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Upper cervical epidural abscess with emphasis on diabetes as a risk factor: a case report

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Abstract

Background An upper cervical spine epidural abscess (UCEA) is an epidural abscess that develops in the area between the occiput and the second cervical spine (axis). It is a rare diagnosis that carries the risk of instability of the atlantoaxial joint, and its management is not well-defined. It is known that the skin is the most common source of infection, and that diabetes mellitus (DM) is the most frequently reported risk factor. In this case, we present a patient diagnosed with UCEA, who achieved full neurological recovery postoperatively despite having neurological deficits for over five days prior to surgery.

Case presentation We report the case of a 56-year-old male patient with no history of any prior medical conditions, who presented with headache, neck pain, and weakness of the left side. The weakness started approximately three days prior to his presentation. His initial work up revealed hyperglycemia and elevated HbA1c of 86 mmol/mol (10%). Magnetic resonance imaging (MRI) of the cervical spine revealed spondylitis of the C2 spine with an abscess at the craniocervical junction. He underwent a two-staged surgical approach: decompression and stabilisation. The patient achieved full motor recovery approximately three months postoperatively.

Conclusions We recommend screening for DM when a spinal epidural abscess (SEA) is diagnosed without readily identifiable risk factors. The optimal management in most SEA cases is surgical, which is particularly true for UCEA because of the risk of atlantoaxial joint instability. Full neurological recovery is possible even when the patient has been having deficits for more than five days.

Keywords Spine surgery, Upper cervical epidural abscess, Epidural abscess, Diabetes mellitus

Background

An epidural abscess (EA) is an extradural collection of suppurative material and represents a neurosurgical emergency. It can be intracranial (IEA) or spinal (SEA). IEAs are less common because the dura is adhered to the periosteum in the cranium, making the epidural space a potential one. In contrast, in the spine, there is a fat-containing space above the dura, particularly in the posterior and lateral aspects. SEA is more common in the thoracolumbar spine, where the epidural space is larger, contains more infection-prone fatty tissue, and has a more extensive extradural venous plexus than the cervical spine [1, 2].

An upper cervical spine epidural abscess (UCEA) is an EA that develops in the area between the occiput and the second cervical spine (axis). This diagnosis is extremely rare with less than 50 reported cases since the beginning of the 18th century. UCEA is especially risky because it carries the risk of instability of the atlantoaxial joint [3].

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Due to its rarity, the management of UCEA is not well defined. In this case report, we present a patient with UCEA in whom a previously unknown diagnosis of diabetes mellitus (DM) was the only risk factor, and who achieved full neurological recovery postoperatively.

Case presentation

A 56-year-old male patient with no history of any prior medical conditions presented to the emergency department in November of 2022 with headache, neck pain, and weakness of the left side. He had experienced a headache and neck pain for over a month, during which time he was evaluated and given different diagnoses and treatments. He then developed weakness in the left side for three days before presentation. His complaints were initially associated with a fever that resolved spontaneously over a few days. A clinical review did not reveal any other symptoms. In particular, the patient did not have photophobia, diplopia, sore throat, ear pain or discharge, tooth pain, or neck rigidity.

He developed a mild COVID-19 infection five months before his presentation and received three doses of the vaccine. The patient had no relevant medical or surgical history. He was a non-smoker and had no history of alcohol or drug abuse. In addition, no recent history of antibiotic use or hospital admission was documented nor reported by the patient.

Examination upon presentation revealed a non-ambulatory, obese (body mass index 31.1 kg/m2) gentleman who was conscious and oriented with a Glasgow Coma Scale of 15/15, afebrile, and hypertensive (vital signs: blood pressure 153/83 mmHg, regular heart rate of 86 bpm, respiratory rate 20, and temperature 36.6 °C). Neurological examination revealed left-sided hemiparesis. On a 0–5 scale (using the Medical Research Council Manual Muscle Testing scale), the power of the left upper limb was 1/5 and that of the left lower limb was 2/5. Left-sided reflexes were diminished with an intact sensation. Examination of the cranial nerves revealed left-sided peripheral facial nerve palsy. The patient did not show signs of meningeal irritation, and systemic examination revealed no further findings. The patient had no evidence of skin or nail infection. Ear, nose, and throat examination was unremarkable.

