17-Year-Delayed Fistula Formation After Elective Spinal Instrumentation: A Case Report

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Received: February 20, 2015; Revised: March 19, 2015; Accepted: April 18, 2015

Introduction: A late-developing infection after an uneventful initial spinal instrumentation procedure is rare. Delayed infection and new fistula formation have been reported from a few months to 13 years. Here we report an unusual 17-year-delayed fistula formation after primary spinal instrumentation. The patient underwent hardware removal surgery with antibiotic therapy as a definitive treatment.

Case Presentation: Here we report an unusual 17-year delayed fistula formation after primary spinal instrumentation due to spinal trauma. He was admitted to Ghaem General Hospital, a chief referral center, Mashhad, North-East of Iran in August 2014. The patient underwent hardware removal surgery with antibiotic therapy as a definitive treatment.

Conclusions: Late inflammation may occur around spinal instruments and results in cutaneous fistula formation. After oral or intravenous antibiotic treatment, total device extraction is the cornerstone of treatment.

Keywords: Delayed; Fistula; Instrumentation

1. Introduction

Postoperative wound infection following spinal surgery poses high morbidity and mortality, and in this regard, posterior instrumentation has been reported the most frequent procedure (1). Total instrument removal and subsequent primary wound closure with systemic antibiotics is the treatment of choice (2, 3). However, the situation becomes problematic where insufficient bony fusion or pseudarthrosis requires spinal stabilization. Primary implant removal with subsequent reinstrumentation is susceptible to complications of the potentially infectious wound.

2. Case Presentation

A 53-year-old man complaining of exudative fluid discharge of his right flank since one month ago, was admitted to Ghaem General Hospital, a chief referral center, in Mashhad City, North-East of Iran in August 2014. On the physical examination, he was not pyretic; a draining fistula was located in his right flank, associated with pain, erythema, and local edema. His past history was positive for a falling accident 17 years ago with burst fracture of the third lumbar vertebra, and subsequent posterolateral fusion and instrumentation. The surgical and postsurgical periods were both uneventful with a 10-year follow up. Fistula tract was demonstrated on the fistulography and CT-fistulography, opening to the first lumbar pedicle screw (Figure 1A, 1B). There was no abnormality in his laboratory tests except for elevated erythrocyte sedimentation rate. Blood and smear samples were collected, which they were both negative for any microorganism. Our patient underwent hardware removal surgery with perioperative empirical antibiotics (cephalexin and ciprofloxacin). During the procedure exudative fluid was discovered at the site of the right first lumbar pedicle screw. Adequate smear and culture were obtained with a specimen withdrawn for pathological studies. All the samples were negative for bacteria. The pathological study was consistent with fibro-muscular tissue and non-specific chronic inflammation. Fistula site healed spontaneously and our patient was discharged 10 days after the operation without any complication (Figure 2A, 2B).

3. Discussion

Spinal fusion and instrumentation infection rate varies from 0.7% to 8.5% in the early postoperative course (4). However, the late onset (longer than one month postoperatively) infection is uncommon, as we demonstrated a 17 years gap between the incident and the fistula formation. Delayed infection rate was reported after instrumented spine surgery from 0.2% to 6.9% (5, 6).

Table 1 summarizes the cases with late infection reported in the literature since 1993. The period between the initial surgical procedure and the inflammatory symptoms
varies from a few months to many years, as Mhaidli et al. reported a case with spontaneous drainage 13 years after the first spinal instrumentation (7). Here we report the longest interval, about 17 years.

*Staphylococcus* species was the most common organism found in the surgical site (8-11), with the next most detected bacteria being *Propionibacterium acnes* (2, 5, 12). The inflammatory reaction against the metallic device may have a role in these cases, so the removal of the instrument is necessary whether the organism is detected or not. Very late inflammation reaction may occur around spinal instruments and results in cutaneous fistula formation. After oral or intravenous antibiotic treatment, total device extraction is the cornerstone of treatment.

