Title Page

A Rare Cause of Dysphagia

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Maryiam Wajid: Revision of manuscript
Oscar W. Cummings: Provision of pathology images

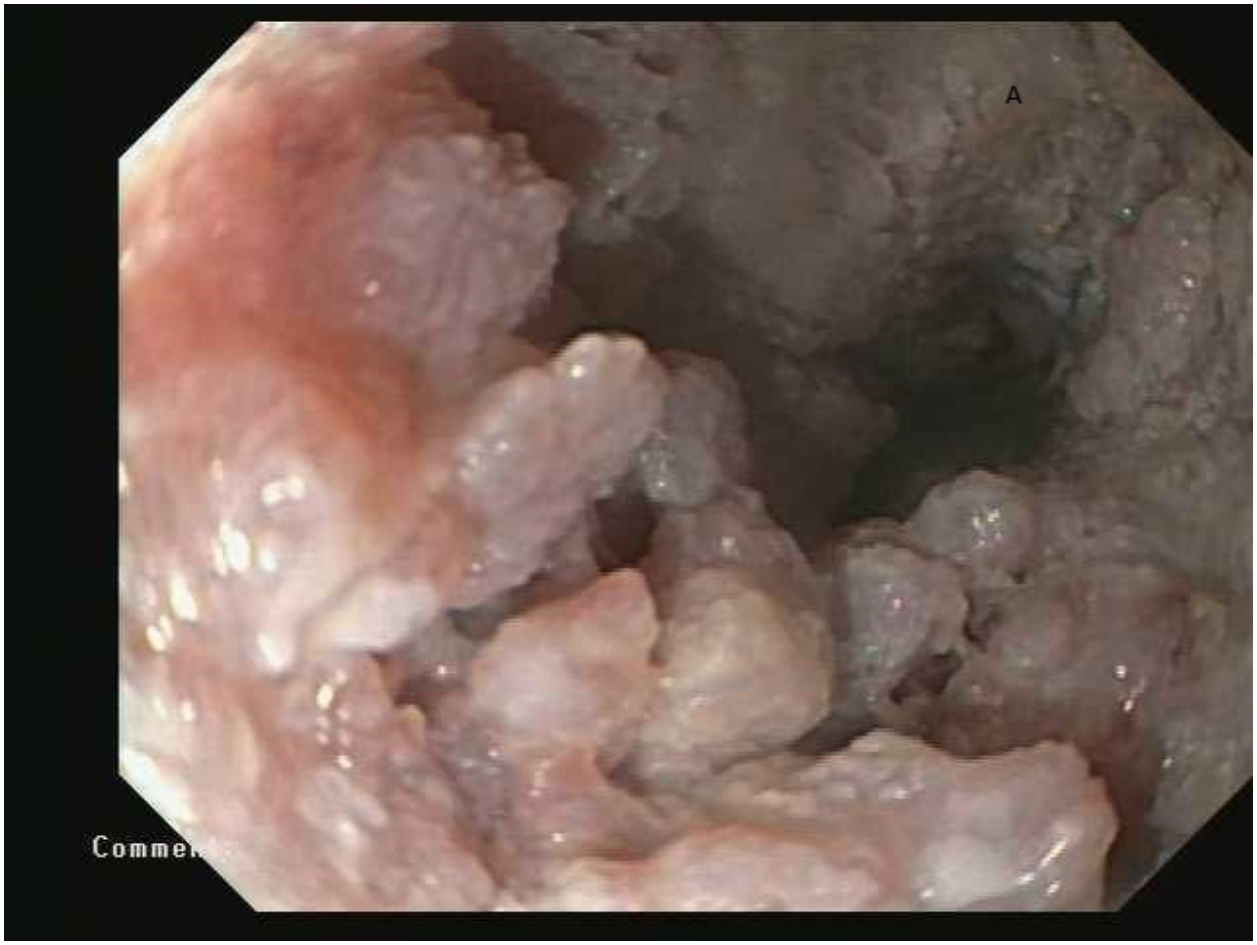
A Rare Cause of Dysphagia

Question: An 86 year old male presented with complaints of dysphagia to solids for 6 years. He was under the care of a general surgeon for the past few years and underwent serial esophageal dilations, however his dysphagia got progressively worse. He reported having solids getting stuck in his esophagus needing to be washed down with liquids. He said that this was an everyday problem and was interfering significantly with his quality of life. Medical history revealed coronary artery disease, congestive heart failure with defibrillator in place, hypertension, cerebrovascular accident, chronic obstructive pulmonary disease and osteoarthritis. He was on aspirin, carvedilol, digoxin, dofetilide, losartan, omeprazole, warfarin, hydrocodone and aclidinium inhaler. He did not smoke cigarettes or drink alcohol, however he did have a previous history of smoking for several years. He did not report any significant weight loss. He underwent an upper endoscopy that showed keratinized esophageal mucosa with whitish discoloration and texture change in the lower third with papillary and filiform protrusions, involving a 6 cm segment of causing significant resistance to upper scope passage (figures A and B). Dilation was performed with Savary dilators up to 51Fr without any improvement in dysphagia. Biopsies showed candidiasis and keratinized squamous hyperplasia. He was treated with a course of fluconazole without any relief in symptoms. He then underwent an endoscopic mucosal resection (EMR) for diagnostic and therapeutic purposes with lovenox bridging.

Answer: Carcinoma Cuniculatum

