

An Unusual Neck Swelling Presenting 19 Years Post Anterior Cervical Surgery



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Submission: June 21, 2017; Published: August 31, 2017

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Abstract

We present a case of a 69-year-old male patient presenting with right sided neck swelling within the scar of previous anterior cervical spine decompression and prosthetic fusion 19 years ago. An MRI showed a sinus tract extending to and communicating with a collection of fluid surrounding the synthetic polymer graft. Seeding of the polymer graft with low grade bacteria post anterior cervical discectomy and fusion resulting in a sinus tract is rare. A delayed presentation of such a complication after 19 years has never been described before. The graft was removed and sinus tract successfully treated following surgery. We have discussed potential complicating factors and potential implications of biofilm and antibiotic resistance in this case.

Keywords: Neck lump; Sinus tract; Cervical vertebrae; Biocompatible materials

Introduction

Anterior cervical discectomy and fixation (ACDF) is usually a safe procedure although occasionally, complications can occur [1]. Formation of sinus tract as a complication has been rarely described. However, such a complication with sinus tract extending up to anterior triangle of neck, presenting as a neck swelling, 19 years after original surgery has never been described before. Multidisciplinary team work is very important when formulating a differential diagnosis and collaboration between departments to ensure the best clinical outcome and experience for the patient.

Case Presentation

A 69-year-old male presented with right sided neck swelling adjacent to the scar of a previous ACDF at C4/C5 vertebral level performed 19 years back. He also had pain radiating to the right ear and mandible. The patient was wheelchair-bound for a few years due to long standing progressive spinal degenerative disease, for which he had previously undergone multiple lumbar spine operations.

Previously, the patient had undergone a C4/5 ACDF with biocompatible osteoconductive polymer (BOP) graft 19 years ago for neck pain and right sided radicular pain. A week after surgery, there was wound infection, successfully treated with a course of Flucloxacillin. A radiograph demonstrated good

position of the graft at the time. Subsequently over the next 10 years, the patient complained of a few episodes of sudden severe weakness of upper and lower limbs that lasted only a few minutes, but these were completely self-resolving and had not occurred recently, these had never been investigated by imaging. However he has had gradual reduction in mobility over 10 years, mainly due to problems related to lumbar spine and he has become wheelchair-bound mainly because of chronic pain and mobility related issues.

Clinical examination during the current presentation showed significant myelopathic changes in upper and lower limbs with brisk reflexes and hypertonia; power was 4+/5 in all four limbs, it had been similar for the last few years with no recent deterioration.

An ultrasound of the neck was initially performed that demonstrated small subcutaneous fluid collection that was aspirated but no organism was grown. MRI revealed the prosthesis at C4/5 to be rotated with some surrounding fluid, while a sinus tract was seen communicating between the skin and fluid around prosthesis (Figure 1). A CT scan showed lucency around the implant with bone resorption (Figure 2). Serum CRP and white cell count (WCC) were normal with no pyrexia or other signs of infection.

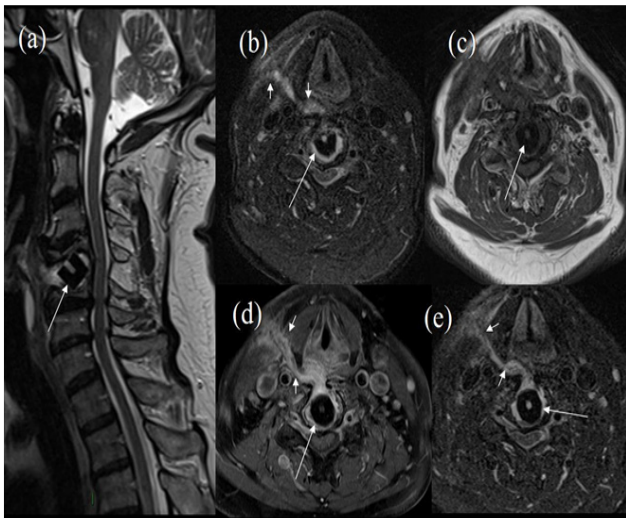


Figure 1: Initial MRI. (a) T2 axials showing rotated disc implant (white arrow) surrounding by fluid. (b) STIR axials, (c) T1 axial and (d,e) Post contrast T1 fat suppressed axials, showing Implant (long white arrow) and the sinus tract (short white arrows). In (b), the sinus tract shows as high signal on STIR (short white arrows). In (d,e), there is enhancement along the sinus tract (small white arrows).



