

Case Report

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Pharyngo Cutaneous Fistula Following Polytrauma

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Abstract

5-15% of patients with undiagnosed Fever of Unknown Origin have a chronic course, especially in long term care settings. It is important to identify the underlying cause as it may be secondary to more sinister underlying causes that often require an intensive and advanced diagnostic evaluation. Acute non-tuberculous retropharyngeal abscess is an infection in one of the deep spaces of the neck; it is rare in adults and usually results from local trauma, such as foreign body ingestion, or following instrumental procedures (laryngoscopy, endotracheal intubation, feeding tube placement, etc.). An esophageal perforation after anterior cervical surgery is also an uncommon but well recognized complication. Esophageal perforation and neck abscess formation are a rare complication of anterior cervical spine surgery. The causes include: (a) Oppression of the esophagus during surgery, due to a clasp held for too long causing a necrosis of the esophagus; (b) Esophagus injured by loose plates and screws.

Therefore, after anterior cervical spine surgery if patients have a high fever, sore throat, swelling incision, and food sediment is was found in the incision, esophageal cutaneous fistula should be considered. The final diagnosis could be done by esophageal radiography. CT scan helps in delineating the location and condition of the implant, extent of an underlying abscess and possible extension of the abscess along the prevertebral space.

We report a 31-year-old, Indian male patient who developed non-tuberculous retropharyngeal abscess during his hospital stay. The retropharyngeal abscess occurred in the context of unknown blunt trauma to esophagus and led to bilateral lower lung collapse and consolidation with secondary bronchiectasis that ultimately proved fatal. This case report describes an unusual but important cause for fever of unknown origin especially in cases where patients have Polymicrobial infections with rare offending agents particularly in patients in acute medical setting.

Keywords: Retropharyngeal abscess; Esophageo cutaneous fistula; Long term care settings; Adult

Abbreviations: FUO: Fever of Unknown Origin; VAP: Ventilator Associated Pneumonia; SpO₂: Oxygen Saturation; TLC: Total leukocyte Count; ESR: Erythrocyte Sedimentation Rate; BUN: Blood Urea Nitrogen; TB-PCR: Tuberculosis Polymerase Chain Reaction Test; CT Neck: Computed Tomography; D2 level: 2nd Post Cervical Spinal Segment; C4 vertebra: 4th Cervical Vertebra; TPN: Total Parenteral Nutrition; Activated PTT: Activated Partial Thromboplastin Time; PT INR: International Normalized Ratio for Prothrombin Time; AD: Anno Domini; C5- C6 level: 5th and 6th Cervical Level

Case Report

Background

Prolonged unexplained fevers have perplexed clinicians from antiquity and remain a challenging exercise in differential diagnosis. The diagnostic workup should be directed by features of the clinical presentations, which almost always suggest an infectious, rheumatic/inflammatory, neoplastic, or miscellaneous disorder.

Retropharyngeal abscess are rare in adults and constitute a serious emergency. Early diagnosis and treatment is very important save the patient's life. Most undiagnosed FUOs are due to a failure to consider a diagnosis or a comprehensive but misguided workup [1].

Two similar cases of delayed prevertebral infection following anterior surgery for traumatic cervical fractures were reported by Kuriloff et al. [2]. Both presented neck swelling and tenderness, associated with dysphagia, fever and clear laboratory evidence of acute inflammation, 2 and 4 months after surgery, respectively. As a pharyngocutaneous fistula was also found in both cases, their treatment consisted of surgical reconstruction of the oesophagus and antibiotics. Another case of delayed neck abscess formation (10 weeks after spinal fusion) was reported by Whitehill [3]. This patient presented fever, neck swelling and the classical laboratory signs of inflammation.

We report an unusual complication of non-tuberculous retropharyngeal abscess that followed an unknown blunt trauma to the esophagus in a patient with quadriplegia.

Case presentation

Mr. X, a 31 year-old Indian male was admitted to high dependency unit of our multi-specialty hospital for long term care; he had Quadriplegia which resulted from neck trauma that he sustained after diving in to the sea. He underwent C5 cervical spine fixation (Figure 1) and was put on tracheostomy with continuous pressure ventilator support for one month, which was then weaned off. He had history of ventilator associated *klebsiella pneumonia* and *pseudomonas aeruginosa pneumoniae* (VAP) and received antibiotics for 14 days as per the culture and sensitivity report. He became afebrile for a short period of time and developed intermittent fever (38°C) after one week full course of antibiotics for VAP and this persisted for 2 months. Along with fever, he also reported headache, myalgia and generalized aches and pains. He had significant weight loss (6 kg) during the three month period of admission. There was however no changes in his appetite, or bowel and urinary function.

