DOI: https://doi.org/10.17816/DD430128

Хронический пищеводный свищ как редкая причина вторичного остеомиелита грудного отдела позвоночника

В.А. Заря, П.В. Гаврилов, М.Е. Макогонова, А.Р. Козак, А.А. Вишневский

Санкт-Петербургский научно-исследовательский институт фтизиопульмонологии, Санкт-Петербург, Российская Федерация

АННОТАЦИЯ

Инфекционные заболевания позвоночника представляют собой воспалительные деструктивные заболевания органа и его структурных элементов в результате инфицирования гематогенным, лимфогенным или контактным путём, в том числе могут являться осложнением хирургического вмешательства. При постановке диагноза крайне важно оценивать в совокупности анамнез, клиническую картину, а также данные лабораторных исследований и лучевой диагностики.

В работе представлен клинический случай вторично развившегося спондилита позвонков ThVII–ThVIII вследствие пищеводного свища. При первичной диагностике спондилит связали со спинальной анестезией, которая проводилась за 6 месяцев до начала заболевания, так как имел место свищевой дефект на коже в поясничной области. По этому поводу трижды проводились оперативные вмешательства в хирургическом стационаре по месту жительства. Данные эндоскопического исследования и жалобы пациентки на связь между приёмами пищи, появлением болей и характером отделяемого из свища не были приняты врачами изначально во внимание. С помощью дополнительного обследования, включающего компьютерную томографию пищевода с пероральным контрастированием и компьютерно-томографическую фистулографию, был установлен основной диагноз «Свищ пищевода», а спондилит грудного отдела позвоночника оказался лишь вторичным осложнением.

Таким образом, окончательный диагноз при болях в спине, обусловленных не только инфицированием, но и являющихся осложнением хирургического вмешательства, должен формулироваться после проведения дифференциальной диагностики с альтернативными заболеваниями позвоночника.

Ключевые слова: остеомиелит; спондилит; свищ пищевода.

Как цитировать:
Заря В.А., Гаврилов П.В., Макогонова М.Е., Козак А.Р., Вишневский А.А. Хронический пищеводный свищ как редкая причина вторичного остеомиелита грудного отдела позвоночника // *Digital Diagnostics.* 2023. Т. 4, № 3. С. 403–410. DOI: https://doi.org/10.17816/DD430128

Рукопись получена: 16.05.2023 **Рукопись одобрена:** 22.08.2023 **Опубликована:** 30.08.2023

DOI: https://doi.org/10.17816/DD430128

Chronic esophageal fistula as a rare cause of secondary osteomyelitis of the thoracic spine

Valeriya A. Zarya, Pavel V. Gavrilov, Marina E. Makogonova, Andrey R. Kozak, Arkadiy A. Vishnevskiy

Saint-Petersburg State Research Institute of Phthisiopulmonology, Saint Petersburg, Russian Federation

ABSTRACT

Infectious diseases affecting the spine are inflammatory destructive diseases that involved the organ and its structural elements as a result of infection by hematogenic, lymphogenic, or contact pathways, including may be a complication of surgical intervention. In arriving at an accurate diagnosis, it is extremely important to evaluate the anamnesis, the clinical picture, as well as the data of laboratory studies and radiation diagnostics in the aggregate.

This article presents a clinical case with the development of secondary ThVII–ThVIII vertebral spondylitis due to esophageal fistula. At the initial diagnosis, spondylitis was associated with spinal anesthesia performed six months prior to onset of the disease, as there was a fistulous defect on the skin in the lumbar region. Consequently, surgical interventions were performed three times in a surgical hospital at the place of residence. The data from the endoscopic examination, as well as the patient's complaints regarding the relationship between meals, the appearance of pain, and the nature of the discharge from the fistula were not taken into account by doctors initially. With the help of an additional examination, including computed tomography of the esophagus with oral contrast and computed tomography fistulography, the main diagnosis was esophageal fistula. Thoracic spondylitis was only a secondary complication.