Laboratory investigations revealed neutrophilic leukocytosis (white blood cell; WBC, 11.8) with a left-shift, elevated C-reactive protein (CRP; 30.8 mg/L), hyponatraemia (127 mmol/L), and fasting hyperglycaemia (174 mg/dL). His HbA1c was 86 mmol/mol (10%). Serum creatinine, potassium, and procalcitonin levels were within normal limits. Computed tomography (CT) of the cervical spine revealed bony erosions of the dens and atlantoaxial joint instability (Fig. 1). MRI revealed C2 spondylitis and an abscess at the craniocervical junction (Fig. 2).

The patient was diagnosed with upper cervical osteomyelitis and an abscess with cord compression. Furthermore, the patient was diagnosed with DM. He was kept on a cervical collar, and blood samples were collected for culture, which later returned negative. Intravenous antibiotics (meropenem and vancomycin) and insulin therapy (insulin glargine and sliding-scale therapy) were initiated.

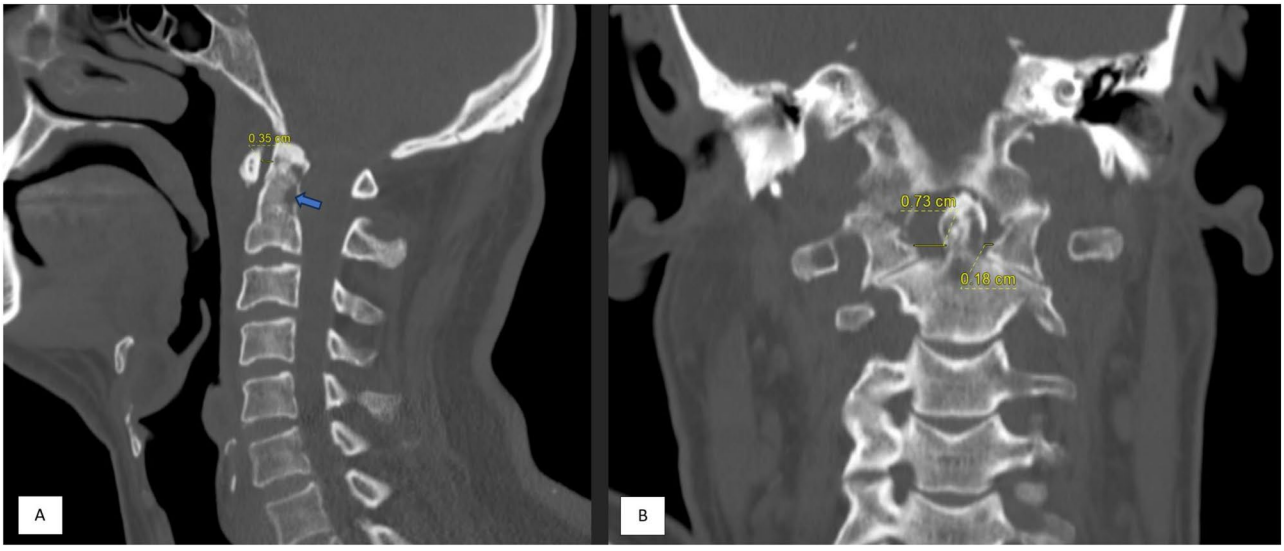


Fig. 1 shows CT images of the cervical spine, which are ideal to visualise bones, assess for atlantoaxial joint (AAJ) stability, and plan for operative fixation when needed. Figure 1**a** is a midline sagittal view of the cervical spine showing bony erosions (arrow) of the dens secondary to osteomyelitis. Also, the anterior atlantodental interval (ADI) is shown to be greater than 2 mm (0.35 cm), which is a sign of AAJ instability. Figure 1**b** is a coronal view of the cervical spine through the dens. It shows significant asymmetry between the right (0.73 cm), and left (0.18 cm) lateral ADI, which is another sign of AAJ instability

