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Multiple Noncontiguous Spondylodiscitis Following Mastectomy Secondary to Breast Carcinoma: Case Report and Literature Review

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ABSTRACT

Background Data: Multiple noncontiguous spontaneous pyogenic spondylodiscitis is rare and has only been explained in the literature by case reports and case series.

Purpose: We present a case report of multiple noncontiguous spontaneous spondylodiscitis caused by *E. coli* involving the cervical and lumbar spine following a mastectomy for breast cancer. We will explain the difficulties in diagnosis, treatment, and follow-up with the concomitant.

Study Design: A case report and literature review.

Case Report: A middle-aged patient with a history of breast carcinoma underwent surgery. Two weeks after mastectomy, the patient developed severe cervical and lumbar spine pain and a low-grade fever (37.5–38.3 °C). Moreover, neurological examination revealed a right-sided antalgic gait, right-sided weakness, and a positive straight leg raising test. Upon presentation, elevated C-reactive protein (CRP) and white blood cells (WBC) were noted. Magnetic resonance imaging (MRI) showed consistent spondylodiscitis at C5-C6 and L3-L4 levels with stenotic features at L4-L5 levels. Surgical treatment of the lumbar region via posterior spinal instrumentation from L3 to L5 levels and decompression was done with a biopsy. Erythrocyte sedimentation rate (ESR) CRP titers were also performed for the follow-up plan, which showed a reduction in 3, 6, and 12 weeks postoperatively. Conservative treatment of the cervical region was undertaken with a complete cure.

Conclusion: Multiple noncontiguous spondylodiscitis after nonspinal surgery is a relatively rare complication requiring a high suspicion index. Surgery is recommended in case of failure of conservative measures, neurological deficit, or mechanical instability as in this case. Furthermore, both clinical examination and blood tests should be used to assess the treatment outcomes. (2021ESJ246)

Keywords: Spondylodiscitis, pyogenic, lumbar spine, infection, mastectomy

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INTRODUCTION

Pyogenic spondylodiscitis (PS) is an infection of the intervertebral disc and the adjacent vertebral bodies primarily due to the hematogenous seeding of a pathogen.[1] It can also originate following spine surgery.[5] PS is a rare complication representing 3–5% of all osteomyelitis cases, with an annual incidence in Europe of 0.4-2.4/100,000.[5] Until now, it remains a diagnostic enigma, and confirming the diagnosis often requires extensive testing. Usually, clinical examination, laboratory, and radiology findings hint towards the diagnosis. Magnetic resonance imaging (MRI) is the radiological test of choice with high accuracy and clear anatomical delineation of epidural space and spinal cord involvement.[8]

Spinal surgery can facilitate the process of discitis via damaging the lower and upper vertebral endplates following disc space curettage, direct introduction of germs, and instability.[18] The most common causes of postoperative discitis are nosocomial infections and direct inoculation during surgery.[13] PS can also originate from a distant infection, for example, endocarditis, abscess, urinary tract infection, pneumonia, or pelvic infection. Other causes include IV drug abuse and nonspinal surgical interventions.[17] A previous study reported a case series of 40 patients with PS following nonspinal surgeries, including gynecological, bariatric, plastic, and general surgeries.[20] Furthermore, the literature reported spontaneous PS cases that involve multiple levels.[6-11] However, multiple noncontiguous spontaneous PS is rare and has only been explained in the literature by case reports and case series.

We present a case report of multiple noncontiguous PS involving the cervical and lumbar spine following a mastectomy for breast cancer.

Case Report:

A 65-year-old female with a history of right-sided breast cancer who underwent surgery in February 2020 presented with severe back and neck pain. The back pain started two weeks following her mastectomy as mild in severity and responding to analgesia. However, the pain became intractable and was not relieved by analgesia, rest, or lying down. The pain started to radiate down the right leg a month after her presentation; on the other hand, the neck pain was mild in severity with no upper limb radiculopathy. The patient denied having dysuria, frequency, urgency, or hematuria. The patient has a history of breast cancer and underwent a right-sided mastectomy two weeks before her initial symptoms. On examination, the patient had an average BMI of 22 and a low-grade fever (37.5–38.3°C). She looked pale and in pain.

