Joint Diseases and Related Surgery

Jt Dis Relat Surg
2023;34(3):731-736

CASE REPORT

A rare case of aggressive pyogenic spondylitis with giant abscesses after vertebral augmentation

Lin Liu, MMed[1], Lei He, MD[1], Ga Long, MMed[2], Min He, MD[1]

[1]Department of Orthopeadic Surgery, The Public Health Clinical Center of Chengdu, Chengdu, China
[2]Department of Orthopeadic Surgery, Dege County People's Hospital, Ganzi Prefecture, China

ABSTRACT

Although *Streptococcus constellatus* (SC), an opportunistic pathogen, can cause abscesses and empyema. The SC infection after vertebral augmentation (VA) can interfere with patients' daily living activities and can be life-threatening in severe cases. A 67-year-old male complained of lumbar pain for two months. The patient underwent percutaneous vertebroplasty of the second and third lumbar vertebrae two months ago. On admission, laboratory and imaging evidence suggested infection of the second and third lumbar vertebrae with bilateral psoas major and left lumbodorsal abscesses. After three weeks of empirical anti-infective therapy, abscess removal and the second and third lumbar vertebrae fusion with iliac bone graft were performed under general anesthesia. Intraoperative pathology and next-generation sequencing (NGS) examination of the pus suggested SC infection, and oral linezolid was given for 12 weeks after surgery. The infection was eventually cured and the patient achieved satisfactory function. In conclusion, pyogenic spondylitis due to SC infection after VA is a life-threatening complication. In addition to infectious disease consultation and routine etiological screening, NGS is important to identify infection with unknown pathogens. Surgery combined with sensitive antibiotics is appropriate for patients with progressive neurological deficits.

Keywords: Abscess, pyogenic spondylitis, *Streptococcus constellatus*, vertebral augmentation, vertebroplasty.

Streptococcus constellatus (SC) is a part of *Streptococcus anginosus* group and is usually colonized in the normal mouth, gut, and urogenital tract flora.[1] However, in some specific cases, it has the potential to spread to physiologically sterile anatomical sites and cause infections. According to its anatomical distribution, SC can cause suppurative infections at multiple anatomical sites, mostly in patients who are immunosuppressed or undergoing invasive procedures. A retrospective cohort study reported that SC was mainly associated with bacteremia, while the presence of abscesses or empyema was uncommon.[2]

Vertebral augmentation (VA) has gradually become one of the main methods for the treatment of osteoporotic vertebral fractures (OVF) due to the advantages of minimal surgical trauma, simple technique, and immediate pain relief.[3] Nevertheless, multiple complications associated with the procedure remain a non-negligible problem. Previous studies have shown that although the incidence of pyogenic spondylitis after VA is not high, the consequences are serious.[4] Due to the presence of polymethylmethacrylate (PMMA), conventional anti-infective treatment measures are insufficient, and surgery inevitably becomes one of the main treatment methods for such patients. Currently, relevant studies on pyogenic spondylitis after VA surgery mainly focus on the high risk factors of postoperative infection, clinical manifestations after infection and clinical diagnosis;[4,5] however, there is no in-depth study on the surgical treatment of such patients.

Received: April 23, 2023
Accepted: May 23, 2023
Published online: September 16, 2023

Correspondence: Min He, MD. Department of Orthopeadic Surgery, The Public Health Clinical Center of Chengdu No. 377, Jingming Road, Jinjiang District, Chengdu, 610061 China.

E-mail: 26771825@qq.com

Doi: 10.52312/jdrs.2023.1182

** The first and fourth authors contributed equally to this manuscript.*

Citation: Liu L, He L, Long G, He M. A rare case of aggressive pyogenic spondylitis with giant abscesses after vertebral augmentation. Jt Dis Relat Surg 2023;34(3):731-736. doi: 10.52312/jdrs.2023.1182.