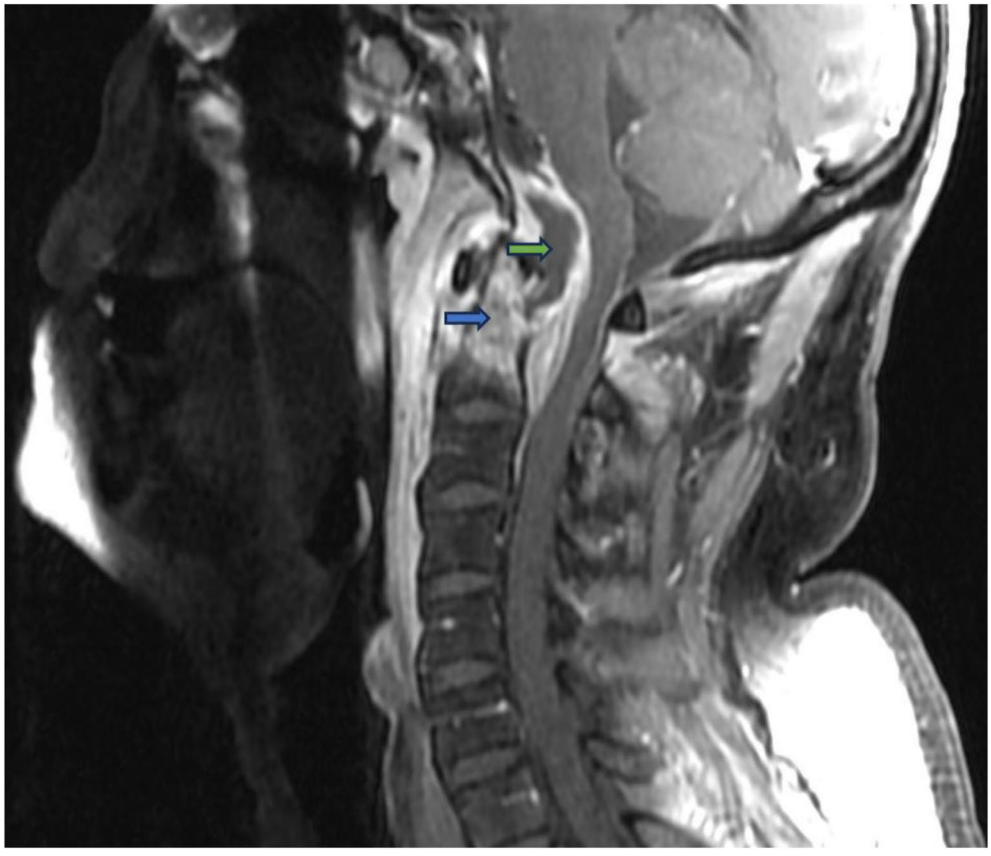
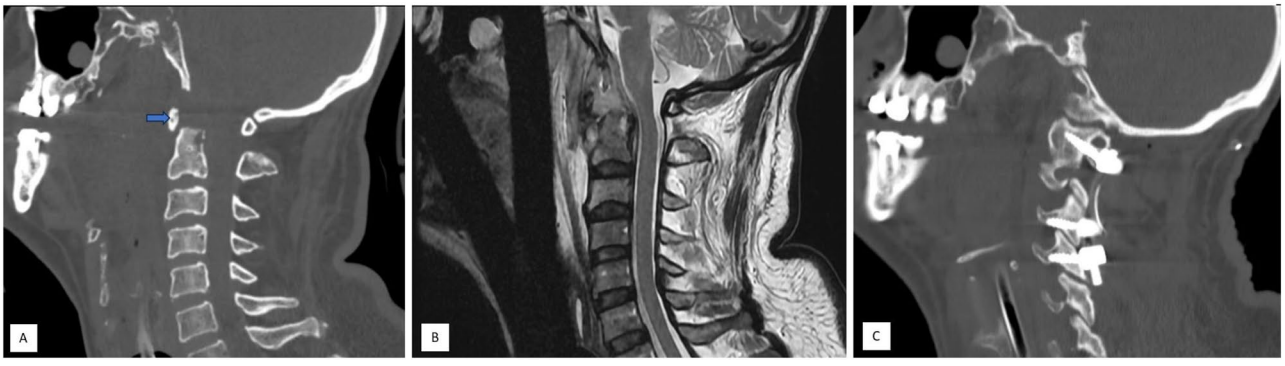


Fig. 2 is a T2-weighted MRI image with contrast enhancement showing a midline sagittal view of the cervical spine. There is hyperintense signal and positive contrast enhancement at the dens (blue arrow), which is indicative of an inflammatory process (osteomyelitis). It also shows s lesion (abscess) that is exerting a mass effect on the upper cervical cord and lower brain stem (green arrow)



