REVIEW

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latrogenic pharyngo-esophageal diverticulum post-anterior cervical discectomy and fusion: A case report and review of literature

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Abstract

Objectives: The purpose of this study is to report a case of iatrogenic pharyngoesophageal diverticulum post-anterior cervical discectomy and fusion (ACDF) surgery, its management and management of postoperative complications. We also did a thorough review of literature about iatrogenic pharyngoesophageal diverticulum which is a rarely encountered complication occurring after a commonly performed surgery; ACDF.

Methods: Here we describe a case of iatrogenic pharyngoesophageal diverticulum post-ACDF surgery. In this paper we also make comparisons to the 23 cases reported in the literature in terms of: presentations, clinical findings, management courses, and complications. This study was approved by the Institution Review Board of Imam Abdulrahman Bin Faisal University, Dammam, Saudi Arabia. (Ref. no.: IRB-2023-01-473). The reported subject provided written informed consent before initiation of this study.

Results: Our case is a 45-year-old male with a history of ACDF surgery 8 years ago. He presented with dysphagia and regurgitations which started 1 year after ACDF. He was labeled as a case of Zenker's diverticulum and underwent multiple failed open and endoscopic surgeries prior to presenting to us. Upon presenting to our center, barium swallow showed the pharyngoesophageal diverticulum. Patient definitive diagnosis of iatrogenic rather than Zenker's diverticulum was established intraoperatively with esophagoscopy which revealed exposed hardware inside the diverticulum. He underwent open diverticulectomy and diverticulopexy. Postoperatively

No papers have been published, posted, or submitted from the same study.

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> he developed pharyngocutanous fistula and right vocal fold palsy, both successfully managed conservatively.

> Conclusion: latrogenic Pharyngoesophageal diverticulum is a rare complication following ACDF, however prolonged dysphagia shall warrant further investigation by contrast studies. Open diverticulectomy with muscle reinforcement is a good management modality. Due to the complicated anatomy secondary to previous operations, we recommend conservative management for patients with postoperative pharyngocutaneous fistula.

KEYWORDS

anterior cervical discectomy and fusion, pharyngocutanous fistula, pharyngoesophageal diverticulum, zenker diverticulum

1 INTRODUCTION

Anterior cervical discectomy and fusion (ACDF) first described in 1958 has become the most frequently performed operation for cervical disc herniations. ACDF surgery is associated with a very favorable outcome in most patients, with 95% of patients experiencing continuous improvement in their symptoms. Mild to moderate transient dysphagia is a commonly reported complication affecting 50% of cases and lasting 1 to 4 weeks postoperatively. A rare complication of ACDF presenting as dysphagia is the formation of an iatrogenic pharyngoesophageal diverticulum. To our knowledge, only 23 cases reported the same complication. In this study, we report a case of iatrogenic pharyngoesophageal diverticulum post-ACDF surgery, its management and management of postoperative complications. We also did a thorough review of literature.

2 **METHODS**

A case report of iatrogenic pharyngoesophageal diverticulum post-ACDF surgery along with its management, complications and management of complications.

Review of literature through a search across different search engines (Pubmed, Google scholar, IAU library) using the following terms: Zenker's diverticulum, pharyngeal diverticulum, traction diverticulum, iatrogenic pharyngeoesophageal diverticulum, pharyngeoesophageal diverticulum, post-ACDF pharyngeoesophageal diverticulum, and anterior cervical discectomy and fusion complications. We obtained 19 case reports and series which displayed a total of 23 cases that reported the development of pharyngeoesophageal diverticulum after ACDF surgery between the years 1991 and 2022.

CASE REPORT 3

A 45-year-old Saudi male patient presented with dysphagia, regurgitation and halitosis started after surgical history of ACDF done 9 years

ago. He developed dysphagia 1 year after the ACDF surgery and was assessed as "giant zenker's diverticulum" in another hospital. He underwent open diverticulectomy 1 year after the ACDF with no improvement. Patient underwent another open + endoscopic diverticulectomy and diverticulopexy 6 years later without improvement. Patient then came to our clinic with the same complaints. Upon examination in our hospital a soft palpable neck swelling was noted on the right side. A flexible nasophayrngolaryngoscopy revealed fullness of the hypopharynx. Contrasted neck computed tomography (CT) scan showed a retropharyngeal air-containing structure that extends from the level of the epiglottis down to the level of T1 vertebra extending to the right-side posterior to the right thyroid lobe (Figure 1). Subsequently, a barium swallow was performed revealing a pharyngoesophageal diverticulum at the cricoid cartilage level (C5/C6 disc space level), measuring about $6.5 \times 4.5 \times 1.5$ cm (Figure 2).