Gait examination revealed a right-sided antalgic gait, and the motor examination of the lower limbs showed increased pain on passive movement of the leg with mild weakness in the right ankle dorsiflexors (power grade 4/5) compared to the left (power grade 5/5). The examination also showed a positive straight leg raise test on the right side and right leg paraesthesia along with the distribution of L5 nerve roots. On the other hand, there were neither weaknesses nor features of upper motor neuron lesion (UMNL) in the upper limbs. Furthermore, there was no hepatosplenomegaly, and the genitourinary examination was unremarkable. Examination of the mastectomy (surgical site) site showed no signs of infection, and there was no clinical evidence of urinary tract infection (UTI) before mastectomy and spine surgery.

Informed consent from the patient was taken to publish her case and from our IRB.

Bloods And Laboratory Results

Upon presentation, the initial blood test showed hemoglobin (HB) = 11.6 mg/dl erythrocyte sedimentation rate (ESR) = 120 mm/hr, C-reactive protein (CRP) = 112 mg/L, and an average white blood cells count (WBC) = $9*10^9$/L. The preoperative blood and urine cultures were negative.

Blood tests after surgery were as follows: WBC = 12000; ESR = 40; CRP = 24. According to clinical suspicion, a contrast MRI of the

spine was requested, which revealed consistent spondylodiscitis at C5-C6 and L3-L4 levels with stenotic features at L4-L5 level (Figures 1,2). For management, the patient tried analgesics and empirical antibiotics at another center, which failed to resolve the symptoms before being referred to our center. Since the patient exhibited signs and symptoms of neural compression in the lower limbs and failure of conservative treatment, surgery was decided.

Surgical Technique:

The plan was to go for posterior spinal instrumentation from L3 to L5 levels, and decompression was done with a disc curettage and biopsy. Thus, the patient was intubated under general anesthesia, and a prone position was established. A confirmation of L3-L5 levels was done via a C-Arm, where the posterior midline incision was performed. Then, dissection of the subcutaneous fascia and muscle was achieved by electrocautery, and fixation of the posterior pedicle screws (Spineway, France) was performed at L3-L5 levels. Decompression was performed via laminectomy and discectomy of L3-L4 and L4-L5 levels, and samples were sent for histopathology, culture, and sensitivity. Eventually, debridement of the disc site was done, the local bone was used as a graft for fusion, the wound was closed in layers, and a suction drain was inserted (Figure 3).

It is worth mentioning that preoperative antibiotics were spared to avoid interaction with the biopsy specimens; however, intraoperative IV antibiotics were given immediately after sampling. Conservative treatment for the cervical spine was decided via a Philadelphia collar.

Postoperative Follow-Up:

Postoperatively, the patient was on empirical antibiotics: 2 gm ceftriaxone, IV, 24 hourly; 1 gm vancomycin, IV, 12 hourly, started at postoperative day 0 and continued up to the point where culture results showed an *E. coli*-type of infection. The therapy was changed according to the sensitivity results of the antibiogram where 1 gm meropenem, IV 8 hourly, was used for three

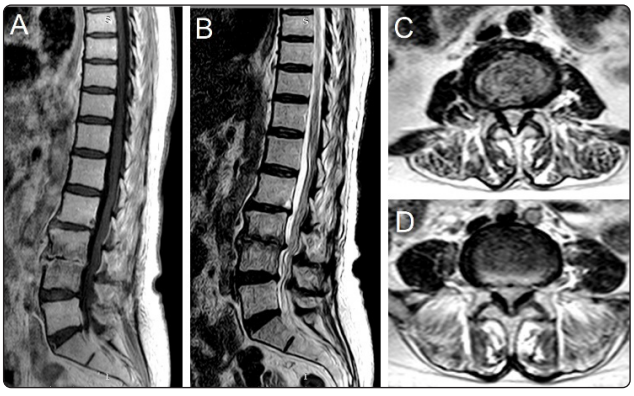

Figure 1. MRI of the lumbosacral spine, (A) T1 image sagittal, (B) T2 image sagittal, (C) T2 image axial L3-L4, and (D) T2 image axial L4-L5, showing the destruction of L3-L4 disc space with abnormal signal intensity at the adjacent endplate and degenerative stenosis of L4-L5 level.