Figs. 3 Shows post-operative imaging. Figures 3**A** and **B** are post-decompression (post-odontoidectomy and abscess evacuation). Figure 3**A** is a CT image showing a midline sagittal view that demonstrates the resected dens (odontoidectomy) behind the anterior arch of the atlas (arrow). Figure 3**B** is a T2-weighted MRI image showing a midline sagittal view that demonstrates abscess evacuation and cord decompression (compare with Fig. 2). Figure 3**C** is post-stabilisation. It shows a paramedian sagittal CT view demonstrating posterior cervical spine fixation of C1, C3, and C4 using screws fixed with rods

The patient was advised to undergo a two-staged surgical approach: decompression and stabilization. First, we performed transoral microsurgical odontoidectomy and abscess evacuation under radiographic guidance (day 0). Intraoperative samples were obtained and sent for culture and sensitivity testing. Postoperatively, the patient was shifted to the intensive care unit (ICU) sedated, and kept on a cervical collar. Intravenous dexamethasone was initiated. Postoperative CT and MRI confirmed abscess drainage and cord decompression (Fig. 3A and B). Nasogastric tube feeding was initiated after radiological assessment.

Five days after the first procedure, we performed posterior cervical spine fixation of C1, C3, and C4 (Fig. 3C).

Postoperatively, the patient was transferred to the ICU, and successfully extubated on the following day (day 6). After extubation, immediate improvement in the patient's facial palsy was noted over the next 48 h. On day 11, he started and tolerated oral feeding. Minimal improvement in hemiparesis was first noted on day 13. He was discharged in a stable condition on day 17.

The WBC count transiently increased to 22 postoperatively (on day 6) and returned to normal (WBC count of 9.8) few days later (on day 12). Also, CRP and serum sodium levels normalized postoperatively (on day 5). Cultures of the samples taken intraoperatively grew S. aureus, and the patient was switched to a single beta-lactam antibiotic therapy (cloxacillin) based on the results of sensitivity testing. In addition, his HbA1c level improved to 58 mmol/mol (7.5%), and the patient was kept on insulin glargine and oral therapy (dapagliflozin/metformin 10/1000 mg once daily).

In his first post-discharge follow-up (on day 41), facial palsy had completely resolved. His left-sided weakness improved to a power of 3–4/5 and he was able to stand with support. Approximately three months postoperatively (on day 99), the patient was able to stand and walk independently with minimal weakness in his left upper limb compared to the right upper limb. In all his subsequent follow-up visits, the patient showed no neurological deficits.

Discussion

An SEA is a collection of pus that enters the epidural space of the spine in one of three ways: haematogenous spread from a distant site of infection, extension from a nearby infection (e.g. vertebral osteomyelitis or psoas abscess), or direct inoculation (via traumatic injury or surgical or anaesthetic procedures). The most common source of infection is the skin (abscesses and furuncles). However, almost one-third of SEA cases have no identifiable source, which was the case in our patient [4]. The reported risk factors for SEA include any condition that could lead to bacteraemia (e.g. intravenous drug abuse, tattooing, infected catheters), any condition that leads to an immunocompromised state of health (e.g. DM, alcoholism, and acquired immunodeficiency), and any mechanism that could directly introduce bacteria into the epidural space (epidural/spinal anaesthesia, spinal surgery, and trauma) [5–7].

DM has consistently been the most frequently reported risk factor for SEA and was the only risk factor in our patient. In the largest meta-analysis of SEA, DM was reported in 15% of the patients [2]. DM is also associated with an increased risk of infection, and a retrospective study conducted in 2018 estimated that DM was responsible for 6% of hospitalisations and 12% of deaths related to infections [8]. DM as a risk factor for infection is not a new observation. It's known that DM compromises the immune system via multiple mechanisms at both the innate and adaptive levels [9].

Nevertheless, to the best of our knowledge, the pathophysiology of this observation, that is, the association between DM and SEA, has not been scrutinised in the literature. In addition to compromising the immune system, DM causes neuropathy, which impairs peripheral sensation and increases the risk of skin injury. Considering that the most common source of infection in SEA is the skin [2], diabetic neuropathy may be a mechanism by which DM increases the risk of SEA. However, this observation requires further study.