Thus, the final diagnosis of back pain and fistula in the lumbar region should be formulated after differential diagnosis with alternative diseases of the spine.

Keywords: osteomyelitis; spondylitis; esophageal fistula.

To cite this article:

Zarya VA, Gavrilov PV, Makogonova ME, Kozak AR, Vishnevskiy AA. Chronic esophageal fistula as a rare cause of secondary osteomyelitis of the thoracic spine. *Digital Diagnostics.* 2023;4(3):403–410. DOI: https://doi.org/10.17816/DD430128

Received: 16.05.2023 **Accepted:** 22.08.2023 **Published:** 30.08.2023

DOI: https://doi.org/10.17816/DD430128

慢性食管瘘作为继发性胸椎骨髓炎的罕见病因

Valeriya A. Zarya, Pavel V. Gavrilov, Marina E. Makogonova,
Andrey R. Kozak, Arkadiy A. Vishnevskiy

Saint-Petersburg State Research Institute of Phthisiopulmonology, Saint Petersburg, Russian Federation

简评

脊柱感染性疾病是由于血源性、淋巴原性或接触性感染（包括手术并发症）引起的器官及其结构元素的炎症性破坏性疾病。在进行诊断时，对病史、临床表现以及实验室检查和放射诊断数据进行评估极为重要。

本文介绍一例因食管瘘引起的ThVII-ThVIII椎骨继发性脊椎炎的临床病例。在最初诊断时，医生认为脊椎炎与脊髓麻醉有关，而脊髓麻醉是在发病前6个月进行的，因为腰部皮肤上有瘘管缺损。这次在居住地的外科医院进行了三次手术治疗。医生最初并没有考虑到内窥镜检查结果以及病人关于进食、疼痛和瘘管分泌物性质之间关系的主诉。在额外检查的帮助下，包括口服造影剂的食道CT扫描和瘘管CT造影，确定了食管瘘的主要诊断，而胸椎脊椎炎只是次要并发症。

因此，在存在背痛的原因不仅是感染，还可能是手术治疗的并发症的情况下，最终诊断应该是在与其他脊柱疾病进行鉴别诊断后再做出的。

关键词：骨髓炎；脊椎炎；食管瘘。

引用本文：

Zarya VA, Gavrilov PV, Makogonova ME, Kozak AR, Vishnevskiy AA. 慢性食管瘘作为继发性胸椎骨髓炎的罕见病因. *Digital Diagnostics*. 2023;4(3):403–410. DOI: https://doi.org/10.17816/DD430128

收到：16.05.2023 **接受：**22.08.2023 **发布日期：**30.08.2023

BACKGROUND

Spinal infections are inflammatory destructive disorders of the spine and its structural components (vertebral bodies, intervertebral discs, ligaments, and intervertebral joints). They can be caused by any bacterial agent due to a hematogenous, lymphogenous, or contact infection, or they can be surgical complications (e.g., postoperative and iatrogenic infections) [1].

Spondylitis can be infectious or non-infectious (aseptic). Infectious spondylitis is caused by bacterial, fungal, or parasitic invasions. Spondylitis can cause hematogenous (septic) or contact infections [2–4], as well as postoperative (iatrogenic) complications [5–8]. Some authors reported that spondylitis caused by esophageal perforation can spread posteriorly, resulting in secondary damage to cervical or thoracic vertebrae. For example, Janssen et al. [9] presented cervical and thoracic spondylodiscitis cases caused by esophageal perforation in patients with a history of esophageal cancer who underwent combination therapy. Some of these patients had a recurrence of esophageal fistula after previous nonsurgical and surgical treatment. The authors also reported a female patient who ingested and independently retrieved a toothpick 2 months before the onset of spondylitis, which was accompanied by an epidural and paravertebral abscess. According to Fonga-Djimi et al. [10], an esophageal perforation caused by a foreign body (a toy car wheel) was worsened by mediastinitis and spondylodiscitis. Wadie et al. [11] presented a clinical case of a child with cervical spondylodiscitis and paravertebral soft tissue phlegmon caused by pin ingestion, which resulted in perforation of the posterior pharynx at the level of the laryngeal aperture. Van Ooij et al. [12] described a female patient with cervical spondylitis caused by a fish bone trapped in the esophagus.