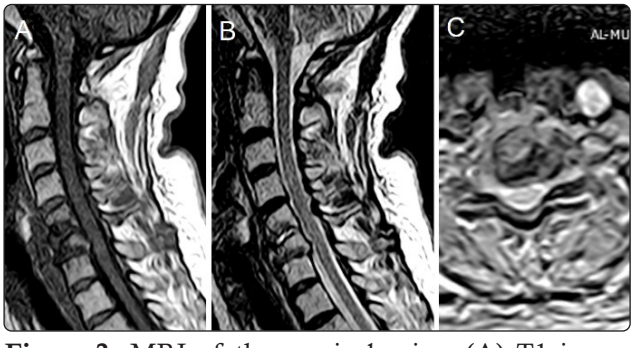

Figure 2. MRI of the cervical spine, (A) T1 image sagittal, (B) T2 image sagittal, (C) T2 image axial C5-C6 showing abnormal signal intensity at the levels of C5-C6 and C6-C7, with the hyperintense area at the disc level of C5-C6.

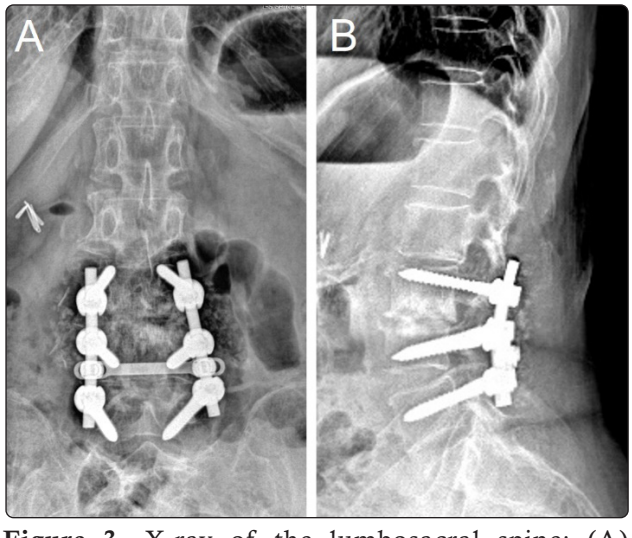

Figure 3. X-ray of the lumbosacral spine: (A) anteroposterior view and (B) lateral view, showing irregularity of L3-L4 disc space and posterior pedicle screws fixation of L3-L5 with decompression and posterolateral fusion.

weeks as it is readily available and susceptible to ongoing infection. Then after, oral antibiotics continue for another three weeks.

ESR and CRP titer tests were also performed for the follow-up plan. The patient remained in the hospital for three days, then discharged home. Removal of the stitching was done two weeks postoperatively. The patient restarted her radiotherapy for her breast cancer six weeks postoperatively. Regular outpatient clinical, radiological, and blood tests at six weeks, three months, six months, and one year were performed and showed improvement in symptoms and decline of ESR and CRP levels. Two weeks postoperatively, blood tests showed ESR = 90 and CRP = 64; six weeks postoperatively, ESR = 76, CRP = 24, and normal WBC; three months postoperatively, ESR = 55 and CRP = 10. Gradual improvement of neck and back pain intensities on the Visual Analog Score (VAS) and the follow-up duration are shown in Figure 4.