Notably, our patient had not been diagnosed with DM before he was diagnosed with UCEA. This raised the question of whether the patient should have been screened for DM. According to the American Diabetes Association (Standards of Care in Diabetes, 2023), screening for DM is recommended starting at 35 years of age, regardless of the risk factors [10]. This indicates that screening our patient and providing proper early treatment for DM could prevent the development of UCEA. In addition, considering that the most commonly reported risk factor for SEA is DM, we suggest screening for DM in any patient who develops this diagnosis without readily identifiable risk factors. Perhaps, screening for DM is warranted in any patient who develops an infection that is unusual in terms of presentation, site, or pathogen.

SEA are usually caused by pyogenic bacteria. S. aureus is the most common pathogen. It is isolated in approximately 73% of all SEA cases [2]. In a recent review of cervical SEA, S. aureus was isolated in 47.9% of cases [11]. This is also true for UCEA; a review on UCEA found S. aureus to be the most common causative pathogen (60% of cases), followed by Streptococcus pneumoniae (5% of cases); in 20% of cases, no pathogen could be identified [3].

Early diagnosis of SEA is paramount to prevent permanent neurological deficits and mortality. However, the condition's rarity and sometimes atypical presentation can make this difficult.

The incidence of SEA is 0.2–2 cases per 10,000 hospital admissions. Among SEA cases, 19% are cervical, and less than 50 cases of UCEA have been reported since the beginning of the 18th century [2, 3].

The classic triad of SEA includes pain, fever, and neurological deficits. Nevertheless, what's classic here isn't the usual. Local pain is the most common symptom and fever is initially absent in over one-third of patients. In addition, the average leukocyte count is 15,700, and normal counts are not uncommon [2].

The patterns and rates of symptom progression also vary. The usual progression of symptoms in SEA is local

pain with possible local tenderness and/or fever, followed by radicular symptoms due to nerve root irritation, then weakness, and paralysis [12].

In UCEA, the most common symptom is neck pain, which is probably a reason for the delayed diagnosis. Our patient experienced neck pain and headache for more than a month before any additional symptoms appeared. However, the occurrence of fever and neck pain should raise the possibility of cervical ESA. In this case, the patient developed a self-resolving fever that lasted for a few days, during which we believe that the diagnosis of cervical SEA should have been actively looked for. Nevertheless, only when the patient developed left-sided weakness, MRI was requested and a diagnosis of UCEA was made [3].

When cervical SEA is confirmed or suspected, detailed general, neurological, cranial nerves, and ENT (ear, nose, and throat) examinations need to be performed, keeping in mind that the absence of additional findings, such as neurological deficits, does not exclude the diagnosis.

They are performed to establish baseline function, assess possible cranial nerve involvement, and rule out possible sources of infection such as skin infection or tonsillitis [13].

The facial palsy in our patient was unlikely to be related to his diagnosis. It might be related to his COVID-19 infection and/or to a DM neuropathy, with the noted improvement was because of the use of dexamethasone rather than the surgery. However, this is a theory that we will not be able to confirm [14, 15].

Gadolinium-enhanced MRI is the diagnostic modality of choice for SEA, with a reported sensitivity and specificity of >90%. T2-weighted images are of particular value because an abscess shows signal uptake, differentiating it from the surrounding normal tissue. In this case, MRI revealed a lesion with a mass effect, a hyperintense signal, and positive enhancement with gadolinium contrast (Fig. 2). The radiological differential diagnoses could include infectious, neoplastic and rheumatological (e.g. rheumatoid arthritis) etiologies [16]. However, microbiological and/or histological assessment would provide the definitive diagnosis.

Additionally, brain imaging is an essential part of the workup once neurological deficits are present to rule out intracranial insults. For our patient, a brain MRI was done and found unremarkable. In addition to MRI, CT is needed for better visualisation of the vertebrae, assessing the need for stabilisation surgery, and for preoperative planning if surgical stabilisation is decided, as was the case in our patient (Figs. 1 and 3C) [17].

The management plan for UCEA can be either conservative or surgical. Conservative management consists of immobilisation and antibiotic therapy. Surgical management includes decompression (with or without stabilisation) and antibiotic therapy. Aspiration combined with antibiotic therapy has been previously described for SEA. However, there are no reports of UCEA cases being treated as such [3].