When an infection spreads to the chest, it causes empyema, pericarditis, and mediastinitis. As a result of spondylitis, empyema and pericarditis may reoccur. Infections of the anterior thoracolumbar and lumbar spines can result in subdiaphragmatic abscess, peritonitis, and psoas abscess.

Appropriate treatment of infectious spondylitis requires an accurate diagnosis of the etiology and pathogen. The use of X-ray diagnostics is important in the diagnosis of spondylitis. However, X-ray results do not guarantee that the nature and etiology of the infection are accurately identified. Thus, it is necessary to consider the medical history, clinical presentation, laboratory findings, and X-ray findings when diagnosing.

This study presents a rare secondary thoracic spine lesion caused by a chronic esophageal fistula.

CASE REPORT

Patient

The patient is a 42-year-old female. Back pain and lumbar fistula complaints initially appeared 3 yr ago. According to the medical history, the patient received spinal anesthesia for a cesarean section 6 months prior. The patient experienced fistula relapses approximately three to five times a year. At the presentation, a magnetic resonance imaging (MRI) revealed bony and fibrous ankylosis ThVII–ThVIII (Figure 1), which could indicate both spondylitis in remission and contact spinal infection.

At different outpatient facilities, the patient had surgery (fistulotomy and abscessotomy) three times. The surgical specimen contained hemolytic streptococcus susceptible to amoxiclav, ampicillin, cefotaxime, vancomycin, doxycycline, and meropenem. The patient received targeted antibiotic medication with no effect. Before admission to the

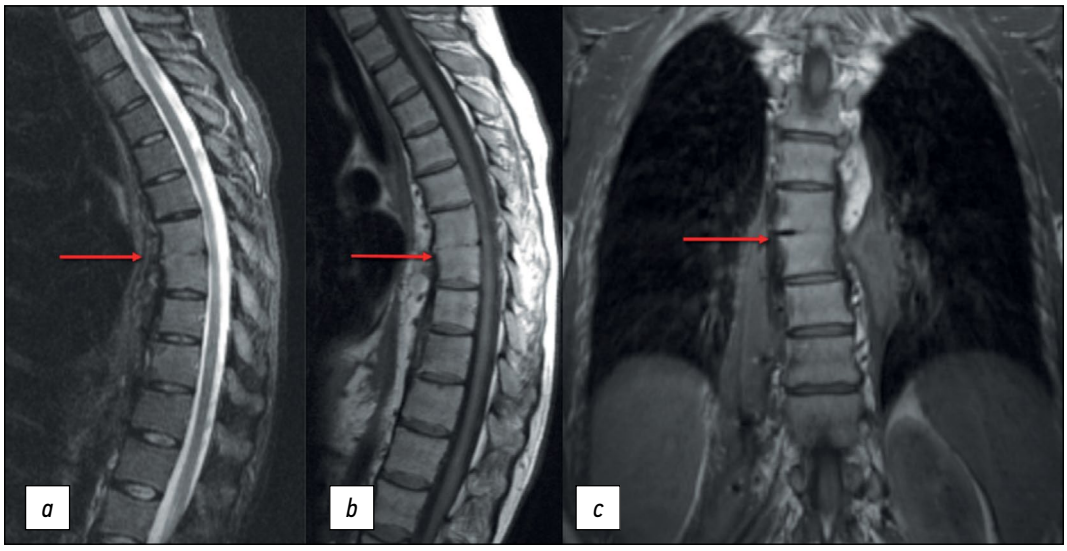

Fig. 1. Thoracic spine MRI: (*a*) STIR mode, sagittal plane; (*b*) T1WI mode, sagittal plane; and (*c*) T1WI mode, coronal plane. The arrows indicate bony and fibrous ankylosis ThVII–ThVIII.

