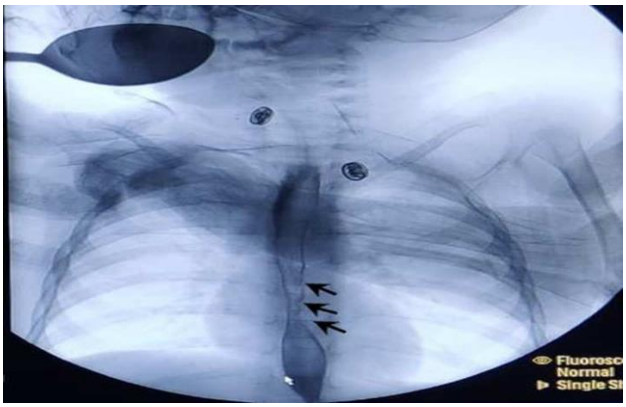

Figure 2: Fluoroscopic image showing multiple esophageal strictures (indicated by black arrows).

DISCUSSION

CES is a rare anomaly in pediatric patients and multiple CES are reported even rarer.[2] Depending on the severity of the stenosis, CES generally becomes symptomatic at around 4 to 10 months of age with introduction of complimentary feeds. Children usually present with failure to thrive, regurgitation of feeds, hypersalivation, foreign body impaction and even respiratory distress during feeding.[3] It is seen more commonly in males, with mean age of onset of symptoms being seven months. [4] CES has been noted to occur more in white populations.[2]

The nature of regurgitant/vomited material can give a clue to site of its origin, like in our case, the presence of uncurdled small volume regurgitant material, generally comes from esophagus, while large volume non-bilious curdled regurgitant/vomitus is often from stomach and bilious vomitus has origins from duodenum. CES is classified into three types – tracheobronchial remnants (TBR), fibromuscular thickening or fibromuscular stricture (FMS) and membranous webbing or esophageal membrane (MS).[5] FMS and EM can occur simultaneously in a patient. TBR occurs due to developmental anomaly and contains cartilage, seromucous glands respiratory epithelium, and non-inflammatory lymphoid tissue. There may be a mechanical obstruction in the esophagus due to cartilage or lymphoepithelial tissue, or it may be functional obstruction due to an aperistaltic segment. In FMS there is a localized hypertrophy of muscular layer and diffuse fibrosis in esophagus along with dysmotility. EM has a normal squamous epithelium and muscular layer. EM is commonly seen in upper or middle third of the esophagus, FMS in middle or lower third, and TBR is seen mainly the lower third of esophagus (within 1 cm of the gastroesophageal junction).[1]

CES has been associated with other congenital malformations also, the most common being esophageal atresia (with or without tracheoesophageal fistula). Cardiac anomalies, anorectal malformations, intestinal atresia, diaphragmatic hernia, duodenal duplication, Meckel's diverticulum, tracheomalacia, chromosomal anomalies (trisomy 21), vesicoureteral reflux, microphthalmos, microgastria, Apert syndrome, palatal cleft, and hemangioma are some of the other malformations associated with CES.

The recommended treatment modality for CES is endoscopic dilatation and surgery. Endoscopic dilatation is effective in MS and FMS, while surgical repair (resection of stenotic segment with end-to-end anastomosis) is required in TBR. Even after a successful treatment, dysphagia occurs frequently, warranting repeated endoscopy sessions and a long follow-up.[6]

CONCLUSION

CES is a rare pediatric anomaly presenting with dysphagia, vomiting, poor weight gain, regurgitation of feeds and respiratory symptoms during feeding, usually occurring with introduction of complimentary feeds.

The nature of vomitus can give us a clue to site of its origin e.g. uncurdled small volume milk usually comes from esophagus. Membranous esophageal stricture has good response to serial endoscopic esophageal dilatations.

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