Figure 2: CT scan, sagittal reconstructed image. The rotated implant is seen (white arrow), surrounded by a lucent area and bone resorption.

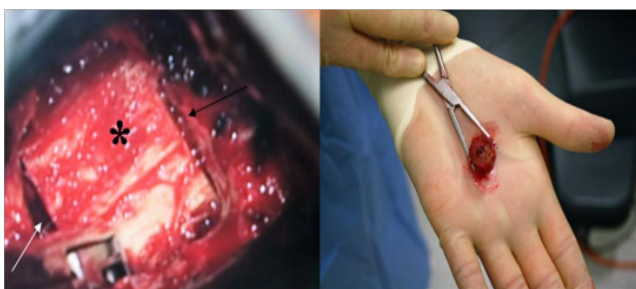


Figure 3: Intraoperative picture (on the left side) showing the exposed implant (*). White arrow represents the superior margin, while black arrow represents the inferior margin. The implant post removal (picture on right side).

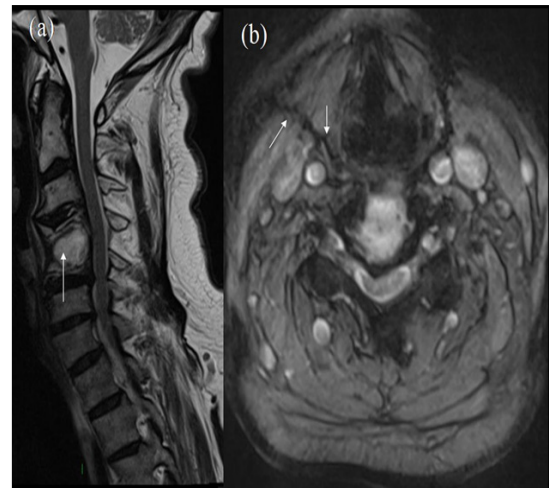


Figure 4: Post operative MRI. (a) T2 sagittal images show the "empty cavity" (white arrow) with otherwise stable appearance of the surrounding bones. (b) T2 gradient echo axial shows the healed sinus tract (white arrows).

The patient was started on IV antibiotics (Flucloxacillin) for 6 weeks and subsequently had a right side anterior cervical wound exploration and C4/5 debridement. Intraoperatively, the cervical disc level was completely calcified on all six sides of the C4/5 fusion (analogy made to the six sides of a dice). The anterior osteophytes were drilled off microscopically and the infected Synthetic Polymer graft was removed (Figure 3). Initially, the plan had been to place an expandable titanium construct in place of the infected C4/5 fusion. However, the remaining 5 sides of the fused C4/5 vertebrae were deemed strong and stable enough to remove the graft without placing a (titanium) foreign body. A precautionary hard collar was placed for 6 weeks instead. Post-operatively, the patient had mild left C5 weakness. Subsequent spinal imaging revealed no new kyphosis or ongoing infection (Figure 4).

All tissue samples from aspiration or from the surgical exploration did not grow any organism even after 7 days of incubation. In the first post-operative day the patient had raised temperature and full septic screen was done. Blood culture showed *Staphylococcus Epidermidis* and *Staphylococcus Hominis* which was resistant to Penicillin and Erythromycin but sensitive to Vancomycin and Daptomycin. Post-operatively, Daptomycin IV was given as per local antibiotic guidelines and microbiology team that continued for six weeks.

Upon clinical review, the neck lump resolved completely. Further physiotherapy and occupation therapy management was arranged for the patient.

Discussion

ACDF is a common procedure for degenerative spinal disease and the complications associated with this procedure can be devastating although most surgical outcomes are satisfactory [1]. Previous complications reported with this procedure are myelopathy, spinal cord, nerve root injury, post-surgical

wound infection, thoracic duct injury, vascular injury, recurrent laryngeal nerve palsy, Horner's syndrome, epidural hematoma, cervical spine instability, discitis, epidural abscess, esophageal perforation, Zenker's Diverticulum and graft migration [1-4].