The patient was taken to the operating room, and endoscopic evaluation revealed an exposed cervical fusion plate and screws eroding from the pharyngeal wall and redundant mucosa (Figure 3). Two retraction pouches were also identified at the level of hypopharynx; small diverticulum from the left side and larger one in the right side that measured approximately 4.5 cm in depth just below the cricopharyngeus muscle. Open exploration from right cervical incision confirmed these findings. Significant adhesions between the diverticulum and surrounding tissue were evident and resection of the diverticulum was performed. Combination of Endostapler and manual suturing were used to ensure adequate pharyngeal repair. Then, the primary repair was reinforced by a sternocleidomastoid muscle rotation flap. During surgery, the neurosurgery team was involved and upon their assessment it was decided to keep the hardware in place and to cover it with an artificial anti-adhesive layer.

Postoperatively, the patient developed a pharyngocutaneous fistula and right vocal cord palsy. A contrast study demonstrated leakage at the pharyngoesophageal junction with fistula tract reaching the skin (Figure 4). Patient was managed conservatively by compressive dressings, IV antibiotics and gastrostomy tube feeding. Within 3 months complete resolution of fistula was ensured and oral diet resumed but there was a remaining non-healing sinus tract which was not

FIGURE 1 CT scan of the neck with contrast showing a retropharyngeal aircontaining structure (arrow) that extends from the level of the epiglottis down to the level of T1 vertebra.

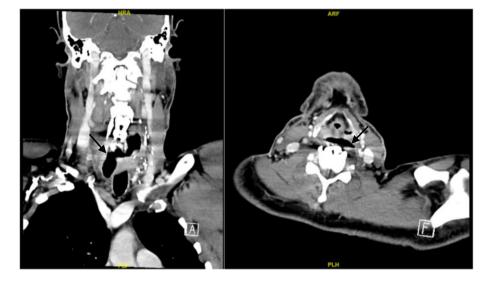






FIGURE 2 Preoperative barium swallow study revealing a pharyngeoesophageal diverticulum at the cricoid cartilage at the C5/C6 level, measuring about $6.5 \times 4.5 \times 1.5$ cm.



FIGURE 3 Intraoperative endoscopic evaluation revealing exposed cervical fusion plate and screws (arrow) eroding from the pharyngeal wall.





FIGURE 4 Postoperative barium swallow study showing leakage (arrow) at the pharyngoesophageal junction with fistula tract reaching the skin.

connected to pharynx or esophagus, and it required simple surgical excision (Figure 5). The right vocal cord palsy was completely resolved after around 5 months of follow up. At 2 years of follow up, the patient was having only very mild dysphagia and according to him he is around 90% better than pre-operative condition.

4 | DISCUSSION

ACDF surgery is one of the most commonly performed spinal procedures. The outcome of ACDF is usually satisfactory, however, complications occasionally arise.³ The most encountered postoperative complication is dysphagia with an incidence widely ranging between 2% and 67%.³ Risk factors of long-term dysphagia post-ACDF include female gender, revision surgery and multi-level surgeries.⁴ A prospective longitudinal study assessing the persistence of dysphagia post-ACDF found that the incidence of dysphagia diminished from 50% to 12.5% at 1 month and 12 months intervals.² Persistent dysphagia, however, should raise the suspicion of iatrogenic pharyngo-esophageal diverticulum which is a rare but possible complication.⁵ This complication carries an estimated incidence of 1.6%.⁶

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FIGURE 5 3 months post-op, showing complete resolution of fistula.

Through literature review, we obtained 19 case reports and series which displayed a total of 23 cases that reported the development of pharyngeoesophageal diverticulum after ACDF surgery between the years 1991 and 2022. Table 1 summarizes the reviewed cases in this paper.