Prior to the accident, Mr. X was a healthy young male and he did not have any other medical problems; in particular there was no history of contact with tuberculosis, animal or insect bite, or any history of

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Figure 1: (Surgical intervention with corpectomy and replacement of body of the C5 vertebra & anterior fixation of C4, C5 and C6 vertebra).

travelling outside of Qatar in the past one year. He was not on any traditional medication. Physical examination revealed an average built young adult male; his vital statistics were unremarkable with a heart rate of 90 beats per minute, supine blood pressure of 120/80 mm Hg, respiratory rate of 18 per minute, regular breathing and an oxygen saturation (SpO₂) of 99% on one liter/minute on oxygen therapy). He was febrile and had temperature between 38°C-39°C on several occasions. He was conscious, and oriented and had grade 2/5 power in his upper limbs and grade 0/5 power in his lower limbs. He had generalized sensory loss below the neck. Examination of his respiratory system revealed reduced chest expansion; on percussion, he had dull note bilaterally in the basal area.

On investigations, he was found to have anaemia (haemoglobin 9.5g/dl) and evidence of infection with a Total Leukocyte Count (TLC) of 25,900/mm³ and neutrophilia of 93% his platelet count was 498000/ul. He had a raised Erythrocyte Sedimentation Rate (ESR) (137 mm in one hour) and C reactive protein (194 mg/L) indicating inflammation. His renal functions were within accepted limits (Blood Urea Nitrogen (BUN) of 5.9 mmol/L, serum Creatinine of 41 mmol/L, serum albumin of 37 g/L and normal electrolytes. Blood cultures were negative, urine cultures positive for *enterococcus faecalis*; Sputum culture from tracheostomy aspirate showed *klebsiella pneumonia* and *pseudomonas aeruginosa pneumonia*.

Serum electrophoresis showed increased gamma globulin chains with no M band suggesting chronic inflammatory pathology. While his Mantoux skin test was positive, quantiferon test was indefinite. So sputum for polymerase chain reaction for the detection of *Mycobacterium tuberculosis* TB-PCR done and this was negative; an infectious disease consultation concurred with the opinion that the patient did not have active tuberculosis.

Plain radiograph and contrast computerized tomography of chest revealed bilateral lower lobe consolidation with patchy ground glass area of air space infiltrate in the rest of right lung and layering right pleural effusion (Figures 2 and 3). Electrocardiogram and Echo cardiogram were normal.

After 90 days of intermittent fever he developed multiple diffuse swellings with poorly defined margins on the right side of his neck with progressive dysphagia. The swellings were non tender and fluctuant. There was no evidence of tracheal shift. He reported severe pain behind his chest. On evaluation, Mr. X had neck stiffness with spasm of spinal muscles. Laryngological examination with direct laryngoscopy was

normal; the Head and Neck Specialist therefore suggested CT neck with contrast to rule out swelling in the retropharyngeal area. This imaging (Figure 4) showed a massive retropharyngeal abscess with air fluid level extending to the base of the skull above and its lower limit extending up to D2 level.

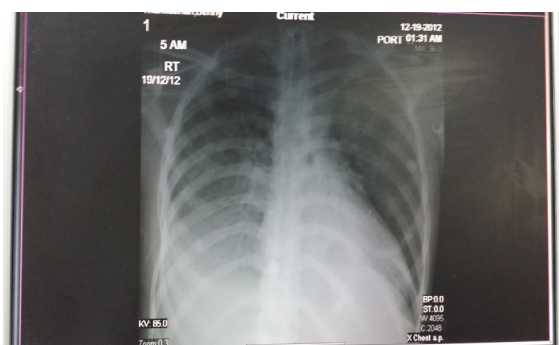


Figure 2: (Chest roentgenogram: Bilateral lung infiltrates with right side pleural effusion).