This patient presented with a 2cmx2cm, right anterior neck lump soft and fluctuant but tender to touch. The common differential diagnosis for midline neck lumps include skin or subcutaneous lesions, enlarged lymph nodes, thyroglossal duct cysts and thyroid masses [5]. Lateral neck lump commonly indicates epidermoid cysts, dermoid cysts, lipomas, lymphangiomas, bronchial cysts, lymphadenopathy, neurogenic tumors, vascular tumors and salivary gland pathologies as common differential diagnosis [5]. A prevertebral abscess presenting as a neck lump after anterior cervical fusion surgery with neck implant is rare, but previous case reports have reported posterior triangle prevertebral abscess discharge and dysphagia secondary to prevertebral abscesses [6].

The incidence of postoperative wound infections in anterior cervical discectomy and fusion is 0.1%-1.6% [7]. Symptoms and signs of infection include neck swelling, pyrexia, elevated inflammatory markers, dysphagia and discharge at the surgical site. Late surgical infection is rare and common causes are esophageal rupture, Zenker's diverticulum, retropharyngeal bacterial colonization and seeding of prevertebral space [4,8].

BOP graft has been frequently used in the past as a vertebral interbody graft for the ACDF procedure. The biocompatible, osteoconductive, biodegradable and synthetic characteristics of a BOP graft are advantageous as compared to an expensive non-homologous graft [9]. Some of the studies have shown a higher rate of intersomatic space collapse and higher surgical complications related directly to the BOP graft [10].

Infection commonly presents immediately after an ACDF procedure as a superficial surgical site infection, osteitis, retropharyngeal abscess, meningitis and epidural abscess [8]. Two cases of delayed prevertebral infection (2-4 months) with retropharyngeal abscess and pharyngocutaneous fistula following cervical spine surgery have been previously reported and the patients presented with neck lump, fever and raised laboratory markers suggesting infection [11]. Another case report documented neck abscess [10] weeks post spinal fusion surgery and the patient also presented with pyrexia, neck lump and raised inflammatory markers [12]. Acute infections occurring in the first three months after surgery are usually caused by virulent microorganisms such as *S. aureus*, whereas delayed infections (3-24 months after surgery) are mostly caused by low virulence microorganisms such as coagulase-negative staphylococci [13] as well as low virulence skin flora like *Propionibacterium* [14] and diphtheroids [10], however there is no further information in literature about any infections happening after longer duration.

Staphylococcus Epidermidis is Gram positive a catalase-positive, coagulase-negative, facultative anaerobe that can grow

by aerobic respiration. It is normal human flora particularly found on skin [15]. *S. epidermidis* infections are generally hospital-acquired [16] particular for patient with catheters or other surgical implants because it is known to form biofilms that grow on these devices [17]. This occurs most commonly on intravenous catheters and medical prostheses and implants [18]. Staphylococcus Hominis is Gram positive, coagulase negative, anaerobic bacteria, second most human normal flora. [19] *S. hominis* is normally found on human skin, but it can cause infections in patient with compromised immune systems [20].

While no organism was grown from the fluid collection, blood cultures were positive. Although it is difficult to be entirely certain, we hypothesize that a biofilm may have formed over the surface of prosthesis. A biofilm is a collection of microorganisms impregnated in a self-produced polysaccharide matrix adherent to a solid biological or non-biological surface [7, 13]. Polysaccharide intercellular adhesin (PIA) produced by staphylococci has been demonstrated to be a crucial factor to form biofilm in implants [21,22]. Prostheses-related infections are now thought to be biofilm-associated infections [23-29] which are highly resistant to antibiotic treatment. [30-35] Reasons have not been fully understood; however few theories and thought has been described like poor antibiotic penetration, nutrient limitation, slow growth, adaptive stress responses etc [36]. In our patient, over the last many years he had several instances of infections (UTI, Skin infection, chest infection etc) due to poor mobility and severe cervical myelopathy. The patient also received many courses of antibiotics and steroids over years. This might explain his weak immune system resulting in susceptibility to infection by *S.Hominis*.

