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

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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 nonspecific 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 [a]

Reference	Number of Patients	Clinical Symptoms	Risk Factors	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/ renal calculi (1), intravenous drug abuse (2)	Paraplegia with neurogenic bladder (2), Pyelonephritis/ renal calculi (1), intravenous drug abuse (2)	*Propionibacterium acnes* (1), *S.aureus* (3), *Streptococcus morvillorium* (1), *S.epidermides* (1)	Instrument removed (3), abscess drainage under CT guidance (1), needle aspiration (1)	Hematogenous seeding	1993
Robertson and Taylor (4)	3	Elevated ESR (2), feeling of malaise (3), abscess in groin (2)	Not stated	Not stated	*Proteus mirabilis* (1), none found (1) not stated (1)	Instrumentation removed	Intraoperative inoculation	1993
Dubousset et al. (13)	18	Incisional swelling and pain, spontaneous drainage	Not stated	Not stated	*S.epidermis* (2), negative (16)	Instrumentation removed	Micromotion; metal fretting causing s sterile inflammation	1994
Viola et al. (5)	8	Wound drainage (2), abscess (4) back pain (8), Elevated ESR (7)	Smoker (3), Malnutrition (1), Neurogenic bladder (2)	Smoker (3), Malnutrition (1), Neurogenic bladder (2)	*S.epidermidis* (6), *Proprionibacterium acnes* (1), negative (13)	Instrumentation removed	Intraoperative inoculation	1997
Antuna et al. (14)	1	Pain, spontaneous drainage	None	None	Not reported	Instrumentation removed	Not stated	1997
Hatch et al. (15)	1	Back pain, tenderness over instrumentation, Westergren sedimentation rate 38%	None	None	Negative	Instrumentation removed	Metal fretting, Chronic inflammation or low virulent bacteria	1998
Wimmer and Gluch (3)	8	Aseptic loosening of hardware, radiolucency around pedicle screws, pain and swelling in 6 patients who had discharging sinus	None	None	Negative (6), no culture taken (2)	Instrumentation removed (6), none (2)	Metal fretting, micromotion	1998
Clark and Shufflebarger (2)	22	Fluctuant mass or drainage, pain rarely a factor, no fever	none	none	*S.epidermides* (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	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)	Reoperation for dislodgement of hook (1), intravenous drug abuse (1)	*Propionibacterium acnes* (12), *S epidermides* (4), *Micrococcus* varians (13), *S. aureus* (1), negative (5)	Instrumentation removed	Intraoperative inoculation	2001

Soultanis et al. (18)	5	local subcutaneous abscess, whereas the remaining patients had a local drainage	instrumentation failure and loosening	instrumentation failure and loosening	coagulase-negative Staphylococci (3), *Acinetobacter baumani* (1), *Peptostreptococcus* (1)	Instrumentation removed	Not stated	2003
Bose (8)	4	Back pain (1), incisional swelling (2), spontaneous drainage (1), abscess (1), hip pain (1), mental confusion (1)	Dental infection (1), neurogenic bladder (1), kidney infection (1), Knee replacement surgery (1)	Dental infection (1), neurogenic bladder (1), kidney infection (1), 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/debridement (2), instrumentation removed (2), antibiotic therapy alone (1)	Hematogenous seeding (4)	2003
Muschik et al. (1)	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, metallurgic reactions, contamination with low-virulence microorganisms	2004
Hahn et al. (6)	7	Sudden Onset Local pain and swelling (7), Sinus Drainage (2)	None	None	*Propionibacterium acnes* (6), Not found (1)	Instrumentation removed	Intraoperativeinoculation	2005
Kowalski et al. (9)	51	Back Pain (33), Wound drainage (16), Sinus tract present (13), Neurologic deficits (8)	Diabetes mellitus (1), Systemic malignancy (6), Hepatic failure (1), Immunosuppressive medication use (8), End-stage renal disease (1)	Diabetes mellitus (1), Systemic malignancy (6), Hepatic failure (1), Immunosuppressive medication use (8), End-stage renal disease (1)	coagulase-negative staphylococci (9), *Propionibacterium acnes* (6), *Staphylococcus aureus* (11), Gram-negative bacilli (1), Streptococci (4), Polymicrobial infection (12), negative (8)	Antibiotic therapy (6), Instrumentation removed (45)	Intraoperative inoculation of low-virulence organisms	2007
Emel et al. (19)	1	Purulent flow developed in the posterior skin scar	L3-Giant cell tumor	L3-Giant cell tumor	*Staphylococcus aureus*	Instrumentation removed	Intraoperative inoculation	2007
Mok et al. (10)	4	Drainage (2), Operative site pain (2)	None	None	*S. epidermidis* (4 of 4), *Propionibacterium acnes* (3 of 4)			2009
Sierra-Hoffman et al. (11)	7	Drainage (4), Fever (3), Erythema (1)	Not specified, For late onset infection	Not specified, For late onset infection	Methicillin-sensitive *Staphylococcus aureus* (3), *Pseudomonas aeruginosa* (2), *Enterococcus faecalis* (2)	Antibiotic therapy (1), Instrumentation removed (6)	Intraoperativeinoculation	2010
Mhaidli et al. (7)	1	Spontaneous drainage	Non Stated	Non Stated	*Propionibacterium acnes*	Instrumentation removed	Intraoperative inoculation	2012
Farshad et al. (20)	7	Sudden Pain, Swelling and Flactuation	Not Stated	Not Stated	*Propionibacterium acnes* (6)	Instrumentation removed	Intraoperative inoculation, metal fretting	2012
Messina et al. (21)	7 (of a series of 23 infections)	Not specifically, stated, wound drainage (19), pain (8), fever (7)	Not Stated	Not Stated	Not specified, for late onset infection	Instrumentation removed	Intraoperative inoculations	2014
Our Case	1	Elevated ESR, Fistula formation	none	none	negative	Instrumentation removed	Not stated	