Pathology from the EMR revealed a well differentiated squamous cell carcinoma, carcinoma cuniculatum. Post EMR, his symptoms improved somewhat but not significantly. Given the keratinization of the mucosa, EMR was technically difficult. He was deemed a poor surgical candidate given his age and co-morbids, however, he continued to have significant symptoms. It was decided to perform cryotherapy for symptom alleviation. This was performed using liquid carbon dioxide as the cryogen. In addition argon plasma coagulation was performed distally over the filiform projections. Post cryotherapy the patient's symptoms improved significantly and his choking episodes improved from daily to monthly. On repeat endoscopy the abnormal mucosal segment was reduced from 6 cm to 2 cm in length. Additional cryoablation was performed. Immediately post cryotherapy, mucosal sloughing of the abnormal mucosa was seen, which was peeled gently using endoscopy forceps. Histology showed hyperkeratosis (figure C), bulbous papillae (figure C) and keratin filled cysts (figure D). Carcinoma cuniculatum is a rare variant of well differentiated squamous cell carcinoma [1]. It was reported for the first time in the esophagus in 2005[2]and only 12 esophageal cases have been reported in the literature so far [1-3]. Dysphagia is the leading presenting symptom. Tumor invasion forms sinuses resembling rabbit burrows which can be seen on macroscopic examination. Diagnosis is exceedingly difficult by endoscopic biopsies, however is possible with EMR. There is no association with human papillomavirus infection. Both exophytic and endophytic growth pattern has been observed. It usually is deeply penetrating without lymph node metastasis [3]. Mitotic figures are rare and atypical mitosis is not seen and hence the indolent clinical course. Cytologic atypia however present is focal and mild and is not associated with an unfavorable outcome after curative resection as neither the deep mural invasion. Long term disease related survival is good with a median of 84 months, and cause of death is typically unrelated to the carcinoma cuniculatum [3]

