Pharyngocutaneous fistula after anterior cervical spine surgery

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Accepted: 12 March 2009 / Published online: 28 March 2009
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Abstract

Pharyngocutaneous fistulae are rare complications of anterior spine surgery occurring in less than 0.1% of all anterior surgery cases. We report a case of a 19 year old female who sustained a C6 burst fracture with complete quadriplegia. She was treated urgently with a C6 corpectomy with anterior cage and plating followed by posterior cervical stabilization at another institution. Post operatively she developed a pharyngocutaneous fistula that failed to heal despite several attempts of closure and

esophageal exclusion with a Jpeg tube. The patient was eventually successfully treated with a three-stage procedure consisting of firstly a posterior approach to reinforce the posterior stabilization of the cervical spine that was felt to be inadequate, secondly an anterior approach with removal of all the anterior instrumentation followed by iliac crest bone graft and thirdly a superior based sternocleidomastoid flap that was interposed between the esophagus and the anterior cervical spine. The patient's fistula healed successfully. However, yet asymptomatic, the anterior iliac crest bone graft resorbed almost completely at 16 months follow up. In light of this complication, we discuss the surgical options for the treatment of pharyngocutaneous fistulae and the closure of this fistula using a superiorly based sternocleidomastoid muscle flap.

Keywords Cervical spine fracture ·
Pharyngoesophageal fistula · Esophageal injuries ·
Anterior cervical spine surgery · Sternocleidomastoid flaps

Grand rounds case presentation

The patient is a 19-year-old female who was an unrestrained passenger in a motor vehicle accident in January 2007. The patient was ejected from the vehicle and suffered a C6 burst fracture with significant canal compromise and spinal cord injury, rendering her as a complete C5 quadriplegic (Fig. 1a). The patient had a C6 corpectomy anterior column reconstruction with a mesh cage and cervical plate followed by a posterior instrumented fusion from C4 to C7 at an outside facility (Fig. 1b). While at the rehabilitation center, she was found to have persistent drainage from her neck (Fig. 2a). A pharyngocutaneous fistula was suspected and she was transferred to our

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Fig. 1 **a** Note severe burst fracture of C6, with evidence of spinal cord injury prior to surgery at outside facility. **b** Construct from outside facility after anterior corpectomy and posterior fixation

Fig. 2 **a** Note opening of pharyngocutaneous fistula. **b** Axial CT of spine at level of fistula. Note the presence of air deep to the skin surface as well as adjacent to the hardware (white arrows)

institution where a CT scan showed evidence of air in front of the instrumentation (Fig. 2b). A barium swallow study confirmed the esophageal injury. Pharyngoesophagoscopy demonstrated a posterior esophageal perforation (Fig. 3) measuring 1.5×2 cm long. Two attempts were made to close the defect primarily, and these were not successful despite the placement of a Jpeg tube to exclude the esophagus. This was followed by the use of an omohyoid flap to close the pharyngeal defect while the instrumentation was retained. This too was not successful.

Diagnostic imaging section

Figures 1–3.

Literature

Pharyngocutaneous fistulae are rarely found in patients who have undergone anterior cervical spine surgery. The

Fig. 3 Note the cervical plate seen from the laryngoscope, hence demonstrating defect in pharyngeal wall

incidence of pharyngeal or esophageal injuries has been reported to be 0.1% after anterior cervical spine surgery [6]. While esophageal injuries may not be extremely rare, the development of an esophageal injury into an esophagocutaneous fistula has been reported fewer than ten times [2, 3, 8, 9]. Pharyngocutaneous or esophagocutaneous fistulae are always associated with injuries to the esophagus. The resultant salivary leakage into surrounding soft tissue, ultimately leads to communication of the salivary tract with the skin and hence a fistula. There are various etiologies for these types of fistulae. Perforations may occur as direct trauma to the esophagus during dissection, decompression of the spine with a burr or fixation of the anterior cervical spine. This injury may not be recognized during the original surgery, making the diagnosis delayed, hence allowing for the development of a fistula. Perforation of the esophagus may also occur as a result of protruding screws from a cervical plate. The management of fistulae in this setting can be challenging since one must balance several factors to allow the patient to achieve the fastest and safest recovery. In particular, the surgeon must aim to create a durable closure of the perforation, control or prevent infection, and maintain the stability of the spine. Since a pharyngocutaneous fistula is the most frequent complication after total laryngectomy [1], head and neck surgeons have developed several techniques to facilitate their repair. It is therefore prudent to involve the expertise of the otolaryngology service to optimize patient results. The use of different flaps such as omental flaps or muscle flaps to repair these fistulae has been recognized as effective treatment in patient who have had anterior spine surgery [5, 7]. We report our case of a pharyngocutaneous fistula and confirm the effectiveness of its treatment through a sternocleidomastoid muscle flap.