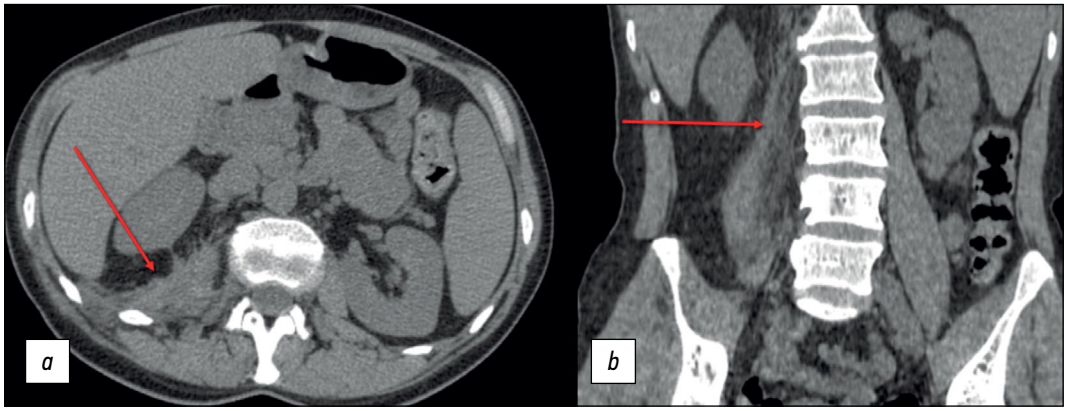

Fig. 2. Lumbar spine MRI: (*a*) soft tissue mode, axial plane, and (*b*) soft tissue mode, coronal plane. The arrows indicate the right psoas muscle abscess.

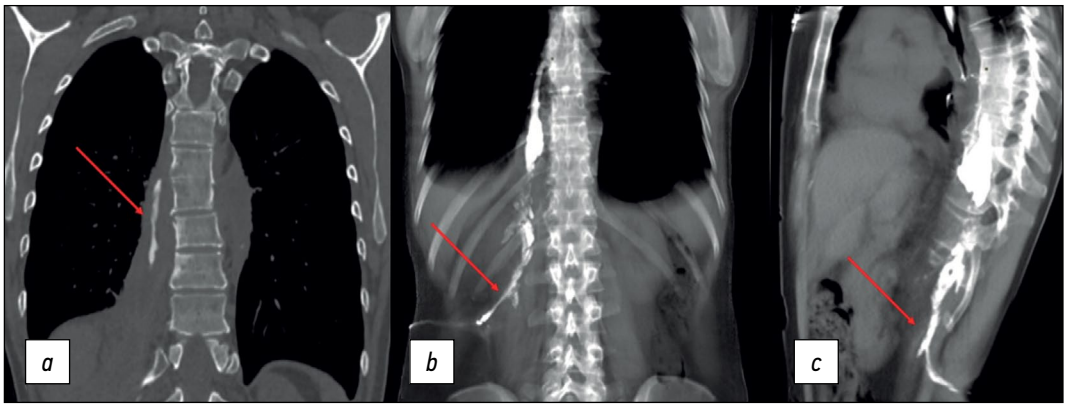

Fig. 3. (*a*) CT esophagography with oral contrast, coronal plane and (*b* and *c*) CT fistulography, multiplanar reconstruction (MPR), coronal and sagittal plane. The arrows indicate the fistula tract from the esophagus to the right paravertebral space, from the ThVII to the ThX level.

St. Petersburg Research Institute of Phthisiopulmonology, a computed tomography (CT) revealed symptoms of prior spondylitis ThVII–ThVIII and a right-sided psoas abscess (Figure 2).