In this article, we present a rare case of a male patient presenting with pyogenic spondylitis due to SC with bilateral paravertebral and left lumbodorsal abscess formation after VA.[6]

CASE REPORT

A 67-year-old male was admitted to hospital complaining of lumbar pain for two months. Percutaneous vertebroplasty (PVP) for osteoporosis with compression fractures of the second and third lumbar vertebrae was performed at the local hospital two months ago. The patient denied a history of infectious diseases such as tuberculosis. The main comorbidity was diabetes mellitus. Since the Visual Analog Scale (VAS) score of lumbar pain after PVP surgery fluctuated between 2 and 4, and there was no fever symptom, and the patient always misinterpreted it as residual pain after minimally invasive surgery. He did not seek medical attention, until his lumbar pain reached 9 points. Due to the consideration of spinal tuberculosis, he was referred to our hospital.

On admission, the patient's initial vital signs were as follows: body temperature 36.5°C, heart rate 133 bpm, respiratory rate 24, blood pressure 136/75 mmHg, and oxygen saturation (SpO_2) 96% on room air. The patient was wheeled into the ward. He maintained a forced hip flexion position. The left thoracic back and waist were visibly swollen, with significant undulation when pressed. When tapping his thoracic back, the pain was obvious, with pressure pain in the spinous processes of the second and third lumbar vertebrae and bilateral paravertebral soft tissue. In addition, the bilateral Ely's tests were positive; however, the feeling of saddle area was normal, and the pathological signs were negative. Cardiac, respiratory, and abdominal examinations were unremarkable.

Preliminary laboratory tests revealed mild anemia, leukocytosis (primarily neutrophilic), mild liver function impairment, hypoproteinemia, and significant elevation of erythrocyte sedimentation rate (ESR), C-reaction protein (CRP), and procalcitonin (PCT) (Table I). In addition, the *Mycobacterium tuberculosis (M. tuberculosis)* immunoglobulin antibody was positive, while the T-cell spot of tuberculosis (T-SPOT.TB) test was negative. Anteroposterior and lateral radiographs of the lumbar spine on admission revealed loss of height at the second and third lumbar vertebrae and secondary kyphosis (Figure 1). In addition to these findings, a lumbar computed tomography (CT) scan showed bone destruction at the second and third lumbar vertebrae, formation of bilateral psoas major abscesses with separation, and left lumbodorsal abscesses (Figure 1).

As the etiological agent was unclear and tuberculosis infection was initially suspected, strong diagnostic anti-tuberculous therapy was administered empirically after admission. The drugs included isoniazid, rifampin, ethambutol, pyrazinamide, levofloxacin, and linezolid. After 10 days of treatment, the white blood cell (WBC), neutrophils, CRP, ESR and PCT decreased (Table I), suggesting that anti-infective treatment was effective. However, anemia was worse than at the time of admission, which was considered to be caused by severe infectious depletion. After three weeks of anti-infection, anemia correction, liver preservation and improvement of nutritional status, the patient's general condition was

TABLE I
Laboratory test results

Item	Admission	10 days after admission	3 weeks after admission	Discharge
WBC (L)	20.45×10^9 ↑↑	6.35×10^9	5.32×10^9	6.24×10^9
Neutrophils (L)	17.62×10^9 ↑↑	4.13×10^9	3.27×10^9	4.05×10^9
Hemoglobin (g/L)	110.0 ↓	86.0 ↓↓	107.3 ↓	115.2 ↓
ALT (U/L)	40 ↑	18	14	63 ↑
AST (U/L)	45 ↑	30	23	72 ↑
Albumin (g/L)	19.2 ↓↓	21.4 ↓↓	28.2 ↓	31.7 ↓
CRP (mg/L)	168.39 ↑↑	78.11 ↑	48.39 ↑	18.28 ↑
ESR (mm/h)	107.0 ↑↑	99.0 ↑↑	57.0 ↑	46.0 ↑
PCT (ng/mL)	1.231 ↑↑	0.843 ↑	0.356 ↑	0.307 ↑

WBC: White blood cell; ALT: Alanine transaminase; AST: Aspartate transaminase; CRP: C-reactive protein; ESR: Erythrocyte sedimentation rate; PCT: Procalcitonin; ↑: Mildly increased level, ↑↑: Highly increased level, ↓: Mildly reduced level, ↓↓: Highly reduced level.