A surgical plan was adopted for most SEA cases. The surgical approach depends on the vertebral level and the location of the abscess (anterior vs. posterior). In this case, the patient underwent a two-stage surgical approach (decompression/stabilization approach): transoral microsurgical odontoidectomy and abscess evacuation (decompressive stage; Fig. 3A and B), followed by posterior cervical spine fixation (stabilisation stage; Fig. 3C).

Stabilisation was required here for two reasons. First, CT of the cervical spine demonstrated atlantoaxial joint instability. This was evident by assessing the atlantodental intervals (ADI). In this case, the anterior ADI was greater than 2 mm, and the lateral ADIs showed extreme asymmetry, and both these findings are indicative of instability [18, 19]. Second, the nature of the decompressive stage, specifically odontoidectomy, destabilised the occipito-antallo-axial complex by severing the stabilising cruciate and alar ligaments [20].

Nonsurgical treatment is recommended for patients who are unable to tolerate surgical stress. In addition, it can arguably be considered for an unwilling patient if the following three conditions are fulfilled: no neurological deficits have developed, a sample for culture and sensitivity testing is obtainable, and regular assessments are performed with MRI being readily available for urgent evaluation should the patient deteriorate. Nevertheless, this approach is risky for UCEA because of the serious risk of atlantoaxial joint instability. However, successful nonsurgical therapy is not unheard of, but only a few cases have been reported in the literature.

A study published in 1985 suggested a third scenario in which a nonsurgical approach is recommended, that is when a patient has been paralyzed for three days or more. In our case, the patient developed severe left-sided hemiparesis (power of the left upper limb 1/5 and left lower limb 2/5) for approximately 5–6 days. However, the patient achieved complete recovery postoperatively [21].

The outcomes of SEA depend on multiple factors. The most important factor is an early diagnosis. The mortality rate has been reported to be as high as 31%, and the outcome of cervical SEA is generally less favourable than that of thoracic or lumbar SEA [22, 23].

Conclusion

UCEA is a rare diagnosis. DM is its most frequently reported risk factor. In addition to the relatively immunosuppressed state of patients with diabetes, diabetic neuropathy could be a contributing factor, an observation that needs to be examined in future studies. We

recommend screening for DM when SEA is diagnosed without readily identifiable risk factors. The optimal management in most SEA cases is surgical, which is particularly true for UCEA because of the risk of atlantoaxial joint instability. Full neurological recovery is possible even when the patient has been having deficits for more than five days.

Abbreviations

ADI	Atlantodental intervals
CT	Computed tomography
CRP	C-reactive protein
DM	Diabetes mellitus
EA	Epidural abscess
IEA	Intracranial epidural abscess
MRI	Magnetic resonance imaging
SEA	Spinal epidural abscess
UCEA	Upper cervical spine epidural abscess
WBC	White blood cell

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Data availability
No datasets were generated or analysed during the current study.

Declarations

Ethics approval and consent to participate
Written informed consent was obtained from the patient.

Consent for publication
Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Competing interests
The authors declare no competing interests.