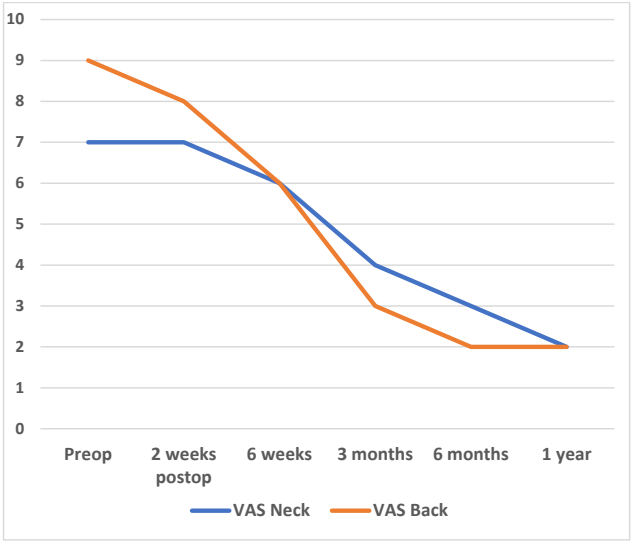

Figure 4. Line chart showing the neck and back pain intensities on the Visual Analog Score (VAS) along with the duration of follow-up.

DISCUSSION

Reaching the diagnosis of spondylodiscitis may be complicated and frequently done on presumptive evidence. The literature has suggested that, in most cases, discitis is caused by a hematogenous spread of microorganisms secondary to bacteremia caused by local or distant surgery. We hypothesize that the hematogenous space driving multiple noncontiguous spondylodiscitis, in this case, was facilitated by the fact that the patient's immune response was deranged secondary to her breast cancer and the stress of her recent mastectomy, and the isolation of *E. coli* from the infection site as *E. coli* is a common cause of UTI and bacteremia is common. Upon carefully reviewing the literature, we found that signs and symptoms are nonspecific in most cases, and back pain is the most common presenting complaint. On the other hand, motor weakness and sensory changes are only found in the minority. Unlike the consensus for an infection, fever is present in less than 50% of spondylodiscitis cases.[14] Examination findings are like those for mechanical back pain, making it a firm differential diagnosis except for localized tenderness, which increases the odds for spondylodiscitis. White blood cell counts might be elevated or normal, and CRP levels are increased in 58%-82% of the patients.[2,3]

If vertebral discitis is suspected, a contrast MRI is the gold standard radiological evaluation for such conditions with sensitivity, specificity, and accuracy of 96%, 92%, and 94%, respectively.[12] Typical MRI findings include a decreased signal intensity of vertebral bodies in T1-weighted images and increased disc signal intensity in T2-weighted images. Still, MRI might be negative early in the disease course, and the recommendations in the literature are to be repeated after 7–14 days if the diagnosis is still likely.[15]

E. coli causing multiple noncontiguous spondylodiscitis is a relatively novel condition. Upon reviewing the literature, few cases were found and were mainly in either tuberculosis (TB) or brucellosis.[4] Multilevel spinal involvement in the latter is rare and accounts for about 3.2–9%.[11]

In both TB and brucellosis, the participation of other musculoskeletal parts can occur.

Staphylococcus aureus is the most common microorganism causing PS in Europe, yet *Mycobacterium tuberculosis* is the most common worldwide. Unique to this case, the causative microorganism was *E. coli*, which is relatively novel to the published literature field, causing multiple noncontiguous PS.[12]

Gentile et al.[7], in their recent systematic review, have highlighted factors to consider for proper management of such cases. These include the presenting symptoms, neurological deficit, mechanical instability, presence of an abscess, and isolation of causative microorganism. The latter is essential for the management plan and can be done via blood culture, which is positive in only 58% of cases. Disc biopsy and CT-guided biopsy are ideal for patients with a negative blood culture. The microorganism can be isolated in 75% of cases, considering that disc biopsy is already a part of the surgical patients.[14]

As the patient has a history of breast carcinoma, the association of CRP and breast cancer is weak, as shown in previous literature.[16,19] In our patient, the high level of ESR and CRP is mainly related to spondylodiscitis, which is more sensitive than breast carcinoma, in addition to other diagnostic criteria of spondylodiscitis (clinical and MRI finding); thus, during follow-up, clinical improvement, including the pain and function, ESR, and CRP are used to assess the treatment outcome.