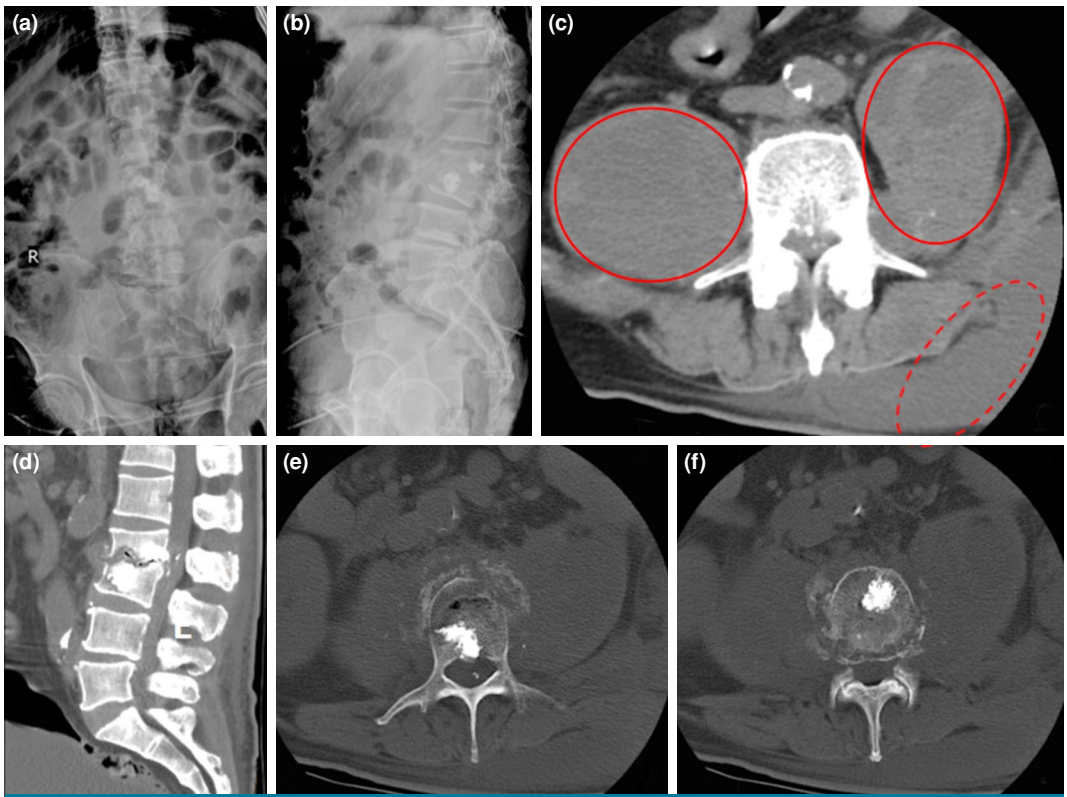

FIGURE 1. (a,b) X-rays revealed wedge-shaped changes in the L2-3 containing bone cement and a mild lumbar kyphosis. **(c)** CT revealed massive bilateral abscess formation in the psoas major muscle with separation within the abscess (red solid line) and a massive abscess in the left lumbodorsal section (red dashed line). **(d)** The sagittal view showed bone destruction and partial loss of vertebral height in L2-3, with a little bone cement visible in the spinal canal. **(e,f)** Cross-sectional CT at the level of L2-3 suggested vertebral bone destruction with spinal canal stenosis.
CT: Computed tomography.

significantly improved (Table I). The infection was effectively controlled and the abscess lesion did not develop further. Therefore, we decided to perform surgical treatment.