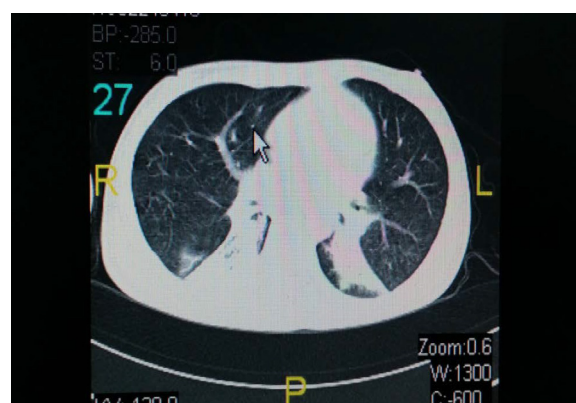


Figure 3: (Contrast computerized tomography of chest: Bilateral lower lobe consolidation with patchy ground glass area of air space infiltrate in the rest right lung).

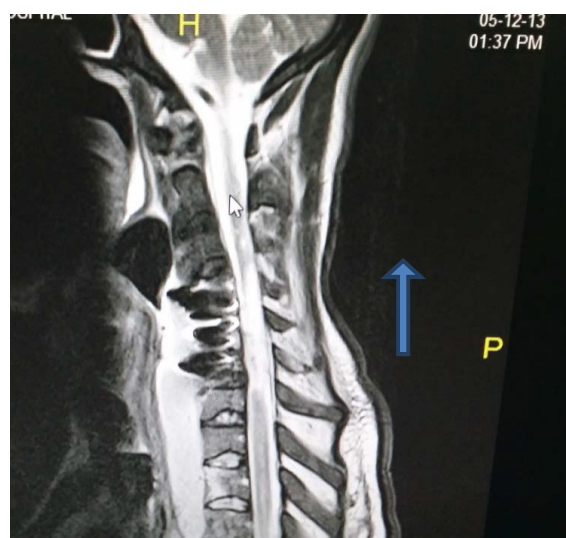


Figure 4: CT (sagittal view) showing retropharyngeal collection.

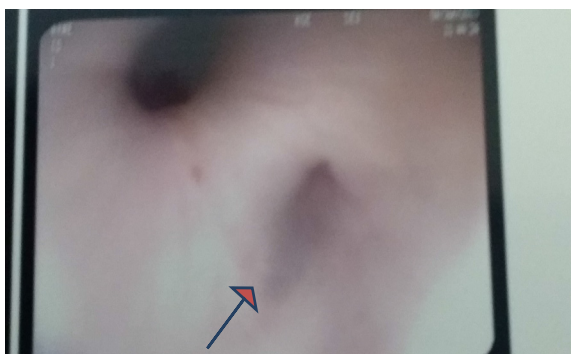


Figure 5: (Esophageal fistulous tract in upper gastro endoscopy).

He underwent upper gastro endoscopy (Figure 5) done by gastroenterologist to look for cause of retropharyngeal abscess. It revealed multiple deep ulcers were seen in esophagus (at least 3), a large fistulous tract that was 15 cms long and made a cutaneous fistula and caused paravertebral abscess with deep cervical extension; it contained food materials/pus coming out, ahead). The stomach and duodenum were normal. The definite management of cervical abscess is immediate drainage; hence the retropharyngeal collection was drained under CT guidance with 5 French Pig tail catheter inserted and deployed at the level of the C4 vertebra (Figure 4). 750 cc of foul smelling, thick pus was drained for 3 weeks and patient was put on total parenteral nutrition (TPN) during these days. There was a gush of gas before the pus came out; it can be due to anaerobic gas forming organism.

A Ryle's tube was inserted later to prevent contamination by food. The culture and sensitivity of pus revealed three organisms: *Klebsiella pneumoniae* with extended spectrum beta lactamase organism, carbapenam (resistant *Pseudomonas aeruginosa* sensitive to Piperacillin Tazobactam, Trimethoprim plus Sulphamethaxole) and *Sacharomyces cervicea* (sensitive to Fluconazole, Caspofungin). Patient was started on intravenous Piperacillin Tazobactam 4.5 gram 8 hourly, Fluconazole 200 mg once daily respectively. Mr. X showed a significant improvement and remained asymptomatic for a period of one month.