Nine out of the 23 cases have reported the development of symptoms over 2 years after ACDF.⁵⁻¹³ However, in the literature, postoperative dysphagia ranged from immediate postoperative discovery to delayed discovery 20 years postoperatively.^{3,7} In our case, dysphagia developed 1 year after the surgery. Allis et al. in a case series postulated that the site of discectomy and fusion occurred most often at the level of C5-C7. Therefore, greater overall frequency of disc disease and surgery at those levels.¹⁴ Similarly, the site of discectomy and fusion was at the level of C5 and C6 in our patient.

Although this complication resembles Zenker's diverticulum, in fact, many of the reported cases have been mistakenly diagnosed as Zenker's diverticulum in their initial assessment, these two entities differ in their pathophysiology. Zenker's diverticulum is a herniation of the hypopharyngeal mucus membrane in the locus minor resistentiae between the muscle fibers in the upper esophageal sphincter region. A well-known theory explained its pathophysiology by the increased pressure in the pharynx during swallowing, secondary to a dysfunction of the cricopharyngeus muscle. In contract, a traction diverticulum results from pulling of the esophageal wall because of an inflammatory process in the surrounding area as reported in a case postcervical esophastomy. 15 In spinal surgery, the spine is exposed through a neck incision by displacing the SCM, jugular and carotid vessels laterally and the infrahyoid muscles and esophagus medially. Self-retaining retractor can damage the esophagus during exposure, and it may lead to weakness of the esophageal wall. The prolonged contact of the screw plates with the pharynx, causes microtrauma due to repeated friction leading to the formation of the diverticulum.6

To the best of our knowledge, only 23 cases of post-ACDF iatrogenic pharyngoesophageal diverticulum have been reported in the literature. The first case being reported in 1991 of a 45-year-old

man who presented 8 months post-ACDF with progressive dysphagia, regurgitation and weight loss. Successfully managed by excision, cricopharyngeal (CP) myotomy and fascia lata interposition. The reported cases ranged between 28 and 70 years with no significant difference between male and female gender (12 vs. 11), respectively, which is consistent with our patient being a 45-year-old male.

Physical examination findings varied, some cases showed no abnormalities at all, others revealed swelling in the neck and three cases exhibited pooling of saliva in the pyriform sinus. 5,15,16 In most reported cases, initial assessment was done using lateral radiographs, which revealed a ballooned esophagus full of gas. CT was a good tool to assess the positioning of cervical plates and the presence of abscesses. However, the clear contours of the pharyngoesophageal diverticulum were only revealed with the use of barium swallow. Thereafter, esophagoscopy was performed to confirm the site of the diverticulum, revealing exposed hardware visible inside the diverticulum. Volkow et al., reported a case that presented with elevated inflammatory markers, upon single-photon emission computed tomography (SPECT) (C4-C6) cervical spine spondylitis was suggested, later hydro soluble esophagogram revealed a posterior pharyngoesophageal diverticulum with a fistula to C6.9 In our case, we started with contrasted neck CT followed by barium swallow, however the provisional diagnosis was Zenker's diverticulum. Intraoperative endoscopy revealed hardware eroding into the posterior diverticulum similar to several of the reported cases in which endoscopy was performed pre or intra operatively.

There are a few considerations in the management of post-ACDF diverticula. First, endoscopic repair was largely unsuccessful in these patients. Though it was attempted in five cases, ^{13–15,17} open cervical approach was the end result of all of the reported cases except one case done by Salam et al. ¹⁸ Conversion from endoscopic to open repair was attributed to two factors; first, the inability to engage the stapler due the thick wall between the pharynx and the diverticulum. Second, the problematic positioning of the endoscope as the ACDF-related diverticulum is not located posteriorly, unlike a typical Zenker's diverticulum. ¹⁷

The second consideration is whether removal of hardware should be considered. In the literature, surgeons opted to remove hardware in most cases (60%) even in those with no dislodgement. Surprisingly, complication rates were higher in those whose hardware was removed (35%) in comparison to those whose hardware has not been removed (25%).

