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CASE REPORT

Spontaneous Fusion After Cervical Disc Arthroplasty: A Case Report and Literature Review

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Objective: To report a rare case of spontaneous fusion (SF) following cervical disc arthroplasty (CDA), to review the related literature, and to propose a new measure to prevent it.

Methods: The course of a patient with SF is described here. The potential causes, risk factors, and preventive measure of SF after CDA published in previous studies have also been reviewed and discussed.

Results: A 63-year-old man presented with a 6-month history of progressive neck pain and developed left C-7 radiculopathy 4 years ago. Magnetic resonance imaging revealed disc herniation at the C6–C7 levels resulting in compression of the left C-7 nerve root. The patient underwent CDA at the C6–C7 levels, during which a PRESTIGE cervical disc device was implanted. He failed to follow-up regularly as recommended postoperatively because he was completely free from the pain in his neck and left upper limb. Four years later, he was readmitted with a 2-month history of occasional neck stiffness. Plain radiographs indicated complete radiographic fusion of the C6–C7 levels with trabecular bone bridging surrounding the cervical disc prosthesis, and dynamic imaging showed no motion. He was seen at regular follow-up visits for up to 60 months without special treatment, as his symptoms of neck stiffness were minor and his symptom has not worsened since then.

Conclusion: SF after CDA is a rare condition that can be attributed to patient- or prosthesisrelated causes, and its risk factors are diverse. SF after CDA did not affect the patient's clinical outcome, and no special treatment was required for it. Practitioners should be aware of this rare complication and advise patients of the risks before performing CDA.

Keywords: adjacent segment disease, cervical disc arthroplasty, spontaneous fusion, heterotopic ossification

Introduction

Anterior cervical discectomy and fusion (ACDF) has become the gold standard for treating degenerative cervical disc disease for the past decades.¹ However, some studies have shown that fusions limit the movement and alter the cervical spine biomechanics in the fused segment, which can result in the occurrence of adjacent segment disease (ASD).^{2,3} As a result, cervical disc arthroplasty (CDA) has emerged as an alternative treatment that may decrease the incidence of ASD through motion preservation.^{4,5} However, spontaneous fusion (SF), may also occur at the surgical segment as a rare complication of CDA.⁶ Herein, we describe a rare case of SF following CDA and review related literature to identify its courses, risk factors, and preventive measures.

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Case Report

Four years ago, a 63-year-old man presented with a 6-month history of progressive neck pain and left C-7 radiculopathy. The physical examination revealed left upper limb hypesthesia and weakness (strength, 4/5). T2-weighted magnetic resonance imaging scans of the cervical spine revealed disc herniation at the C6–C7 levels resulting in compression of the left C-7 nerve root (Figure 1).

After conservative treatment failed, surgery was recommended for the patient. Cervical anterior discectomy at the C6–C7 levels was performed via a left-sided approach to avoid injury of the recurrent laryngeal nerve. After discectomy and amputation of the posterior longitudinal ligament, the C6–C7 intervertebral space was distracted and held by a Caspar distractor. An unambiguous and dissociative herniated disc penetrating the posterior longitudinal ligament was found and removed completely during the decompression process. A drill was used to drill and open the bone canals to prepare the endplates. Subsequently, a prosthesis cervical disc device (PRESTIGE disc prosthesis; Medtronic Sofamor Danek) was implanted at the C6–C7 level (Figure 2).

Postoperatively, the patient failed to follow-up regularly as recommended, because he was completely free from pain in the neck and left upper limb, and the muscle strength of his left upper limb improved to normal immediately postoperatively. Four years later, he was readmitted with a 2-month history of occasional stiffness of the neck. Findings from the neurological examination were negative. Plain radiographs indicated complete osseous fusion of the C6–C7 levels with trabecular bone bridging surrounding the cervical disc prosthesis, and dynamic imaging showed no motion (Figure 3), which was further confirmed by computed tomography (CT) (Figure 4). We

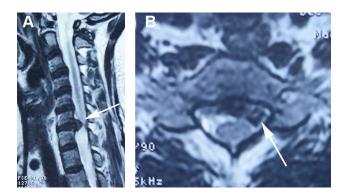


Figure I Preoperative MRI. Sagittal (A) and axial (B) MRI scans revealed a C6–C7 disc herniation resulting in compression of the left C-7 nerve root (arrow).



Figure 2 Intraoperative fluoroscopy. A prosthesis cervical disc device was implanted at C6–C7.

inferred that the patient had developed SF after CDA. He was seen at regular follow-up visits for up to 60 months without special treatment, as his symptoms of neck stiffness were minor and his symptom has not worsened since then. No specific radiological evidence of ASD in this patient was found at his last follow-up.