After general anesthesia, the patient was placed in the right lateral position, and the extent of the superficial lesion was roughly drawn according to the fluctuating sensation (Figure 2). During the operation, an oblique incision of approximately 15 cm in length was made in the left lumbar region. Exploration revealed a significant swelling of the left psoas major muscle forming an abscess cavity. The abscess in the left lumbar region reached up to the scapular region and anteriorly to the surface of the pectoralis major muscle. After blunt separation, the abscess cavity was separated and a large amount of necrotic tissue and about 3,700 mL of brown pus had accumulated in it (Video 1). The pus was sucked up and necrotic tissue was scraped out. A few specimens were taken for pathological examination. A 6-cm-long auxiliary oblique incision was made in the left anterior chest wall in the same position, and the pus cavity was incised layer by layer. The pus cavity was sequentially flushed with a large amount of hydrogen peroxide and saline, and a drainage tube was placed. Subsequently, a left lateral position was chosen and an oblique incision of about 15 cm was made on the right side of the lumbar side, and the right psoas major muscle was found to be swollen and forming an abscess cavity with an accumulation of necrotic tissue and about 300 mL of brown pus, which was aspirated and the necrotic tissue was scraped away, and a small amount of specimen was retained for pathological examination. Free bone and necrotic tissue were removed from the L2 and 3 vertebrae, and a bone groove was cut in the L2-3 vertebrae to measure the length of the groove. Subsequently, the right iliac crest was subcutaneously freed and the required iliac bone was routinely cut and set aside. The iliac bone was trimmed appropriately and, then, inlaid and pressed into the bone groove, and the bone graft was

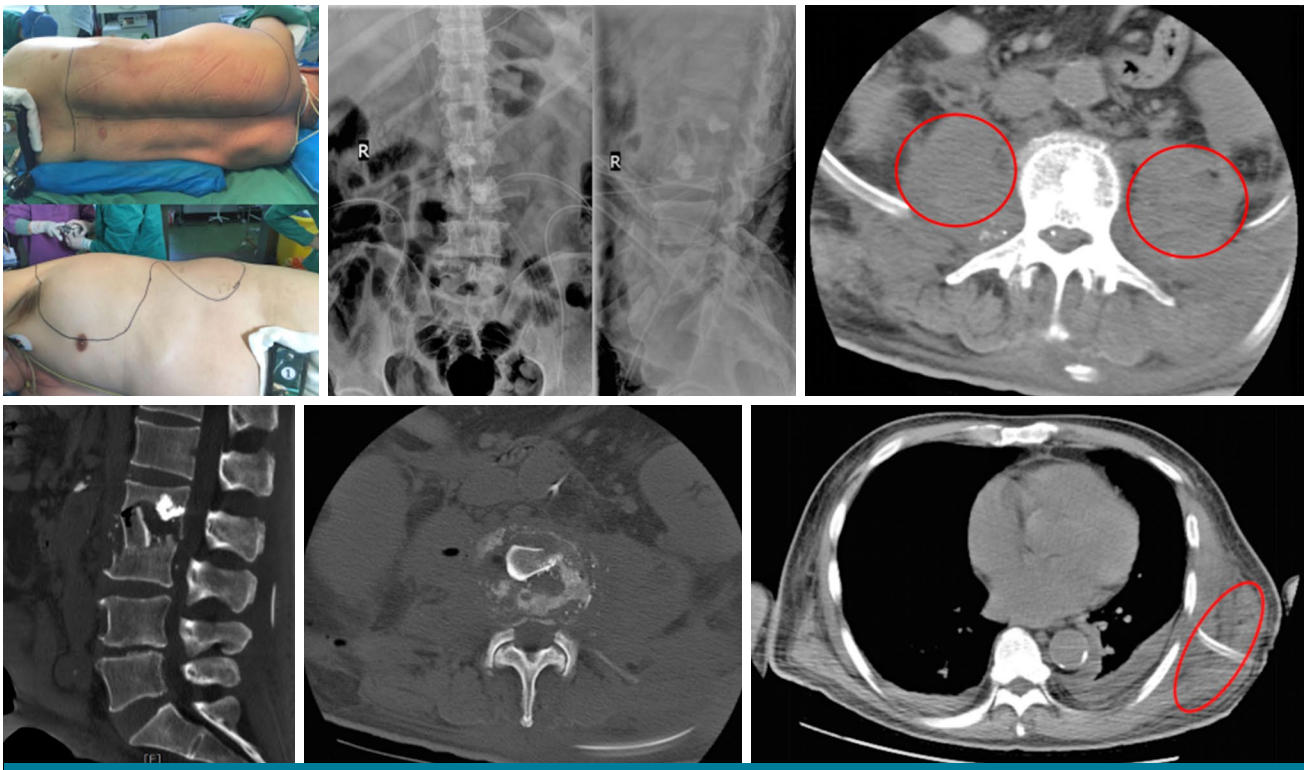

FIGURE 2. The scope of the superficial abscess can be seen in the gross view, that is, the left lumbodorsal section and anterior chest wall (black line area on patient). Radiographs immediately after surgery showed correction of lumbar kyphosis and satisfactory placement of bone graft. Computed tomography revealed significant reduction of abscesses in the bilateral psoas major, left lumbodorsal section and left chest wall (red solid line). The implanted iliac bone was located in the L2-3 intervertebral space, and spinal stenosis was improved.