Even though the pathophysiology of the presented cases seems to be distinctive from that of Zenker's diverticulum, CP myotomy was performed in 65% of the reported cases. Allis et al. postulated that CP myotomy may decrease pulsion forces that could have contributed to the formation of the diverticulum. However, no significant dysphagia was reported as a complication in those who did not undergo CP myotomy.¹⁴

Closure of perforation may be obtained using stapler, manual suturing or a combination of the two. Esophageal repair dehiscence was noted more among cases that were repaired using manual suturing than cases repaired using stapler. Generally, diverticulectomy

pen, Hardware covered with artificial antiadhesive layer. osure with combination of endo- stapler and manual suturing ernocleidomastoid muscle flap attempted onverted to open ardware removed suppriority pedicled sternocleidomastoid muscle flap pen omyotomy done superiority addiced sternocleidomastoid muscle flap pen omyotomy done osure was done using interrupted sutures
Endoscopic repair attempted Converted to open Hardware removed CP myotomy done A superiorly pedicled sternocleidomastoid muscle flap Open CP myotomy done Closure was done using interrupted sutures
Open CP myotomy done Closure was done using interrupted sutures Onen
Hardware removed Closure using 4-0 vicryl Iliac bone graft with anterior plating
Open Hardware removed CP myotomy done

(Continues)

TABLE 1 (Continued)

o Z	No. Study	Patient demographics	Interval between ACDF surgery and presentation	Presenting symptoms	Site of ACDF	Findings on physical examination	Surgical Intervention	Postoperative complications	Follow up period and status at last follow up
_	Almre et al. ¹⁰	53-year-old Male	18 years	Progressive dysphagia	C6-C7	Did not reveal any abnormalities or signs of infection.	Open Hardware removed CP myotomy done The esophageal wall defect manually sutured using 3-0 PDS (polydioxanone) suture and the neck incision was closed in layers by a running suture.	Day 3 post-op: the patient developed high fever and esophageal suture line failure was suspected. Wound was opened, no leak was detected. The wound was irrigated and drained. Postoperative antibacterial treatment for 10 days.	At 1 year follow up patient had no complaints
∞	Solerio et al. ¹¹	41-year-old Male	7 years	Hyperthermia, right latero-cervical swelling, and a fistulous orifice at the site of the cervical surgical scar	C4-C6	ΣΖ	Open Hardware removed CP myotomy done Sternocleidomastoid muscle flap done	The patient had a salivary fistula. A small dehiscence of the proximal stapler line. After an unsuccessful treatment with fibrin glue, a right cervicotomy was performed.	A contrast study performed on the 10th postoperative day showed no evidence of fistula. The patient was eventually moved to a rehabilitation center.
6	Tian et al ¹²	31-year-old Male	7 years	Progressive dysphagia, odynophagia, dry cough, recurrent fever, weight loss, and bulging in the neck with swallowing	C4-C5	Neck bulge with swallowing	Open Hardware removed Esophagus closed with interrupted sutures Sternohyoid and omohyoid muscle flaps	None	2.5 years in good condition no complaints
10	Park et al. ¹³	54-years-old Female	3 years	Dysphagia and irritation in the neck	C5-C7	Σ	Open CP myotomy done Closure using Stapler with 4–0 vicryl suture reinforcement NPWT was used.	None	Σ
11	Allis et al. ¹⁴ Case 1	56-year-old Female	1 year	Regurgitation and chocking spells	C5-C6	ΣΖ	Open Hardware removed CP myotomy done A transverse cervical artery myofascial flap	None	At 32 months, asymptomatic