During the additional history taking, the patient reported an association between his food intake (mostly liquid food), the onset of pain, and the type of discharge from the lumbar fistula tract. Due to these symptoms, a CT esophagography with oral contrast revealed a fistula tract at the ThVII level extending from the esophagus to the right paravertebral space up to the ThX level and loculated empyema on the right. An additional CT fistulography was performed to determine the length of the fistula tract, with a contract solution (iopromide 370) injected into the lumbar fistula located on the right at the LIII level. The CT fistulography revealed a fistula tract extending from the right psoas major muscle to the ThVII level. The fistula tract linked with the esophageal lumen at the same level, where the contrast agent was visible (Figure 3).

A follow-up thoracic and lumbar spine MRI performed in the hospital revealed stable changes in the ThVII–ThVIII vertebral bodies and bilateral paravertebral abscesses (Figure 4).

All laboratory findings were unremarkable, except for the erythrocyte sedimentation rate (30 mm/h). To determine further treatment strategy, an esophagogastroduodenoscopy (EGD) was performed, which revealed a fistula tract along the left posterior wall, up to 0.5 cm long, bordered with esophageal epithelium and covered by granulation tissue. The diagnosis of an esophageal fistula in the middle third was confirmed. A post hoc analysis of earlier EGD findings showed that the observed esophageal defect had previously been identified and classified as a diverticulum. However, this information was not taken into consideration in other outpatient facilities.

Thus, a multidisciplinary team made the following diagnosis based on the clinical and X-ray findings: esophagopleural fistula of the lower third of the esophagus. On the right, there is chronic loculated empyema. Chronic contiguous osteomyelitis ThVII–ThVIII with a fistula. The patient was sent to the thoracic surgery department for the excision of an esophageal fistula.

DISCUSSION

Spondylitis infections can be caused by various factors, including dental caries, ENT infections, phlegm,

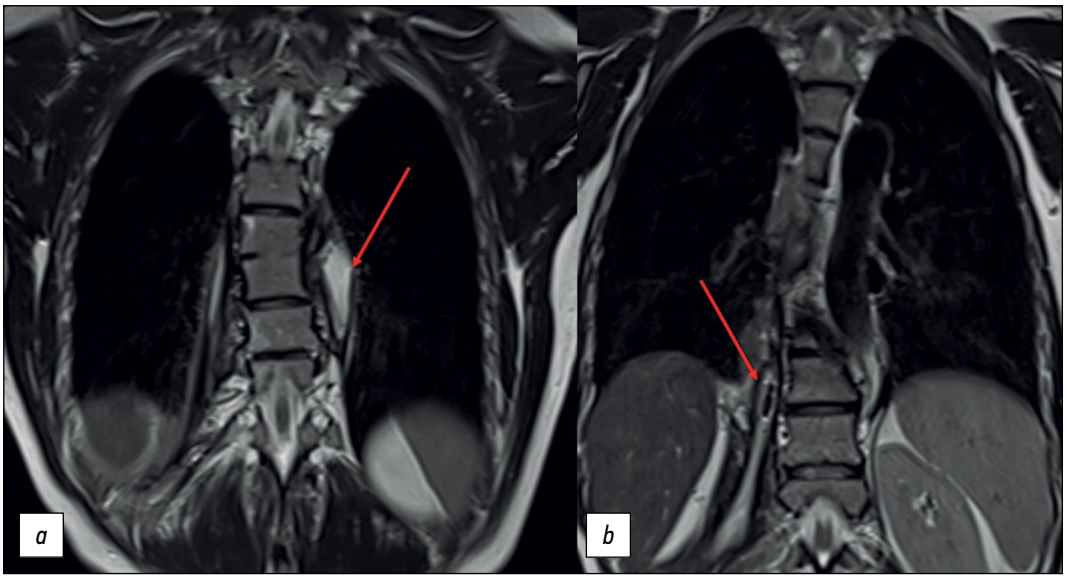

Fig. 4. Thoracic and lumbar spine MRI: T2WI mode, coronal plane. Bony and fibrous ankylosis ThVII–ThVIII and paravertebral abscesses (arrows) on the left (*a*), with air bubbles on the right (*b*).

and endocarditis. Structural damage to the spine can be caused by hematogenous or contact infections, including penetrating injuries, such as iatrogenic injury. Given the available data, the following questions arise: why was the therapy ineffective, and was the primary cause spondylitis or esophageal fistula?