### Table 1. Summary of Late Infections Reported After Instrumented Spine Surgery<sup>a</sup>

| Reference                  | Number of Patients | Clinical Symptoms                                                                 | Risk Factors                                                                 | Findings From Culture                                                                 | Treatment                          | Suspected Cause            | Year of Publication |
|----------------------------|--------------------|-----------------------------------------------------------------------------------|------------------------------------------------------------------------------|--------------------------------------------------------------------------------------|------------------------------------|--------------------------|----------------------|
| Heggeness et al. (12)      | 6                  | Back pain (3), fever (3), spontaneous drainage of painful swelling (4), retroperitoneal abscess (1), fluctuation mass (1) | Paraplegia with neurogenic bladder (2), Pyelonephritis (2), renal calculi (1), intravenous drug abuse (2) | *Propionibacterium acnes* (1), *S. aureus* (1), *Streptococcus morbillorum* (1), *S. epidermidis* (1) | Instrument removed (3), abscess drainage under CT guidance (1), needle aspiration (1) | Aseptic loosening of hardware (1), retroperitoneal abscess (1) | 1993                 |
| Dubousset et al. (13)      | 18                 | Incisional swelling and pain, spontaneous drainage                               | Not stated                                                                  | *S. epidermidis* (2), negative (16)                                                  | Instrumentation removed            | Micromotion; metal fretting causing sterile inflammation | 1994                 |
| Viola et al. (5)           | 8                  | Wound drainage (2), abscess (4) back pain (8), Elevated ESR (7)                 | Smoker (1), Malnutrition (1), Neurogenic bladder (2)                         | *S. epidermidis* (6), *Propionibacterium acnes* (1), negative (13)                   | Instrumentation removed            | Intraoperative inoculation | 1997                 |
| Antuna et al. (14)         | 1                  | Pain, spontaneous drainage                                                      | None                                                                        | Negative (6), no culture taken (2)                                                   | Instrumentation removed            | Metal fretting, Chronic inflammation or low virulent bacteria | 1998                 |
| Wimmer and Gluch (3)       | 8                  | Aseptic loosening of hardware, radiculopathy around pedicle screws, pain and swelling in 6 patients who had discharging sinus | None                                                                        | Negative (6), no culture taken (2)                                                   | Instrumentation removed            | Metal fretting, micromotion | 1998                 |
| Clark and Shuflebarger (2) | 22                 | Fluctuant mass or drainage, pain rarely a factor, no fever                     | none                                                                        | *S. epidermidis* (6), *Enterococcus* (2), *S. aureus* (1), *Propionibacterium acnes* (3) | Instrumentation removed            | Intraoperative inoculation | 1999                 |
| Weinstein et al. (16)      | 3 (of a series of 46 infections) | Not specifically stated for 3 patients                                         | Not specifically stated                                                      | Not specifically stated; *S. aureus* found in 34 of 46                              | Instrumentation removed            | Not stated                | 2000                 |
| Richards and Emara (17)    | 23                 | Spontaneous drainage (15), fluctuance (6), pain (9), fever (3)                 | Reoperation for dislodgement of hook (1), intravenous drug abuse (1)        | *Propionibacterium acnes* (12), *S. epidermidis* (4), *Micrococcus varians* (11), *S. aureus* (1), negative (5) | Instrumentation removed            | Intraoperative inoculation | 2001                 |
| Authors          | Year | Case Number | Diagnosis                                                                 | Symptoms and Findings                                                                 | Infection Site | Pathogen(s)                                                                 | Treatment                                                                 | Duration |
|------------------|------|-------------|---------------------------------------------------------------------------|--------------------------------------------------------------------------------------|----------------|-----------------------------------------------------------------------------|---------------------------------------------------------------------------|----------|
| Etemadrezaei H et al. (18) | 2003 | 5           | Local subcutaneous abscess, remained patients had a local drainage        | Instrumentation failure and loosening                                                | Instrumentation failure and loosening | Coagulase-negative Staphylococci (3), Acinetobacter baumannii (1), Peptostreptococcus (1) | Instrumentation removed                                                   | Not stated |
| Bose (6)         | 2003 | 4           | Back pain (1), incisional swelling (2), spontaneous drainage (3), abscess (1), hip pain (2), mental confusion (1) | Dental infection (1), neurogenic bladder (1), kidney infection (2), Knee replacement surgery (1) | Dental infection (1), neurogenic bladder (1), kidney infection (2), Knee replacement surgery (1) | S. aureus (2), not tested (1), S. aureus suspected but no organisms found probably due to long-term course of antibiotics | Irrigation and debridement (2), Instrumentation removed (2), Antibiotic therapy alone (1) | Hematogenous seedling (4) |
