Challenges in Cricopharyngeal Dysfunction Complicated by multiple mechanisms : A Case Report

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Introduction

- Cricopharyngeal dysfunction (CPD) is characterized by the failure of the cricopharyngeal muscle to relax or close, leading to difficulty in swallowing and an increased risk of choking.
- We report a progressive case of dysphagia due to CPD, further complicated by Ossification of the Cervical Anterior Longitudinal Ligaments (OALL) and advanced esophageal cancer.

Patient presentation (72/M)									
Chief complaint	Difficulty swallowing both solid and liquid materials								
Onset	2-months before outpatient visit								
Associated symptom	Weight loss of 10kg due to reduced food intake, general weakness								
Past history	diagnosed with diabetes mellitus and hypertension 20 years before our outpatient visit								
	Manual muscle test (MMT): Grade 4+/4+ Range of motion (ROM): Passively full ROM								
Differential diagnosis	Upper gastrointestinal endoscopy : no specific findings Chest and abdomen CT scans : no specific findings								
	Whole spine CT scan : Ossification of Cervical Anterior Longitudinal Ligaments (OALL) from C3 to T1								
	Video Fluoroscopic Swallowing Study (VFSS) : Penetration-Aspiration Scale (PAS) Grade 5 for rice and liquid, and Grade 3 for all semisolid materials associated with mild CPD and severe osteophyte formation								
Surgical treatment	Removal of OALL from C3 to C7 was performed a month after the initial visit								

Postoperative outcome and management



Figure 1-1. (a) Lateral cervical spine radiograph and (b) sagittal cervical spine computed tomography (CT) showing anterior cervical osteophytes from C3 to T1. The anterior cervical osteophytes at the C3 and C4 levels are the most prominent (arrows) and (c) Axial CT slices through C4 show prominent osteophytes.



Figure 1-2. Lateral cervical spine radiographs obtained on preoperative state (d), post operative day 1 (e), and 3 (f). Prevertebral soft tissue swelling (bidirectional arrows) shows aggravation of severe prevertebral edema panel f compared with panels d and e.

 On the third postoperative day, VFSS showed worsening dysphagia, particularly CPD, and esophageal to pharyngeal reflux (PAS grade 5 for pudding, and not testable for other materials due to aspiration). Additionally, a cervical X-ray showed severe prevertebral edema at C3-7.
Electrostimulation and gradual balloon dilatation (3~5cc), three times a week were performed. On the 26th postoperative day, VFSS showed improvement (PAS grade G5 for cup drinking, G3 for 5cc fluid, G1 for other materials, but CPD persisted) and prevertebral edema, leading to the patient's discharee.

	PAS ^a)				Vallecular residue b)				Pyriformis sinus residue			
Study time	Pre	P3	P26	P106	Pre	P3	P26	P106	Pre	P3	P26	P106
Pudding	3	5	1	5	2	1	1	3	1	3	2	3
Rice porridge	3	NT ^{c)}	1	7	2	NT	1	3	1	NT	2	3
Curd-type yogurt	3	NT	1	NT	2	NT	1	NT	1	NT	2	NT
Steamed rice	5	NT	1	NT	2	NT	1	NT	1	NT	3	NT
Thin liquid (2cc)	5	NT	1	NT	1	NT	1	NT	1	NT	2	NT
Thin liquid (5cc)	5	NT	3	NT	2	NT	2	NT	1	NT	2	NT
Thin liquid (large amount)	5	NT	5	NT	2	NT	2	NT	1	NT	3	NT

Table 1. Summary of results of the video fluoroscopic swallowing study. (PAS, Penetration-Aspiration scale; Pre, Pre-operation, P3, postoperative day 3; P26, postoperative day 26, P106, postoperative day 106. NT, Not testable)

A month after discharge

 A month after discharge, the patient reported worsening dysphagia and resumed weight loss of 6kg and was admitted for gastrostomy and further evaluation. VFSS showed severe CPD (PAS grade G7 for semi-solid, G5 for pudding).



Figure 2. Esophagogastroduodenoscopy shows polypoid mass on upper esophagus(a).

Neck CT (b) and Positron Emission Tomography-Computed Tomography (PET-CT) (c) and (d) identified an 11cm avid mass in the esophagus and multiple lymph node involvement. Asterisk (*) indicates mass in the esophagus in Neck CT (a), PET-CT (c) and (d).

 A consultation with an otorhinolaryngologist led to myomectomy, which a polypoid mass was detected. A mass biopsy and debulking revealed squamous cell carcinoma. A Positron Emission Tomography-Computed Tomography (PET-CT) scan identified an 11cm avid mass in the esophagus and multiple lymph node involvement. The patient was transferred to the Department of Hematology and Oncology for Continuous Renal Replacement Therapy.

Conclusion

 This case underscores the challenges associated with dysphagia, primarily CPD, that are exacerbated by OALL, post-operative complications of OALL removal, and upper thoracic esophageal cancer. The progressive and severe dysphagia with weight loss should prompt serial evaluations, not only with VFSS but also with neck or chest CT and endoscopy.