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Follow up period and status at last follow up	Asymptomatic at 13 months of follow- up	At 10 months follow up, occasional regurgitation and a stable pharyngeal diverticulum. No significant dysphagia and did not desire treatment at that time.	At 18 months follow up, he experiences occasional sticking of food.	Σ	Asymptomatic at last follow up	None
Postoperative complications	None		Right recurrent nerve palsy was noted.	ΣZ	Neck abscess requiring I&D and antibiotics, esophageal leak identified. Open repair revealed a 2 cm dehiscence at the inferior border of the esophageal repair.	Open CP myotomy Esophagus was closed in 2 layers with 3–0 Vicryl and 3–0 silk interrupted sutures.
Surgical Intervention	Endoscopic repair attempted Open CP myotomy done	Fistula was managed with intravenous antibiotics and tube feeding	Open CP myotomy done A piece of fascia lata was secured in front of the cervical spine to reconstruct prevertebral fascia	Open Hardware removed CP myotomy done	Endoscopic repair attempted Converted to open CP myotomy The defect was closed in 2 layers with running 3-0 Vicryl and interrupted 3-0 silk sutures.	Yes
Findings on physical examination	Σχ	Σ	Pooling of saliva in pyriform sinuses.	Salivary stasis in the pyriform sinuses.	Σ	Σ
Site of ACDF	C4-C5 and C5-C6	C5-C7	C6-C7	C6-C7 C6-C7	C5-C6	Σ
Presenting symptoms	Progressive dysphagia and regurgitation unresponsive to her reflux regimen	Left neck abscess	Dysphagia, regurgitation and weight loss.	Progressive dysphagia, sensation of blockage on swallowing, regurgitation and coughing after swallowing.	Odynophagia, dysphagia and regurgitation	Dysphagia, globus sensation and regurgitation
Interval between ACDF surgery and presentation	2 years	1 year	8 months	2 years	Several months later	6 months
Patient demographics	59-year-old Female	50-years-old Female	44-years-old Male	35-year-old Male	28-years-old Female	63-year-old Male
Study	Allis et al. ¹⁴ Case 2	Allis et al. ¹⁴ Case 3	Goffart et al. ¹⁵	Rebol Ferrer et al. ¹⁶	Alyssa et al.	Alyssa et al. ¹⁷ Case 2
ġ	12	13	4	15	16	17

(Continues)

TABLE 1 (Continued)

	Interval betw Patient ACDF surger demographics presentation	Interval between ACDF surgery and presentation	Presenting symptoms	Site of ACDF	Findings on physical examination	Surgical Intervention	Postoperative complications	Follow up period and status at last follow up
							Methylene blue was used to identify leak	
Salam et al. ¹⁸	36-years-old Female	2 years	Dysphagia	C5-C6	Tenderness over the right side of the neck	Endoscopic (Dohlman's endoscopic diathermy technique) Hardware removed	None	At 3 months, swallowing improved and satisfactory
	62-years-old Male	Σ	Incidental finding of esophageal perforation	C5/C6	None	Open Hardware removed CP myotomy done Closure done using staples Rotational sternocleidomastoid flap done	Σ	2 weeks, no complaints
im et al. Case 2 ¹⁹	Female Female	Σχ	Right sided neck pain and odynophagia	Σ	Σχ	Open Hardware removed CP myotomy done Closure done using staples Rotational sternocleidomastoid flap	None	ΣΖ
	Male Male	2 years	Dysp hagia	C5-C7	Σ	Endoscopic repair attempted Converted to open Hardware removed CP myotomy done A left forearm fascia free flap was placed to achieve a vascularized layer between the spine and esophageal closure	Mild infection treated with oral antibiotics and mild subcutaneous emphysema localized to the neck.	At 1 year Asymptomatic. Flexible endoscopy and esophagram at 18 months revealed an irregular but patent esophageal lumen
	43-years-old Female	2 years	Odynophagia, intermittent fevers, weight loss and mild neck pain	C4-C5, C5-C6 and C6-C7	Minimal pain with neck movement and hoarseness	Open Hardware removed Closure was done using resorpable single-layer sutures.	1 h after arrival to the recovery room, the patient deteriorated into pulseless electrical activity. Advanced cardiac life support protocol was initiated but	Patient deceased

o Z	No. Study	Interval betw Patient ACDF surgen demographics presentation	Interval between ACDF surgery and presentation	Presenting symptoms	Site of ACDF	Findings on physical examination	Surgical Intervention	Postoperative complications	Follow up period and status at last follow up
								resuscitation was not successful.	
23	Joanes et al. ²²	23 Joanes et al. ²² 31-years-old 2 years Male	2 years	Dysphagia, regurgitation, C5-T1 cough, and weight-loss.	C5-T1	ΣΖ	Open Hardware removed	None	3 years later patient was asymptomatic
24	24 Dobran et al. ²³	45-year-old 3 months Female	3 months	Cervical and brachial pain, fever and neurological worsening	C6-C7	Σχ	Open Hardware removed Esophagus sutured with a patch of sternohyoid muscle	Diverticulum still present Stable spastic tetraparesis Nutrition thro gastrostomy	Stable spastic tetraparesis Nutrition through gastrostomy