While the exact cause of such delayed presentation of infected BOP implant is uncertain, the fact that there was viable bone around prosthesis would suggest a long standing low grade infection. Since there is a history of wound infection a week after initial surgery, the infection could have occurred then, although it is possible that it happened at a later stage during other infections (chest etc).

In conclusion, we have presented a case of a patient developing a sinus tract extending from an infected disc implant to the anterolateral neck, 19 years after initial surgery, such a long interval has never been reported before. We have also highlighted the potential causes of a delayed presentation including characteristics of organisms involved, mechanism of infection and also discussed other factors complicating this diagnosis. Such cases can be difficult to treat and benefit from a multidisciplinary approach of otolaryngology and neurosurgery besides imaging and microbiology support for optimum outcomes.

References

1. Fountas K, Kapsalaki E, Nikolakakos L, Smisson H, Johnston K, et al. (2007) Anterior cervical discectomy and fusion associated complications. Spine 32(21): 2310-2317.

2. Bertalanffy H, Eggert H (1989) Complications of anterior cervical discectomy without fusion in 450 consecutive patients. *Acta Neurochir* 99(1-2): 41-50.
3. Pompili A, Canitano S, Caroli F, Caterino M, Crecco M, et al. (2002) Asymptomatic esophageal perforation caused by late screw migration after anterior cervical plating. *Spine* 27(23): E499-E502.
4. Summers L, Gump W, Tayag E, Richardson D (2007) Zenker Diverticulum. *Journal of Spinal Disorders & Techniques* 20(2): 172-175.
5. Simo R, Leslie A (2006) Differential diagnosis and management of neck lumps. *Surgery (Oxford)* 24(9): 312-322.
6. Mathesul A, Deokate P, Chandanwale A, Bartakke G, Bhise S, et al. (2015) Late prevertebral abscess with sinus following anterior cervical corpectomy and fusion. *Asian Journal of Neurosurgery* 10(3): 272-276.
7. Fountas KN, Kapsalaki EZ, Nikolakakos LG, Smisson HF, Johnston KW, et al. (2007) Anterior cervical discectomy and fusion associated complications. *Spine (Phila Pa 1976)* 32(21): 2310-2317.
8. Christiano LD, Goldstein IM (2011) Late prevertebral abscess after anterior cervical fusion. *Spine* 36(12): E798-E802.
9. Korovessis P, Repantis T, Vitsas V, Vardakastanis K (2012) Cervical spondylodiscitis associated with oesophageal perforation: a rare complication after anterior cervical fusion. *European Journal of Orthopaedic Surgery & Traumatology* 23(S2): 159-163.
10. Weinstein MA, McCabe JP, Cammisa FP (2000) Postoperative spinal wound infection: a review of 2,391 consecutive index procedures. *J Spinal Disord* 13(5): 422-426.
11. Vrouenraets B, Been H, Brouwer-Mladin R, Bruno M, van Lanschot J, et al. (2004) Esophageal Perforation Associated with Cervical Spine Surgery: Report of Two Cases and Review of the Literature. *Digestive Surgery* 21(3): 246-249.
12. Whitehill R, Sirna E, Young D, Cantrell R (1985) Late esophageal perforation from an autogenous bone graft. Report of a case. *The Journal of Bone & Joint Surgery* 67(4): 644-645.
13. Trampuz A, Widmer AF (2006) Infections associated with orthopedic implants. *Curr Opin Infect Dis* 19(4): 349-356.
14. Richards BS, Herring JA, Johnston CE, Birch JG, Roach JW, et al. (1994) Treatment of adolescent idiopathic scoliosis using Texas Scottish Rite Hospital instrumentation. *Spine (Phila Pa 1976)* 19(14): 1598-1605.
15. <http://textbookofbacteriology.net/staph.html>.
16. Levinson W (2010) Review of Medical Microbiology and Immunology (11th edn), pp. 94-99.