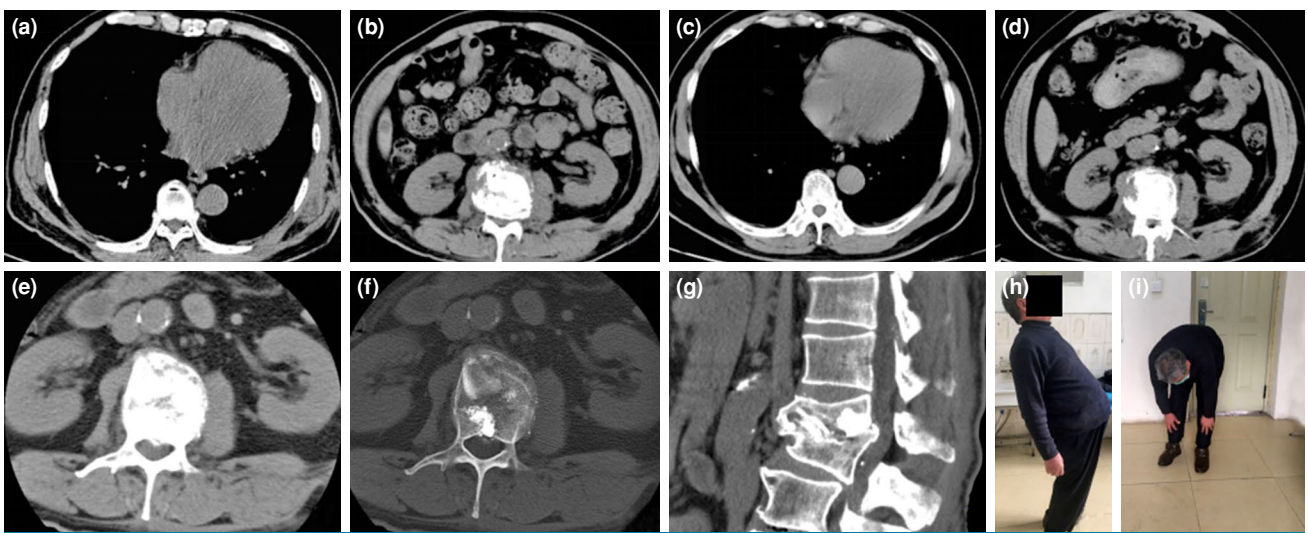

FIGURE 3. (a,b) A repeat CT examination two months after surgery showed a significant reduction of the abscess. **(c,d)** The abscess almost completely disappeared at five months postoperatively with no signs of recurrence. **(e-g)** One year after surgery, CT showed that bony fusion of the implanted iliac bone and disappearance of the abscess. **(h,i)** The patient obtained satisfactory result.

CT: Computed tomography.

checked for stability. After complete hemostasis of the operated field, the wound was flushed with large amount of hydrogen peroxide and saline in turn, and a plasma tube was placed and the incision was closed layer by layer. The amount of bleeding during the operation was about 400 mL.

Immediate postoperative imaging showed significant reduction of abscess, correction of lumbar kyphosis, and satisfactory position of the implanted iliac bone (Figure 2). *Mycobacterium tuberculosis* deoxyribonucleic acid (DNA) test of intraoperative pus and TB Xpert were negative, while pathological examination suggested tissue necrosis with acute inflammatory changes. Next-generation sequencing (NGS) examination of the pus suggested SC, and the general bacterial culture of the drainage was negative on three occasions. Based on these findings, septic spondylitis caused by Streptococcus was considered and, therefore, oral linezolid 600 mg bid anti-infective treatment was given for 12 weeks according to the drug sensitivity test.