Rationale for treatment

As the patient had failed two previous attempts of direct repair and a previous omohyoid flap it was thought that more extensive surgery had to be performed. The patient

was therefore presented at a conference between the spine surgery and otolaryngology teams.

Several issues had to be assessed: the stability of the overall construct, the foreign body environment constituted by the anterior plate and cage, the need for coverage and interposition of tissue material between the cervical spine and the esophagus, and the need to reconstruct the anterior column defect after cage removal. Furthermore the overall nutritional status of the patient had to be considered. The stability was assessed carefully, and a CT scan with reconstruction could already show some radiolucency around the C7 lateral mass screws. Furthermore, lateral mass screws in C7 are not biomechanically very strong [4]. The need to remove the anterior plating system and the cage would increase the instability of the cervical spine. It was therefore decided that the patient would require a revision of her posterior instrumentation. To achieve healing of the fistula it was thought that we would need to do further debridement, eliminate all foreign metallic bodies in the front, have nothing protruding in the front of the spine behind the esophagus, and have a thick interposition material between the spine and the esophagus that will be repaired and patched with a muscle flap.

Procedure

To achieve all these goals the patient had a three-stage procedure during the same anesthesia. First, the patient was placed prone and had a distal extension of her posterior cervical instrumentation down to T3. During the posterior surgery the C6 and C7 lateral mass screws were indeed found to be loose. However, the proximal part of the instrumentation was found to be solid. It was therefore decided to extend the instrumentation with at least four points of fixation and to crosslink the previous rods with dominoes. The posterior instrumentation was therefore extended to T3 with side to side dominoes. The wound was then closed.

Second after the patient had been flipped supine a tricortical iliac crest bone graft of adequate length was

Fig. 4 **a** Hemostat is inserted into the esophageal wall defect. **b** After removal of the plate and cage, iliac crest bone was inserted in the corpectomy defect (*black arrow*)

harvested from the middle of the iliac crest in view of the reconstruction of the anterior column.

Third, the pharyngocutaneous fistula site was exposed and excised with the neck incision. Subplatysmal dissection was performed and fibrotic tissue was debrided. The sternocleidomastoid muscle was mobilized off the sternal and clavicular heads. Further dissection was carried out to expose the cervical instrumentation and the pharyngeal defect (Fig. 4a). The anterior plate and cage were removed

and the wound was debrided and irrigated. The iliac bone graft was sized to fit into the corpectomy site (Fig. 4b). At this point the sternocleidomastoid muscle flap was secured to the pharyngeal perforation. Fibrin tissue adhesive was placed between the perforation and the muscle flap. The wound was then closed in appropriate fashion. The Jpeg tube was left in place for another 8 weeks to maintain adequate esophageal exclusion and nutritional balance.

Procedure imaging section

Figures 4, 5, 6.

Outcome

After surgery, her wound was monitored with no evidence of salivary leakage. She did, however, develop a temperature, and her sputum was found to have methicillin resistant *Staphylococcus aureus*. The infectious disease service, suggested that the patient should continue on vancomycin and metronidazole for a period of 3 weeks following surgery. On postoperative day seven, there was no evidence of salivary leak and she was sent to the rehabilitation facility. A barium swallow study performed 2 weeks afterwards did not show evidence of any fistula and the patient was allowed to resume her diet while the Jpeg tube was removed. She was then evaluated 2 months after surgery in otolaryngology

Fig. 5 Note the mobilized sternocleidomastoid muscle and associated flap that has been attached to the pharyngeal defect (white arrow)

Fig. 6 Two months postoperative X-rays seem to show incorporation of the iliac crest bone graft

Fig. 7 Note resorption of iliac crest bone graft at 16 months follow up (*white arrow*)

clinic where a laryngoscope was used to assess the status of the fistula. It was completely closed at that time, and the patient was eating a swallowing solid food without difficulty. Her neck wound had healed well. Sixteen months after surgery, she continues to do well, eating without any difficulties and without any evidence of fistula recurrence. She has, however, still a tracheostomy due to her C5 quadriplegia. Plain films reveal a stable instrumented construct; however, the iliac crest bone graft showed evidence of an almost complete resorption. No further surgery is done as the patient is totally asymptomatic without any symptoms or fever (see Fig. 7).

Conclusion

This case demonstrates the validity of a solid stabilization of the cervical spine in the setting of infection. This case also demonstrates the use of a thick muscle flap for esophageal fistula repair after cervical spine surgery. However, the anterior bone resorption raises some controversy regarding the surgical technique of anterior debridement with grafting of the anterior column defect. Also the bone resorption raises the question of possible persistent low-grade infection.