A history of epidural anesthesia is the main reason supporting infectious spondylitis as the primary process. In such cases, epidural anesthesia is administered at the LIII–LV level. The affected vertebrae in our case are significantly higher. Another factor suggesting spondylitis as the causative cause is a paravertebral abscess on the left (contralateral to the esophageal fistula). Simultaneously, there was no explanation for the severe inflammatory changes in paravertebral tissues. This clinical presentation of spondylitis is not typical. In general, vertebrae are more affected than soft tissues around them. Furthermore, the patient denied any esophageal injury that may lead to a fistula.

Another factor suggesting an esophageal origin of the process was a fistulous contact between the paravertebral abscess and the esophagus (fistula tract diameter up to 5 mm). Endoscopy revealed an esophageal diverticulum and an intact esophageal wall (which was not involved in the inflammatory process); the fistula was bordered by esophageal epithelium. Moreover, the patient associated the pain syndrome with food intake, and the discharge from the lumbar fistula tract resembled previously ingested food or drink.

The long-term disease makes determining the primary process impossible. Considering all available data, esophageal perforation is proposed as the leading cause, followed by spondylitis and paravertebral abscess. Despite the patient denying any esophageal injury, the endoscopic results are most consistent with an esophageal injury caused by an ingested foreign body (such as a fish bone).

CONCLUSION

The cause of the pathological process (esophageal fistula) was unknown, and the patient was treated symptomatically (surgery and antibacterial medication) while in the hospital.

The availability of several modern imaging and surgical procedures does not eliminate the need for a thorough history taking and clinical presentation examination.

Previous diagnoses should be reviewed for compliance with diagnostic criteria in cases when appropriate therapy is ineffective.

Differential diagnosis is critical for systematically assessing possible alternative diagnoses before concluding.

The common logical fallacy of *post hoc ergo propter hoc* (a false conclusion that confuses co-occurrence with causality) should also be considered.

ADDITIONAL INFORMATION

Funding source. This article was not supported by any external sources of funding.

Competing interests. The authors declare that they have no competing interests.

Authors' contribution. All authors made a substantial contribution to the conception of the work, acquisition, analysis, interpretation of data for the work, drafting and revising the work, final approval of the version to be published and agree to be accountable for all aspects of the work. V.A. Zarya — collecting material, writing the article; P.V. Gavrilov — concept and design of the work, final editing; M.E. Makogonova — analysis of the data obtained, writing the text; A.R. Kozak — collecting material, writing the article; A.A. Vishnevskiy — concept and design of the work, final editing.

Consent for publication. Written consent was obtained from the patient for publication of relevant medical information and all of accompanying images within the manuscript in Digital Diagnostics Journal.