In our case, failure of conservative treatment, neurological deficit, and mechanical instability of the lumbar spine were the main factors behind deciding a surgical intervention for the lumbar spine.

It is worth mentioning that the patient had preexisting spinal stenosis. However, further clinical and radiological investigations, such as CT scans and MRIs, have shown facet arthrosis and degenerative changes. Thus, debridement, open biopsy, and spine instrumentation were undertaken to stabilize the unstable segment. The surgical decision was to include the stenotic segment (L4-L5) infusion to decrease the risk of adjacent segment disease (ASD).

On the other hand, conservative management includes bed rest, brace utilization, analgesia, and antibiotics according to culture and sensitivity. For the follow-up, clinical improvement and laboratory results of ESR and CRP are indicated.

CONCLUSION

Multiple noncontiguous spondylodiscitis after nonspinal surgery is a relatively rare complication that requires a high index of suspicion. Surgery is recommended in case of failure of conservative measures, neurological deficit, or mechanical instability as in this case. Clinical improvement and blood tests should be used to assess the treatment outcome.

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الملخص العربي

التهاب العمود الفقري المتعدد الغير متتابع بعد عملية ازاله الثدي بسبب سرطان الثدي، حالة مرضية ومراجعة للمقالات العلمية

البيانات الخلفية: يعد التهاب الفقار الفقاري القيحي العفوي غير المتجاور أمرًا نادرًا وقد تم شرحه فقط في الأدبيات من خلال تقارير الحالة وسلسلة الحالات.

الغرض: نقدم تقرير حالة عن التهاب الفقار الفقاري العفوي غير المتجاور الناجم عن الإشريكية القولونية التي تشمل العمود الفقري العنقي والقطني بعد استئصال الثدي بحثًا عن سرطان الثدي. سنشرح الصعوبات في التشخيص والعلاج والمتابعة مع المصاحب

تصميم الدراسة: تقرير حالة ومراجعة الأدبيات.

تقرير الحالة: خضعت مريضة في منتصف العمر ولديها تاريخ من الإصابة بسرطان الثدي. بعد أسبوعين من استئصال الثدي، أصيب المريض بألم شديد يشمل العمود الفقري العنقي والقطني وحمى منخفضة الدرجة (37.5-38.3 سيلزى). كشف الفحص العصبي عن مشية مسكنة للجانب الأيمن، وضعف في الجانب الأيمن، واختبار إيجابي لرفع الساق المستقيمة. عند التقديم، لوحظت مستويات مرتفعة من بروتين سي التفاعلي وخلايا الدم البيضاء. أظهر التصوير بالرنين المغناطيسي التهاب الفقار الفقاري الثابت عند مستويات الفقرات العنقية الخامسة والسادسة والقطنية الثالثة والرابعة مع تضيق في مستوى الفقرات القطنية الرابعة والخامسة. تم إجراء العلاج الجراحي للمنطقة القطنية عن طريق الأجهزة الخلفية للعمود الفقري من مستويات الفقرة القطنية الثالثة الى الخامسة وإزالة الضغط بأخذ خزعة. تم أيضًا إجراء فحص البروتين التفاعلي سي ومعد ترسيب كرات الدم الحمراء لخطة المتابعة التي أظهرت انخفاضًا في 3، 6، 12 أسبوعًا بعد الجراحة. تم إجراء العلاج المحافظ لمنطقة الرقبة بنتائج مرضية.

الخلاصة: يعد التهاب الفقار الفقاري المتعدد غير المتجاور بعد الجراحة غير العمود الفقري من المضاعفات النادرة نسبيًا التي تتطلب درجة عالية من الشك. يوصى بإجراء الجراحة في حالة فشل الإجراءات المحافظة، أو العجز العصبي، أو عدم الاستقرار الميكانيكي كما في هذه الحالة، يجب استخدام التحسين السريري واختبار الدم لتقييم نتيجة العلاج.