At the time of discharge, the patient's infection was effectively controlled, with a significant decrease in all inflammatory indicators and correction of anemia (Table I). During the postoperative follow-up, the abscess was gradually reduced (Figure 3). At one-year postoperative follow-up, the patient had a relatively satisfactory outcome, with no sensory or motor deficits of the lower extremities (Figure 3). Repeated CT showed bony fusion of the implanted iliac bone and complete disappearance of the abscess, despite the presence of secondary lumbar kyphosis with spinal stenosis (Figure 3).

VIDEO 1. An intraoperative view showing a large discharge of pus.

DISCUSSION

In this case, we report a patient of pyogenic spondylitis caused by SC infection with bilateral psoas major abscess and a large abscess in the left lumbodorsal section and chest wall. The identification of SC is sometimes difficult due to the prolonged anaerobic culture environment.[7] As a result, the general bacterial culture and tuberculosis detection of the intraoperative pus were negative, and no valid pathogenetic basis for pathological examination was found. In recent years, with the innovation of detection techniques, more and more technical tools have been applied to the study of the *Streptococcus anginosus* group.[7,8] Among them, NGS can sequence thousands to billions of DNA fragments simultaneously, making it possible to rapidly identify pathogens in culture-negative specimens and providing a new idea for pathogen detection.[9] Etiological identification in the case was achieved by NGS examination of intraoperative pus.

The case in this study was an infection after VA. Although its incidence is not high,[10] its consequences are catastrophic. Previous authors have suggested the use of bone cement mixed with tobramycin for VA,[11] while the others have suggested the use of intravenous prophylactic antibiotics in the perioperative period to prevent such complications in high-risk patients.[12] However, the clinical efficacy of these approaches has yet to be confirmed in a large sample of prospective comparative studies. Considering the patient's symptoms, history and treatment, we suspected that the VA failed due to the presence of ongoing infection. For the prevention of such complications, our experience was as follows: *(i)* If the levels of inflammation indicators were elevated before VA, careful screening for local or systemic infection and rheumatic diseases was required. Vertebral augmentation surgery was considered only after the infection was cured for two weeks. *(ii)* The fractured vertebra was examined for infectious disease, combined with magnetic resonance imaging (MRI) and CT. If the vertebral infection could not be ruled out, conservative treatment was recommended for two weeks, and the changes of the

disease were closely observed. Some authors have suggested routine radionuclide bone scintigraphy in patients with suspected OVF,[12] which has unique advantages in identifying infectious diseases, tumors, and fractures of the spine.[13] *(iii)* Strict asepsis was maintained during VA surgery.

Obtaining etiological evidence is essential for the treatment of infectious diseases. As our case was due to an SC infection, routine cultures were difficult to obtain the causative organism. Since the patient's infection was very severe on admission, we empirically administered strong anti-infective therapy and initially mistook the infection for *M. tuberculosis*. Fortunately, the anti-infective treatment was effective and septic shock did not develop. The literature reported that the Streptococcus anginosus group were susceptible to cefotaxime, vancomycin, and teicoplanin.[14] Clindamycin, doxycycline, amoxicillin, and metronidazole were used to treat SC infection.[15,16] Although SC is not a common pathogen causing abscesses, its pathogenicity still requires attention in clinical practice, particularly in immunocompromised patients.

In conclusion, pyogenic spondylitis due to SC infection after VA surgery is a serious complication. In addition to infectious disease consultation and routine etiological screening, NGS is important to identify infection with unknown pathogens. In patients with progressive neurological deficits, surgery combined with the use of sensitive antibiotics is one of the effective treatment options.

Patient Consent for Publication: A written informed consent was obtained from patient.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

Author Contributions: Idea/concept: L.L.; Design: L.H.; Control/supervision: M.H.; Data collection and/or processing: G.L., L.L.; Analysis and/or interpretation: L.L., L.H.; Literature review: L.L., L.H., M.H.; Writing the article: L.L., L.H.; Critical review: M.H.

Conflict of Interest: The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

Funding: The authors received no financial support for the research and/or authorship of this article.