However after one month total parenteral nutrition through central venous catheter, he developed right jugular vein thrombosis with septic shock. Repeat complete haemogram showed haemoglobin 8.6 g/dl, TLC 30000/mm³ and neutrophilia of 75%, platelet count 410000/ul, BUN: 10.2 mm/L, serum creatinine: 123 mm/L, serum sodium 134 mm/L, serum potassium: 5.2 mm/L; activated PTT: 50 seconds, PT INR:1.1 seconds;

Despite the fact that Mr. X was receiving higher antibiotics as per the culture report, repeated culture revealed the same organisms now resistant to antibiotics. The patient's condition further deteriorated probably due to Polymicrobial sepsis syndrome and complication of TPN; he died after 9 months of admission to high dependency long term care unit.

Discussion

A fascial envelope surrounds the structures of neck. Several potential spaces are present in the fascial envelope and retropharyngeal space is one of them. These spaces are continuous with spine, paravertebral space and mediastinum. Hence the infections in neck may spread up to the paravertebral space and mediastinum [4]. Deep neck infections have been recognized and described since the time of Galen in the second century AD [5]. These infections were frequently encountered in the pre-antibiotic era. However since the advent of

antibiotics, these have become quite rare, more so in adults and can cause considerable morbidity [5]. In adults, an acute nontuberculous retropharyngeal abscess mostly develops as a result of trauma to the pharynx and the oesophagus, either by a foreign body or endoscopy [6-9]. However, it may rarely develop following dental infections [7-10] or pyogenic osteomyelitis of cervical spine [11]. A recent study [12] holds upper respiratory tract infections as the most common etiological predisposing factor responsible for retropharyngeal abscess in adults also, presumably due to spread of infection to a persistent retropharyngeal node as in children.

In our case, although there was no evidence of any trauma to the cervical area or penetrating injury on history and clinical examination, it is possible that blunt trauma during the accident or tracheostomy may have caused a break in the mucosal lining with subsequent seeding of infection in the retropharyngeal space.

The usual clinical features of acute retropharyngeal abscess include sore throat, dysphagia, fever and midline pharyngeal swelling. In more severe cases external neck swelling or neck rigidity may be present. Sometimes hoarseness, stridor and respiratory obstruction may also develop either due to anterior displacement of posterior pharyngeal wall by the abscess or secondary laryngeal oedema [13].

In our case the symptoms appeared two months after the prolonged fever, it can be due to severe sensory loss below the neck secondary to trauma causing spinal cord compression at C5- C6 level. Microbiology of non-tuberculosis retropharyngeal abscesses often reveals mixed isolates involving both aerobic and anaerobic bacteria [14,15]. Predominant causative aerobes are *Streptococci*, *Staphylococcus aureus* and *Klebsiella*, while predominant anaerobes are *Bacteroides* and *Peptostreptococcus* species [9,15]. In the present case *Klebsiella*, *pseudomonas* and *Saccharomyces cerviceae* were the causative organisms.

In another similar case reported as a young male who underwent anterior cervical fusion for a compressed fracture of the C5 vertebra developed postoperatively partial extrusion of the bone graft, followed by progressive dysphagia and retropharyngeal emphysema. Although no definite perforation of the oesophagus or pharynx was detected at reoperation, an extensive pharyngocutaneous fistula formed subsequently through the operative wound. Open drainage in association with broad spectrum antibiotics, continuous nasopharyngeal suctioning, stopping of oral intake and gastrostomy feeding resulted in closure of the fistula [16]. Radiograph of the neck in lateral position during deep inspiration with neck fully extended is the most valuable tool in the diagnosis of retropharyngeal abscess. Classical radiological changes suggesting pathology in the retropharyngeal space include: increased thickness of the paravertebral soft tissues, air or air fluid level in the soft tissue and loss or reversal of the cervical spine curvature [10]. CT scan is very useful for the early diagnosis of neck abscess and for the follow-up [17].

Retropharyngeal space communicates inferiorly with the mediastinum making spread of infection possible. Due to the enzymatic action of organisms and aspiration fluid content through tracheostomy stoma, lung parenchyma may get invaded secondarily leading to the development of bilateral consolidation with underlying collapse, like in the present case [18,19-22].

Ideal treatment of acute non-tuberculous retropharyngeal abscess would include immediate drainage by transoral or trans-cervical approach along with appropriate systemic antibiotics depending on the culture and sensitivity results [13,20,21].