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Original surgery and treatment

When considering options for treatment in subaxial spine injuries there are still varied opinions as to definitive treatment. One must take into account several factors: the morphology of the injury; the discoligamentous complex; the neurologic exam of the patient. When describing burst fractures in the cervical spine they can either be compression burst fractures with no ligamentous disruption or burst fracture dislocations having a significant posterior element injury which may include the posterior ligament, facet capsule or facet and lateral mass fractures. Significant retropulsion of the posterior vertebral body demonstrates a high degree of discoligamentous disruption associated with this injury and is often the source of residual spinal cord compression. This patient has a complete spinal cord injury at C6 with a C5 neurologic level. The only original imaging we have is a lateral cervical MRI, which shows considerable retropulsion. Unfortunately with the single sagittal T2 image we do not see any posterior ligamentous injury, or lateral mass fracture. The assumption of a posterior complex injury was made here due to the degree of retropulsion. The indication for surgical decompression

then is given to facilitate neurological recovery within the zone of partial preservation (root recovery) [2]. In a patient with a C5 level lesion, the possible recovery of C6 or C7 can often make a patient much more independent thus enabling him to partially use the upper extremity. Modern day treatment would include anterior cervical C6 corpectomy, vertebral replacement, grafting and an anterior plate either locked or dynamic. In simple burst fractures without any evidence of posterior injury this would be sufficient. In unstable burst fractures many surgeons would supplement their treatment with posterior fixation within the zone of injury, which is usually one level above to one below, i.e. C5–C7. The choice of vertebral replacement can be autograft iliac crest, allograft strut or a cage filled with the bone from the corpectomy. Although a second graft harvest incision is sometimes painful, in a paraplegic patient this is most unlikely. A possible additional infection site may nevertheless be the reason to advocate avoidance of homologous iliac graft. It is also important to remember that burst fractures by nature tend to heal with immobilization only and in the past or in countries without modern aggressive treatment this fracture would likely have consolidated with a kyphotic deformity, whether with early traction, Halo or a brace.

The complication of esophageal perforation is well discussed in the article as being related to anterior spine surgery with plating. One other risk factor that has not been mentioned is the possibility of ischemia secondary to retraction of the surrounding tissue caused by retractors which I would also like to mention [1, 9]. Secondly, there are more than 60 articles in Pub Med in the last 25 years that relate esophageal perforation to anterior cervical spine surgery all during the time of modern cervical spine surgery with anterior plating. Prior to that several articles have commented on the incidence of esophageal or pharyngeal injury as related to cervical spine fractures themselves or the mechanism that caused the original injury. It is often easy to forget that a patient with a severe cervical injury may already have a tear, stretch or other soft tissue injuries to the structures of the neck and that subsequent trauma of surgery aggravates the situation, or the injury is originally missed in an intubated patient [7, 8, 4].

Approach to complication and subsequent treatment

The approach to the resultant complication of esophageal perforation after anterior cervical spine surgery with plating is well described in this article with (1) diagnosing the injury (2) diversion of alimentation, a J-Tube, (3) removing the instrumentation (4) healing the perforation (5) eliminating infection (6) and healing the cervical spine.

There were two failed attempts at primary closure of the defect with J-Tube diversion but retention of the

instrumentation and a third attempt with an omohyoid flap with retention of the plate. These failed and may have benefitted from removal of the anterior plate especially if it was the offending problem or if the problem was aggravated by ongoing infection.

In the latest surgical procedure, the authors collaborated with the ENT service who are most experienced in this area. They also established preoperatively that the posterior instrumentation was loose on CT scan, found the inferior portion of the construct to be loose at exploration, and restabilized the posterior spine down to T3. The lateral mass screws in C7 had become loose due to the small lateral mass and pedicle screws were preferable here. The second stage of the procedure was anterior where the anterior instrumentation was removed and the cage, irrigated and debrided the area and replaced it with autograft bone.

There is no mention of taking bone samples at the time of definitive surgery for microbiology. This maybe would have helped to reveal an osteomyelitis which normally is treated with 6 weeks of intravenous antibiotics. In this case it could have been Methacillin Resistant *S. aureus* and the antibiotic Vancomycin would have been employed for six consecutive weeks.

For esophageal perforations, the literature describes several methods in repairing the perforation, from monitoring small perforations with esophageal diversion, primarily suturing the defect, closing the defect with the use of a flap of local muscle (omohyoid or sternocleidomastoid muscle), omentum or esophageal diversion with jejunum [3, 5, 6]. Here the patient had a series of attempts to treat the problem which started with esophageal diversion and primary repair and ended with esophageal diversion, hardware removal, sternocleidomastoid flap and iliac crest bone grafting. This final treatment encompassed all the principles mentioned above with eventual resolution of the problem.

The endpoint was a healed fistula and a pain free patient with anterior graft resorption possibly from osteomyelitis. I agree with now waiting and following the patient along to see what happens. She is likely fused posteriorly with this long posterior construct. I would consider following the patient with CRP or sed rates and have a low threshold for

redebriement if the patient becomes symptomatic or falls into kyphosis.

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