| Muschik et al. (1) | 2004 | 45          | Wound sinus and spontaneous drainage of fluid (40), local pain (38), swelling (34), redness (28), Fever > 38.0°C (7) | History of allergic predisposition, protracted postoperative fever, and nonunion of the fusion | History of allergic predisposition, protracted postoperative fever, and nonunion of the fusion | Staphylococcus aureus (6), Staphylococcus epidermidis (2) | Instrumentation removed                                                   | Implant bulk, metalurgical reactions, contamination with lowvируulence microorganisms |
| Hahn et al. (6)  | 2005 | 7           | Sudden Onset Local pain and swelling (7); Sinus Drainage (2)              | None                                                                                 | None | Propionibacterium acnes (6), Not found (1)                                  | Instrumentation removed                                                   | Intraoperative inoculation                                                   |         |
| Kowalski et al. (9) | 2007 | 51          | Back Pain (33), Wound drainage (16), Sinus tract present (13), Neurologic deficits (8) | Diabetes mellitus (1), Systemic malignancy (6), Heparic failure (1), Immunosuppressive medication use (6), End-stage renal disease (1) | Diabetes mellitus (1), Systemic malignancy (6), Heparic failure (1), Immunosuppressive medication use (6), End-stage renal disease (1) | Coagulase-negative staphylococci (9), Propionibacterium acnes (6), Staphylococcus aureus (9), Gram-negative bacilli (1), Streptococci (4), Polymicrobial infection (12), Negative (8) | Antibiotic therapy (6), Instrumentation removed (45) | Intraoperative inoculation of lowvируulence organisms |
| Emel et al. (19) | 2007 | 1           | Purulent flow developed in the posterior skin scar                         | L3-Giant cell tumor                                                                  | L3-Giant cell tumor | Staphylococcus aureus                                                        | Instrumentation removed                                                   | Intraoperative inoculation                                                   |         |
| Mok et al. (10)  | 2009 | 4           | Drainage (2), Operative site pain (2)                                     | None                                                                                 | None | S. epidermidis (4 of 4), Propionibacterium acnes (3 of 4)                    | Antimicrobial therapy (1), Instrumentation removed (6) | Intraoperative inoculation                                                   |         |
| Sierra-Hoffman et al. (11) | 2010 | 7           | Drainage (4), Fever (3), Erythema (1)                                    | Not specified, for late onset infection                                             | Not specified, for late onset infection | Methicillin-resistant Staphylococcus aureus (3), Pseudomonas aeruginosa (2), Enterococcus faecalis (2) | Antibiotic therapy (1), Instrumentation removed (6) | Intraoperative inoculation                                                   |         |
| Mhaidli et al. (7) | 2012 | 1           | Spontaneous drainage                                                      | Non Stated                                                                           | Non Stated | Propionibacterium acnes                                                     | Instrumentation removed                                                   | Intraoperative inoculation                                                   |         |
| Farshad et al. (20) | 2012 | 7           | Sudden Pain, Swelling and Fluctuation                                     | Not Stated                                                                           | Not Stated | Propionibacterium acnes                                                     | Instrumentation removed                                                   | Intraoperative inoculation                                                   |         |
| Messina et al. (21) | 2014 | 7           | (of a series of 23 infections)                                            | Not specifically, stated, wound drainage (19), pain (8), fever (7)                  | Not Stated | Not specified, for late onset infection                                      | Instrumentation removed                                                   | Intraoperative inoculation                                                   |         |
| Our Case         |      | 1           | Elevated ESR, Fistula formation                                            | None                                                                                 | None | Negative                                                                     | Instrumentation removed                                                   | Not stated                                                                 |         |

* Abbreviation: ESR: erythrocyte sedimentation rate.
Authors’ Contributions

Hamid Etemadrezaei developed the original idea, revisied the manuscript, supervised the treatment and was guarantor. Samira Zabihiyan prepared the manuscript and finally revised the manuscript. Aidin Shakeri prepared the manuscript, helped in follow up of the patient and acquisition of data. Babak Ganjeifar helped in follow up of the patient, acquisition of data, abstracted the findings, and revised the manuscript.

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