Conclusions

Perforation of the pharynx or oesophagus following blunt trauma to neck is a rare complication. This case illustrates the importance of having a systematic approach to the assessment and management of prolonged fever with poly microbial infections in a non-immuno-compromised individual. The pattern of fever, repeated clinical assessment and use of diagnostic investigation and multidisciplinary care team work are helpful in making a strong diagnosis. Delay in diagnosis may occur because of lack of awareness of this complication and is associated with a high mortality.

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References

1. Rueth N, Shaw D, Groth S, Stranberg S, D'Cunha J, et al. (2010) Management of cervical esophageal injury after spinal surgery. *Ann Thorac Surg* 90: 1128-1133.
2. Kuriloff DB, Blaugrund S, Ryan J, O'Leary P (1987) Delayed neck infection following anterior spine surgery. *Laryngoscope* 97: 1094-1098.
3. Whitehill R, Sirna EC, Young DC, Cantrell RW (1985) Late esophageal perforation from an autogenous bone graft. Report of a case. *J Bone Joint Surg Am* 67: 644-645.
4. Oliphant M, Wiot JF, Whalen JP (1976) The cervicothoracic continuum. *Radiology* 120: 257-262.
5. Frank I (1921) Retropharyngeal abscess: *JAMA*. 77: 517-22.
6. Raj TB, Zarod AP (1985) Acute non-tuberculous retropharyngeal abscess in adults (case reports of three patients). *J Laryngol Otol* 99: 1297-1300.
7. Goldenberg D, Golz A, Joachims HZ (1997) Retropharyngeal abscess: a clinical review. *J Laryngol Otol* 111: 546-550.
8. Tannebaum RD1 (1996) Adult retropharyngeal abscess: a case report and review of the literature. *J Emerg Med* 14: 147-158.
9. Pontell J, Har-El G, Lucente FE (1995) Retropharyngeal abscess: clinical review. *Ear Nose Throat J* 74: 701-704.
10. el-Sayed Y, al Dousary S (1996) Deep-neck space abscesses. *J Otolaryngol* 25: 227-233.
11. Karkanevatos A, Beasley NJ, Swift AC (1997) Acute non-tuberculous retropharyngeal abscess in an adult. A case report and review of the literature. *J Laryngol Otol* 111: 169-171.
12. Pickles JM1 (1988) Retropharyngeal abscess complicating a neck wound (a case report). *J Laryngol Otol* 102: 552-553.
13. Singh I, Chanda R, Gupta KB, Yadav SP (2003) Fatal pyothorax: a rare complication of retropharyngeal abscess. *Indian J Chest Dis Allied Sci* 45: 265-268.
14. Brook I (1987) Microbiology of retropharyngeal abscesses in children. *Am J Dis Child* 141: 202-204.
15. Gidley PW, Ghorayeb BY, Stiernberg CM (1997) Contemporary management of deep neck space infections. *Otolaryngol Head Neck Surg* 116: 16-22.
16. Jamjoom (1997) Pharyngo cutaneous fistula following anterior spinal fixation. *British Journal of Neurosurgery*. 69-74
17. Barratt GE, Koopmann CF Jr, Coulthard SW (1984) Retropharyngeal abscess--a ten-year experience. *Laryngoscope* 94: 455-463.
18. Kruyt PM, Boonstra A, Fockens P, Reeders JW, van Lanschot JJ (1996) Descending necrotizing mediastinitis causing pleuroesophageal fistula. Successful treatment by combined transcervical and pleural drainage. *Chest* 109: 1404-1407.
19. Lokman S, Sani A, Sidek DS (1993) Pyopneumothorax--a rare sequelae of retropharyngeal abscess. *J Laryngol Otol* 107: 460-462.
20. Kirse DJ, Roberson DW (2001) Surgical management of retropharyngeal space infections in children. *Laryngoscope* 111: 1413-1422.
21. Schuler PJ, Cohnen M, Greve J, Plettenberg C, Chereath J, et al. (2009) Surgical management of retropharyngeal abscesses. *Acta Otolaryngol* 129: 1274-1279.
22. Kwong JC, Howden BP, Charles PG (2011) New aspirations: the debate on aspiration pneumonia treatment guidelines. *Med J Aust* 195: 380-381.