REFERENCES

1. Koruga N, Rončević A, Soldo Koruga A, Sabadi D, Drenjančević D, Prica A, et al. Aggressive pyogenic spondylitis caused by S. constellatus: A case report. Diagnostics (Basel) 2022;12:2686. doi: 10.3390/diagnostics12112686.
2. Kobo O, Nikola S, Geffen Y, Paul M. The pyogenic potential of the different Streptococcus anginosus group bacterial species: Retrospective cohort study. Epidemiol Infect 2017;145:3065-9. doi: 10.1017/S0950268817001807.
3. Fu X, Li YM, Tian P, Xu GJ, Li ZJ. Comparison between unilateral curved and bilateral straight percutaneous vertebral augmentation in the treatment of osteoporotic vertebral compression fractures: A meta-analysis. Jt Dis Relat Surg 2023;34:237-44. doi: 10.52312/jdrs.2023.967.
4. Chen YJ, Chen HY, Hsu HC. Re: Infection after vertebroplasty or kyphoplasty. A series of nine cases and review of literature. Spine J 13:1809-17. Spine J 2014;14:1366. doi: 10.1016/j.spinee.2014.01.057.
5. Liao JC, Lai PL, Chen LH, Niu CC. Surgical outcomes of infectious spondylitis after vertebroplasty, and comparisons between pyogenic and tuberculosis. BMC Infect Dis 2018;18:555. doi: 10.1186/s12879-018-3486-x.
6. Atik OŞ. Which articles do the editors prefer to publish? Jt Dis Relat Surg 2022;33:1-2. doi: 10.52312/jdrs.2022.57903.
7. Walkty A, Embil JM, Nichol K, Karlowsky J. An unusual case of Streptococcus anginosus group pyomyositis diagnosed using direct 16S ribosomal DNA sequencing. Can J Infect Dis Med Microbiol 2014;25:32-4. doi: 10.1155/2014/170517.
8. Xiao K, Zhao D, Luo Y, Si Y. Streptococcus constellatus causing brain abscess identified by metagenomics next-generation sequencing. Surg Infect (Larchmt) 2022;23:207-8. doi: 10.1089/sur.2021.338.
9. Guo LY, Feng WY, Guo X, Liu B, Liu G, Dong J. The advantages of next-generation sequencing technology in the detection of different sources of abscess. J Infect 2019;78:75-86. doi: 10.1016/j.jinf.2018.08.002.
10. Park JW, Park SM, Lee HJ, Lee CK, Chang BS, Kim H. Infection following percutaneous vertebral augmentation with polymethylmethacrylate. Arch Osteoporos 2018;13:47. doi: 10.1007/s11657-018-0468-y.
11. Shapiro SA. Cranioplasty, vertebral body replacement, and spinal fusion with tobramycin-impregnated methylmethacrylate. Neurosurgery 1991;28:789-91. doi: 10.1097/00006123-199106000-00001.
12. Zhang S, Wang S, Wang Q, Yang J, Xu S. Debridement and corpectomy via single posterior approach to treat pyogenic spondylitis after vertebral augmentation. BMC Musculoskelet Disord 2021;22:591. doi: 10.1186/s12891-021-04478-0.
13. Okazaki T, Nakagawa H, Yagi K, Hayase H, Nagahiro S, Saito K. Bone scintigraphy for the diagnosis of the responsible level of osteoporotic vertebral compression fractures in percutaneous balloon kyphoplasty. Clin Neurol Neurosurg 2017;152:23-7. doi: 10.1016/j.clineuro.2016.11.007.
14. Jacobs JA, Stobberingh EE. In-vitro antimicrobial susceptibility of the "Streptococcus milleri' group (Streptococcus anginosus, Streptococcus constellatus and Streptococcus intermedius). J Antimicrob Chemother 1996;37:371-5. doi: 10.1093/jac/37.2.371.
15. Rams TE, Feik D, Mortensen JE, Degener JE, van Winkelhoff AJ. Antibiotic susceptibility of periodontal Streptococcus constellatus and Streptococcus intermedius clinical isolates. J Periodontol 2014;85:1792-8. doi: 10.1902/jop.2014.130291.
16. Rams TE, Degener JE, van Winkelhoff AJ. Antibiotic resistance in human chronic periodontitis microbiota. J Periodontol 2014;85:160-9. doi: 10.1902/jop.2013.130142.