REFERENCES

1. Mushkin AYu, Vishnevsky AA. Clinical recommendations for the diagnosis of infectious spondylitis (draft for discussion). *Medical Alliance*. 2018;(3):65–74. (In Russ).
2. Fowler VG, Justice A, Moore C, et al. Risk factors for hematogenous complications of intravascular catheter-associated Staphylococcus aureus bacteremia. *Clin Infect Dis. 2005*;40(5):695–703. doi: 10.1086/427806
3. Lu YA, Hsu HH, Kao HK, et al. Infective spondylodiscitis in patients on maintenance hemodialysis: A case series. *Ren Fail*. 2017;39(1):179–186. doi: 10.1080/0886022X.2016.1256313
4. Choi KB, Lee CD, Lee SH. Pyogenic spondylodiscitis after percutaneous endoscopic lumbar discectomy. *J Korean Neurosurg Soc*. 2010;48(5):455–460. doi: 10.3340/jkns.2010.48.5.455
5. Hsieh MK, Chen LH, Niu CC, et al. Postoperative anterior spondylodiscitis after posterior pedicle screw instrumentation. *Spine J*. 2011;11(1):24–29. doi: 10.1016/j.spinee.2010.10.021
6. Hanci M, Toprak M, Sarioğlu AC, et al. Oesophageal perforation subsequent to anterior cervical spine screw/plate fixation. *Paraplegia*. 1995;33(10):606–609. doi: 10.1038/sc.1995.128
7. Orlando ER, Caroli E, Ferrante L. Management of the cervical esophagus and hypofarinx perforations complicating anterior cervical spine surgery. *Spine*. 2003;28:E290–E295. doi: 10.1097/00007632-200308010-00023
8. Pompili A, Canitano S, Caroli F, et al. Asymptomatic esophageal perforation caused by late screw migration after anterior cervical plating: Report of a case and review of relevant literature. *Spine*. 2002;27:E499–E502. doi: 10.1097/00007632-200212010-00016
9. Janssen I, Shiban E, Rienmüller A, et al. Treatment considerations for cervical and cervicothoracic spondylodiscitis associated with esophageal fistula due to cancer history or accidental injury: A 9-patient case series. *Acta Neurochir (Wien)*. 2019;161(9):1877–1886. doi: 10.1007/s00701-019-03985-3
10. Fonga-Djimi H, Leclerc F, Martinot A, et al. Spondylodiscitis and mediastinitis after esophageal perforation owing to a swallowed radiolucent foreign body. *J Pediatr Surg*. 1996;31(5):698–700. doi: 10.1016/s0022-3468(96)90677-6
11. Wadie GM, Konefal SH, Dias MA, McLaughlin MR. Cervical spondylodiscitis from an ingested pin: A case report. *J Pediatr Surg*. 2005;40(3):593–596. doi: 10.1016/j.jpedsurg.2004.11.023
12. Van Ooij A, Manni JJ, Beuls EA, Walenkamp GH. Cervical spondylodiscitis after removal of a fishbone. A case report. *Spine (Phila Pa 1976)*. 1999;24(6):574–577. doi: 10.1097/00007632-199903150-00015