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References

1. Chow F. Brain and spinal epidural abscess. Continuum (Minneap Minn). 2018. https://doi.org/10.1212/CON.0000000000000649.
2. Reihsaus E, Waldbaur H, Seeling W. Spinal epidural abscess: a meta-analysis of 915 patients. Neurosurg Rev. 2000. https://doi.org/10.1007/pl00011954.
3. Al-Hourani K, Al-Aref R, Mesfin A. Upper Cervical Epidural Abscess in Clinical Practice: diagnosis and management. Global Spine J. 2016. https://doi.org/10.1055/s-0035-1565260.
4. Rubin M. Spinal Epidural Abscess. In: Merck Manuals Professional Edition. 2023. https://www.merckmanuals.com/professional/neurologic-disorders/spinal-cord-disorders/spinal-epidural-abscess. Accessed 23 Dec 2023.
5. Broner FA, Garland DE, Zigler JE. Spinal infections in the immunocompromised host. Orthop Clin North Am. 1996;27:37–46.
6. Sillevis Smitt P, Tsafka A, van den Bent M, de Bruin H, Hendriks W, Vecht C, Teng-van de Zande F. Spinal epidural abscess complicating chronic epidural analgesia in 11 cancer patients: clinical findings and magnetic resonance imaging. J Neurol. 1999. https://doi.org/10.1007/s004150050460.
7. Still JM, Abramson R, Law E. Development of an epidural abscess following staphylococcal septicemia in an acutely burned patient: case report. J Trauma. 1995;38:958–9.
8. Carey IM, Critchley JA, DeWilde S, Harris T, Hosking FJ, Cook DG. Risk of infection in type 1 and type 2 diabetes compared with the General Population: a matched cohort study. Diabetes Care. 2018. https://doi.org/10.2337/dc17-2131.
9. Zhou K, Lansang MC et al. Diabetes Mellitus and Infections. [Updated 2021 Mar 16]. In: Feingold KR, Anawalt B, Blackman MR,. Endotext [Internet]. South Dartmouth (MA): MDText.com, Inc.; 2000. https://www.ncbi.nlm.nih.gov/books/NBK569326/
10. American Diabetes Association. Standards of Care in Diabetes-2023 abridged for primary care providers. Clin Diabetes. 2022. https://doi.org/10.2337/cd23-as01.
11. Papadakis SA, Ampadiotaki MM, Pallis D, Tsivelekas K, Nikolakakos P, Agapitou L, Sapkas G. Cervical spinal epidural abscess: diagnosis, treatment, and outcomes: a Case Series and a literature review. J Clin Med. 2023. https://doi.org/10.3390/jcm12134509.
12. Eusner AP. Nontuberculous spinal epidural infections. N Engl J Med. 1948. https://doi.org/10.1056/NEJM194812022392301.
13. Azizi SA, Fayad PB, Fulbright R, Giroux ML, Waxman SG. Clivus and cervical spinal osteomyelitis with epidural abscess presenting with multiple cranial neuropathies. Clin Neurol Neurosurg. 1995. https://doi.org/10.1016/0303-8467(95)00036-j.
14. Lima MA, Silva MTT, Soares CN, Coutinho R, Oliveira HS, Afonso L, et al. Peripheral facial nerve palsy associated with COVID-19. J Neurovirol. 2020. https://doi.org/10.1007/s13365-020-00912-6.
15. Stamatiou I, Papachristou S, Papanas N. Diabetes Mellitus and Bell's Palsy. Curr Diabetes Rev. 2023;19(1):e080322201913. https://doi.org/10.2174/1573399818666220308161023. PMID: 35260056.
16. Pettersson H, Larsson EM, Holtås S, et al. MR imaging of the cervical spine in rheumatoid arthritis. AJNR Am J Neuroradiol. 1988;9(3):573–7.
17. Sendi P, Bregenzer T, Zimmerli W. Spinal epidural abscess in clinical practice. QJM: Monthly J Association Physicians. 2008. https://doi.org/10.1093/qjmed/hcm100.
18. Rojas CA, Bertozzi JC, Martinez CR, Whitlow J. Reassessment of the craniocervical junction: normal values on CT. AJNR. Am J Neuroradiol. 2007. https://doi.org/10.3174/ajnr.A0660.
19. Chen Y, Zhuang Z, Qi W, Yang H, Chen Z, Wang X, Kong K. A three-dimensional study of the atlantodental interval in a normal Chinese population using reformatted computed tomography. Surg Radiol Anat. 2011. https://doi.org/10.1007/s00276-011-0817-7.
20. Goel A. Indicators of atlantoaxial instability. J Craniovertebr Junction Spine. 2021. https://doi.org/10.4103/jcvjs.jcvjs_55_21.
21. Leys D, Lesoin F, Viaud C, Pasquier F, Rousseaux M, Jomin M, Petit H. Decreased morbidity from acute bacterial spinal epidural abscesses using computed tomography and nonsurgical treatment in selected patients. Ann Neurol. 1985. https://doi.org/10.1002/ana.410170408.
22. Maslen DR, Jones SR, Crislip MA, Bracis R, Dworkin RJ, Flemming JE. Spinal epidural abscess. Optimizing patient care. Arch Intern Med. 1993;153:1713–21.
23. Epstein N. Diagnosis, and treatment of cervical epidural abscess and/or cervical vertebral osteomyelitis with or without Retropharyngeal Abscess; a review. Surg Neurol Int. 2020. https://doi.org/10.25259/SNI_294_2020.

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