(Continued)

TABLE 1

Abbreviation: NM, not mentioned

should be followed by soft tissue reinforcement. Furthermore, a technique applied by Alyssa et al. is utilizing methylene blue to check for leaks intra-operatively.¹⁷

Muscle reinforcement was implemented in several of the reported cases. Four used rotational sternocleidomastoid flaps. ^{5,11,19} Other approaches included the use of sternohyoid muscle, forearm fascia free flap and fascia lata. However, in cases where obtaining muscle reinforcement is difficult Park et al. recommend the use of negative pressure wound therapy. ¹³

Postoperative complications are similar to those occurring with Zenker's diverticulum repair, including recurrent laryngeal nerve palsy with open surgery. Second, rupture of sutures may present as salivary fistula and can progress to life-threatening mediastinitis.⁶ Some authors have reported higher postoperative fistula rates in patients with pharyngoesophageal diverticulum post-ACDF than those with Zenker's diverticulum, due to poor esophageal healing following spinal surgery. 13 Close postoperative assessment is imperative to detect complications like mild infections, abscess formation, leakage and residual diverticulum. Simple infections can occur regardless of the presence of leak often presenting with fever and subcutaneous emphysema limited to the neck, and they can be successfully treated with oral antibiotics, irrigation and drainage. 10,16 However, leakassociated abscesses require incision and drainage in addition to revision surgery. ¹⁷ Some complications could be avoided intraoperatively. for instance Goffart et al. did not positively identify the recurrent laryngeal nerve and subsequently reported the occurrence of right recurrent larvngeal nerve palsy. 15 Additionally, some complications like leaks and residual diverticula can be detected with a postoperative barium swallow. Lastly, similarly to our case, Solerio et al. reported a case of postoperative salivary fistula in which fibrin glue was unsuccessful, thus cervicotomy and muscle flap were employed. 11 However, our patient was successfully managed conservatively with antibiotics, wound care and gastrostomy tube feeding to allow for complete resolution of the fistula.

Postoperative feeding in the immediate postoperative period is achieved by either nasogastric tube feeding or through gastrostomy. In complication-free patients gradual switching to oral feeding was done over a period of 5–10 days. Expectantly, switching to oral feeding required a longer time in complicated cases ranging between 2 and 6 weeks. ^{10,11,17,20,21} The follow up period ranged between a few weeks to 3 years, with the majority being asymptomatic at last follow up, while two cases reported minimal dysphagia or sticking of food that required no further intervention. ^{14,15}

5 | CONCLUSION

latrogenic pharyngo-esophageal diverticulum post-ACDF is a rare but challenging complication. Prolonged dysphagia following ACDF surgery should warrant further investigation by contrast studies especially barium swallow. Moreover, open diverticulectomy with muscle reinforcement at the suture site is a good modality for managing these patients. Postoperative pharyngocutanous fistula rates are high in

these patients, especially if it is revision surgery. Due to the complicated anatomy secondary to previous operations, we recommend conservative management for patients with postoperative pharynocutaneous fistulas.

5.1 | Strengths and limitations

To the best of our knowledge this is the most extensive review of iatrogenic pharyngoesophageal diverticulum post-ACDF done so far, making comparisons and summarizations of a total of 23 cases. 5-23 This study thoroughly displays the management course of a patient presenting with this iatrogenic complication, from clinical presentation, diagnosis, intra-operative considerations to complications and postoperative course. Limitations of our study include missing data in some of the included cases, especially older ones. Additionally, generally case reports carry a limited possible generalization and difficulty in establishment of cause-effect relations.

CONFLICT OF INTEREST STATEMENT

Authors declare no conflicts of interests that could have influenced the work reported in this paper.

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