[a] Abbreviation: ESR: erythrocyte sedimentation rate.

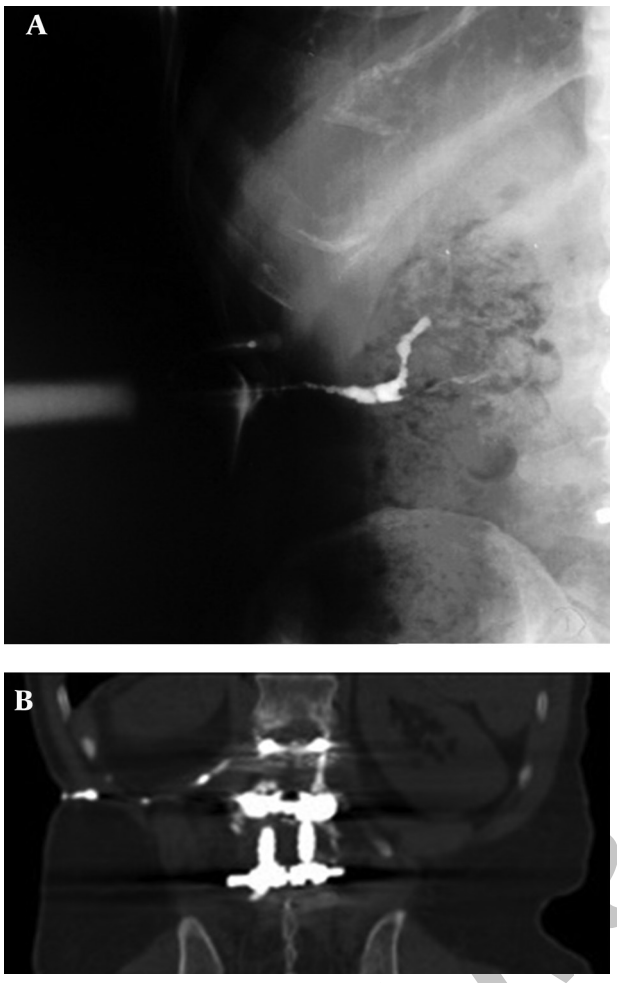

Figure 1. Fistulography; A, CT Fistulography; B, Coronal view.

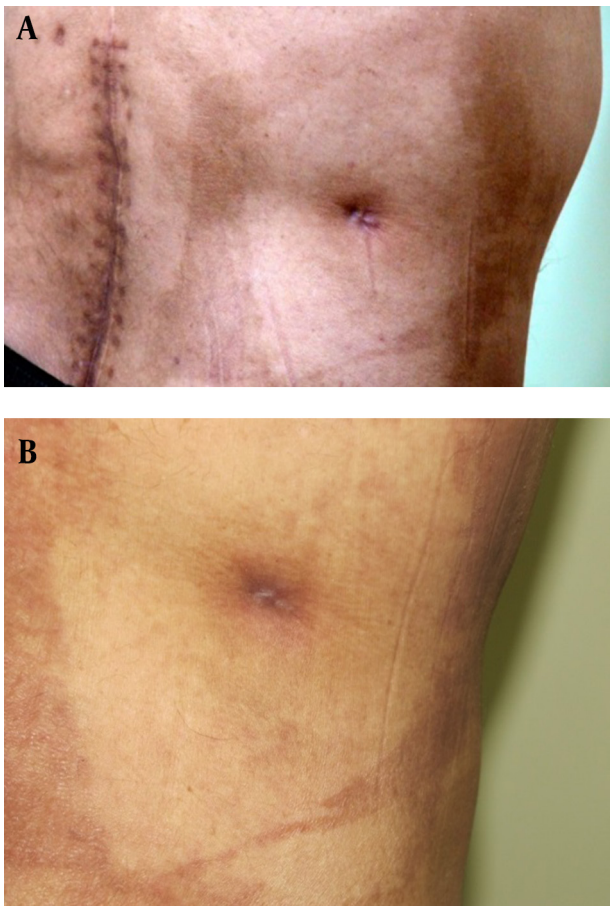

Figure 2. A, Fistula Drainage preoperatively; B, Resolved Fistula Drainage 6 Months Follow-up

Authors' Contributions

Hamid Etemadrezaei developed the original idea, revised the manuscript, supervised the treatment and was guarantor. Samira Zabihyan 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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