СПИСОК ЛИТЕРАТУРЫ

1. Мушкин А.Ю., Вишневский А.А. Клинические рекомендации по диагностике инфекционных спондилитов (проект для обсуждения) // Медицинский альянс. 2018. № 3. С. 65–74.
2. Fowler V.G., Justice A., Moore C., et al. Risk factors for hematogenous complications of intravascular catheter-associated Staphylococcus aureus bacteremia // Clin Infect Dis. 2005. Vol. 40, N 5. P. 695–703. doi: 10.1086/427806
3. Lu Y.A., Hsu H.H., Kao H.K., et al. Infective spondylodiscitis in patients on maintenance hemodialysis: A case series // Ren Fail. 2017. Vol. 39, N 1. P. 179–186. doi: 10.1080/0886022X.2016.1256313
4. Choi K.B., Lee C.D., Lee S.H. Pyogenic spondylodiscitis after percutaneous endoscopic lumbar discectomy // J Korean Neurosurg Soc. 2010. Vol. 48, N 5. P. 455–460. doi: 10.3340/jkns.2010.48.5.455
5. Hsieh M.K., Chen L.H., Niu C.C., et al. Postoperative anterior spondylodiscitis after posterior pedicle screw instrumentation // Spine J. 2011. Vol. 11, N 1. P. 24–29. doi: 10.1016/j.spinee.2010.10.021
6. Hanci M., Toprak M., Sarioğlu A.C., et al. Oesophageal perforation subsequent to anterior cervical spine screw/plate fixation // Paraplegia. 1995. Vol. 33, N 10. P. 606–609. doi: 10.1038/sc.1995.128
7. Orlando E.R., Caroli E., Ferrante L. Management of the cervical esophagus and hypopharynx perforations complicating anterior cervical spine surgery // Spine. 2003. Vol. 28. P. E290–E295. doi: 10.1097/00007632-200308010-00023
8. Pompili A., Canitano S., Caroli F., et al. Asymptomatic esophageal perforation caused by late screw migration after anterior cervical plating: Report of a case and review of relevant literature // Spine. 2002. Vol. 27. P. E499–E502. doi: 10.1097/00007632-200212010-00016
9. Janssen I., Shiban E., Rienmüller A., et al. Treatment considerations for cervical and cervicothoracic spondylodiscitis associated with esophageal fistula due to cancer history or accidental injury: A 9-patient case series // Acta Neurochir (Wien). 2019. Vol. 161, N 9. P. 1877–1886. doi: 10.1007/s00701-019-03985-3
10. Fonga-Djimi H., Leclerc F., Martinot A., et al. Spondylodiscitis and mediastinitis after esophageal perforation owing to a swallowed radiolucent foreign body // J Pediatr Surg. 1996. Vol. 31, N 5. P. 698–700. doi: 10.1016/s0022-3468(96)90677-6
11. Wadie G.M., Konefal S.H., Dias M.A., McLaughlin M.R. Cervical spondylodiscitis from an ingested pin: A case report // J Pediatr Surg. 2005. Vol. 40, N 3. P. 593–596. doi: 10.1016/j.jpedsurg.2004.11.023
12. Van Ooij A., Manni J.J., Beuls E.A., Walenkamp G.H. Cervical spondylodiscitis after removal of a fishbone. A case report // Spine (Phila Pa 1976). 1999. Vol. 24, N 6. P. 574–577. doi: 10.1097/00007632-199903150-00015

AUTHORS' INFO

*** Pavel V. Gavrilov,** MD, Cand. Sci. (Med.);
address: 2-4 Ligovskiy avenue, 191036 Saint Petersburg, Russia;
ORCID: 0000-0003-3251-4084;
eLibrary SPIN: 7824-5374;
e-mail: spbniifrentgen@mail.ru

ОБ АВТОРАХ

*** Гаврилов Павел Владимирович,** канд. мед. наук;
адрес: Россия, 191036, Санкт-Петербург, Лиговский пр-т, д. 2-4;
ORCID: 0000-0003-3251-4084;
eLibrary SPIN: 7824-5374;
e-mail: spbniifrentgen@mail.ru

* Corresponding author / Автор, ответственный за переписку

Valeriya A. Zarya;
ORCID: 0000-0001-7956-3719;
e-mail: zariandra@mail.ru

Marina E. Makogonova, MD, Cand. Sci. (Med.);
ORCID: 0000-0001-6760-2426;
eLibrary SPIN: 6342-8967;
e-mail: MakogonovaME@gmail.com

Andrey R. Kozak, MD, Cand. Sci. (Med.);
ORCID: 0000-0002-3192-1430;
e-mail: andrkozak@mail.ru

Arkadiy A. Vishnevskiy, MD, Dr. Sci. (Med.);
ORCID: 0000-0002-9186-6461;
eLibrary SPIN: 4918-1046;
e-mail: vichnevsky@mail.ru

Заря Валерия Алексеевна;
ORCID: 0000-0001-7956-3719;
e-mail: zariandra@mail.ru

Макогонова Марина Евгеньевна, канд. мед. наук;
ORCID: 0000-0001-6760-2426;
eLibrary SPIN: 6342-8967;
e-mail: MakogonovaME@gmail.com

Козак Андрей Романович, канд. мед. наук;
ORCID: 0000-0002-3192-1430;
e-mail: andrkozak@mail.ru

Вишневский Аркадий Анатольевич, д-р мед. наук;
ORCID: 0000-0002-9186-6461;
eLibrary SPIN: 4918-1046;
e-mail: vichnevsky@mail.ru