17. Salyers, Abigail A, Whitt Dixie D (2002) Bacterial Pathogenesis: A Molecular Approach, (2nd edn), ASM Press, Washington.
18. Bek-Thomson M, Lomholt HB, Kilian M (2008) Acne is Not Associated with Yet-Uncultured Bacteria. *J Clin Microbiol* 46(10): 3355-3360.
19. Kloos W, Schleifer K (1975) Isolation and Characterization of Staphylococci from Human Skin. *International Journal of Systematic Bacteriology* 25: 62-79.
20. Kloos WE, George CG, Olgiate JS, Van Pelt L, McKinnon ML, et al. (1998) *Staphylococcus hominis* subsp. *novobiosepticus* subsp. nov., a novel trehalose- and N-acetyl-D-glucosamine-negative, novobiocin- and multiple-antibiotic-resistant subspecies isolated from human blood cultures. *International Journal of Systematic Bacteriology* 48 Pt 3: 799-812.
21. Kristian SA, Golda T, Ferracin F, Cramton SE, Neumeister B, et al. (2004) The ability of biofilm formation does not influence virulence of *Staphylococcus aureus* and host response in a mouse tissue cage infection model. *Microb Pathog* 36(5): 237-245.
22. Olson ME, Garvin KL, Fey PD, Rupp ME (2006) Adherence of *Staphylococcus epidermidis* to biomaterials is augmented by PIA. *Clin Orthop Relat Res* 451: 21-24.
23. <https://grants.nih.gov/grants/guide/pa-files/PA-03-047.html>
24. Yang L, Liu Y, Wu H, Song Z, Høiby N, et al. (2012) Combating biofilms. *FEMS Immunol Med Microbiol* 65(2): 146-157.
25. Stoodley P, Kathju S, Hu FZ, Erdos G, Levenson JE, et al. (2005) Molecular and imaging techniques for bacterial biofilms in joint arthroplasty infections. *Clin Orthop Relat Res* 437: 31-40.
26. Costerton JW (2005) Biofilm theory can guide the treatment of device-related orthopaedic infections. *Clin Orthop Relat Res* 437: 7-11.
27. Nguyen LL, Nelson CL, Saccente M, Smeltzer MS, Wassell DL, et al. (2002) Detecting bacterial colonization of implanted orthopaedic devices by ultrasonication. *Clin Orthop Relat Res* 403: 29-37.
28. Costerton JW, Montanaro L, Arciola CR (2005) Biofilm in implant infections: its production and regulation. *Int J Artif Organs* 28: 1062-1068.
29. Gristina AG, Costerton JW (1985) Bacterial adherence to biomaterials and tissue. The significance of its role in clinical sepsis. *J Bone Joint Surg Am* 67(2): 264-273.
30. Zimmerli W (2006) Infection and musculoskeletal conditions: Prosthetic-joint-associated infections. *Best Pract Res Clin Rheumatol* 20(6): 1045-1063.
31. Hengzhuang W, Wu H, Ciofu O, Zhijun Song, Niels Høiby, et al. (2011) Pharmacokinetics/pharmacodynamics of colistin and imipenem on mucoid and nonmucoid *Pseudomonas aeruginosa* biofilms. *Antimicrob Agents Chemother* 55(9): 4469-4474.
32. Hengzhuang W, Wu H, Ciofu O, Song Z, Høiby N, et al. (2012) In vivo pharmacokinetics/pharmacodynamics of colistin and imipenem in *Pseudomonas aeruginosa* biofilm infection. *Antimicrob Agents Chemother* 56(5): 2683-2690.
33. Hoiby N, Krogh JH, Moser C, Song Z, Ciofu O, et al. (2001) *Pseudomonas aeruginosa* and the in vitro and in vivo biofilm mode of growth. *Microbes Infect* 3(1): 23-35.
34. Hoiby N, Ciofu O, Johansen HK, Song ZJ, Moser C, et al. (2011) The clinical impact of bacterial biofilms. *Int J Oral Sci* 3(2): 55-65.
35. Hoiby N, Bjarnsholt T, Givskov M, Molin S, Ciofu O, et al. (2010) Antibiotic resistance of bacterial biofilms. *Int J Antimicrob Agents* 35(4): 322-332.
36. Stewart PS (2002) Mechanisms of antibiotic resistance in bacterial biofilms. *Int J Med Microbiol* 292(2): 107-113.



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DOI: 10.19080/OAJNN.2017.6.555678

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