1. Goh, G.H., et al., *Carcinoma cuniculatum of the esophagus and tongue: report of two cases, including TP53 mutational analysis.* Head Neck Pathol, 2014. **8**(3): p. 261-8.
2. De Petris, G., M. Lewin, and T. Shoji, *Carcinoma cuniculatum of the esophagus.* Ann Diagn Pathol, 2005. **9**(3): p. 134-8.
3. Landau, M., et al., *Esophageal carcinoma cuniculatum: report of 9 cases.* Am J Surg Pathol, 2012. **36**(1): p. 8-17.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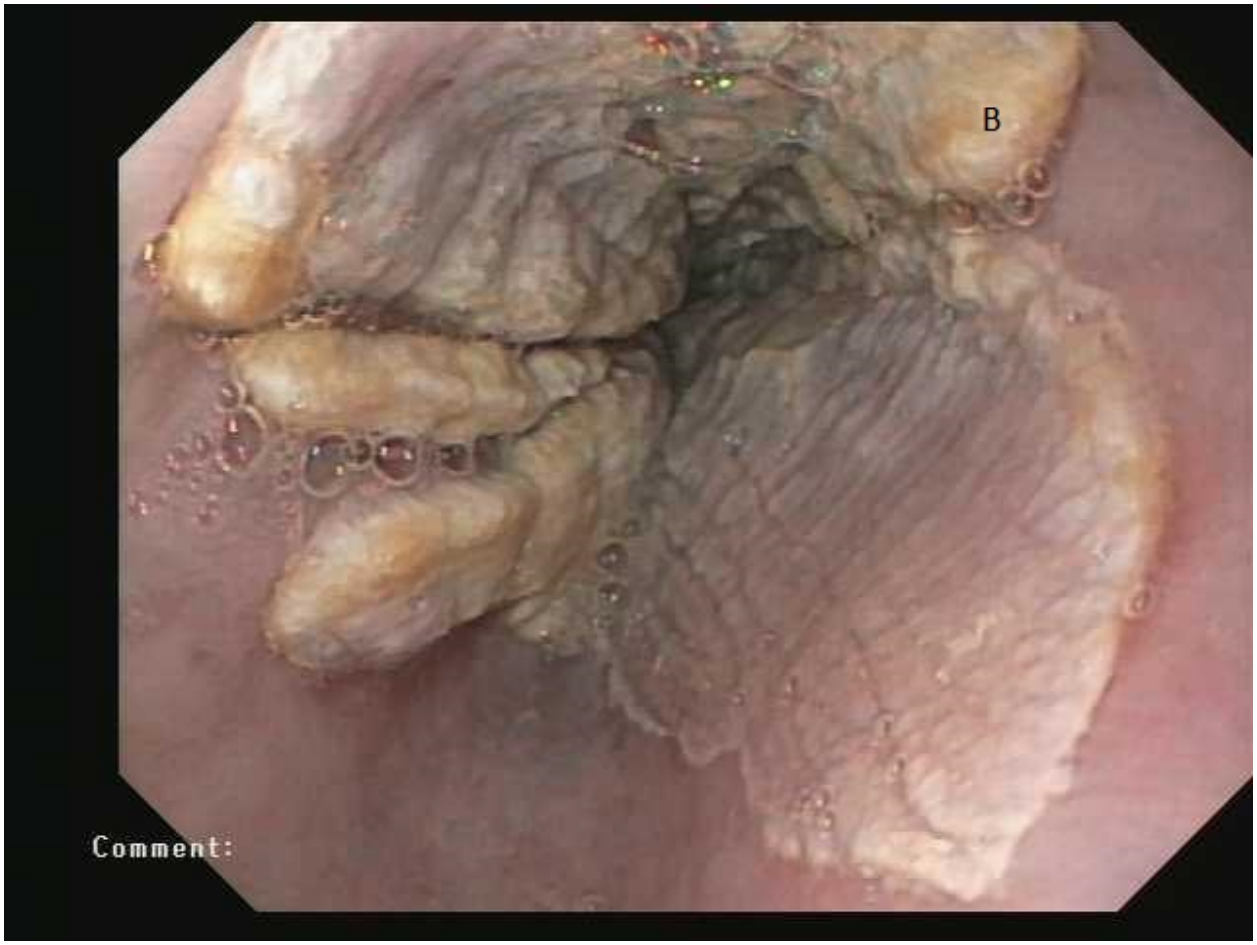
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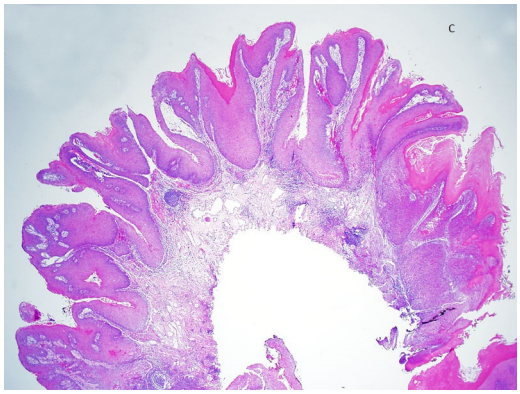
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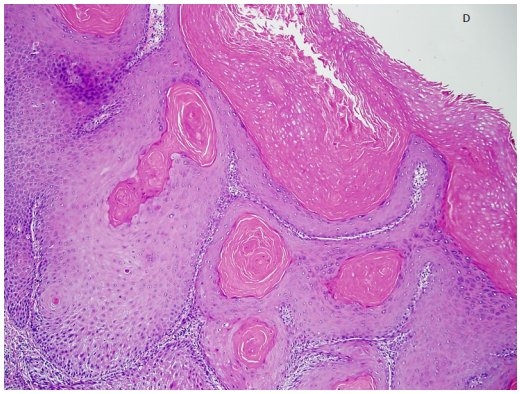
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