Discussion

Considering the excellent surgical results, ACDF has become more popular over the past few decades for the treatment of degenerative cervical spondylosis.¹ However, during the long-term follow-up, it was found that ACDF increased the compensatory activity of adjacent segments due to lack of motion in the fused segments.² Excessive compensatory activity may result in ASD, which may require a second surgery.³ As a result, CDA has emerged as an alternative that can not only achieve similar outcomes as that of ACDF but also reduce the incidence of ASD through motion preservation, which has been proved by many high-quality clinical studies.^{5,7,8} Many biomechanical studies have shown that compared with ACDF, CDA can maintain normal range of motion, intradiscal pressure, and facet joint contact pressure.^{9–11}

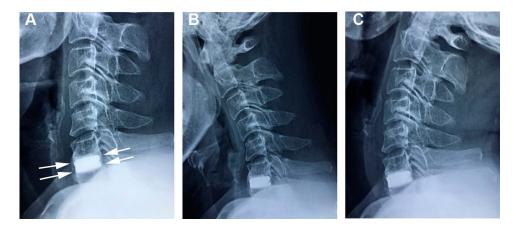


Figure 3 Plain radiographs obtained 4 years postoperatively. Lateral (A) imagings showed a complete radiographic fusion of the C6–C7 level with trabecular bone bridging surrounding the cervical disc prosthesis (arrows). No motion of C6–C7 was observed on flexion (B) and extension (C) imagings.



Figure 4 Computed tomography confirmed the cervical disc prosthesis was surrounded bone bridging (arrows).

Heterotopic ossification (HO), the formation of bone outside the skeletal system, was first proposed and categorized in the field of hip or knee arthroplasty by McAfee et al in 2003,¹² and classification of HO after CDA was

modified by Mehren et al in 2006.¹³ Under this system, HO was divided into 5 classes (0-IV) according to the severity of ossification. Class IV indicates that bony fusion formation is present at the replacement segment and flexion and extension activities of the replaced segment are limited. SF is a complete bony ankylosis or fusion without movement of the CDA in active flexion and extension, and should be categorized as class IV HO. As a relatively rare complication of CDA, SF or class IV HO after CDA has been reported a few times since it was first reported in 2005.^{14–26}

The exact incidence of SF after CDA is still unclear, and it varies greatly in the literature.²⁶ Oh et al reported that the incidence of SF after CDA was 3.3% (2/60).²⁷ However, Tu et al reported that its incidence was 1.9% (1/52).²⁶ This difference can be attributed to multiple factors. Some studies have identified a definite difference in the SF occurrence rate between different prosthetics types used in CDA.^{21,28} Moreover, detection tools may affect the incidence. Tu et al thought that CT was a more sensitive and accurate detector than plain radiography.²⁶ Some other researchers owed this difference to the dynamic progressive nature of HO partially, which means that it may increase over time.²⁹ Besides, the inter-observer error is also a factor that cannot be ignored.²⁸

The reasons for SF after CDA can be attribute to the result of patient- or prosthesis-related causes. With regard to the prosthesis-related causes, all cervical artificial discs can be divided into 3 categories according to its mechanism of motion: "constrained," the device is within the physiological range of motion; "semiconstrained," the device is outside of the normal range of motion; and "unconstrained," the device has no mechanical stop.²¹ Heary et al deemed that there was a strong association

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between the development of SF and degree of activity of the implanted prosthesis. If the motion of CDA is limited, then the artificial disc may behave like a cage and result in arthrodesis, so they thought that SF is more likely to occur with a constrained prosthesis.¹⁴ Besides, mismatch between the prosthesis size and intervertebral space size is another prosthesis-related cause.⁶ With respect to the patient-related causes, patients' underlying diseases, such as diffuse idiopathic skeletal hyperostosis, may predispose them to bridging bone developing in a scenario in which progression to ossification would not have occurred in most individuals.¹⁶

Several theories have been proposed to explain the risk factors of SF after CDA. Leung et al concluded that male sex and increased age were 2 possible risk factors for the development of SF.^{19,30} However, Yang et al showed that all factors including age, sex, body mass index, smoking, and alcohol were not associated with class IV HO.³¹ Moreover, some surgical related factors are also considered as the risk factors, such as direct trauma to the longus colli, excessive drilling on the endplates and removal of bone, and bone dust and osteogenesis after endplate milling.^{21,23,32} Among them, residual bone dust after endplate preparation was considered to be the most important factor by some researchers.¹⁷

Lee et al thought that strict surgical indication for CDA was the first step to prevent HO formation and subsequent SF formation.¹⁸ Osteophytes can usually be observed in patients with severe cervical degenerative disease, which may ultimately lead to arthrodesis between 2 adjacent vertebral bodies.33 Thus, patients with osteophytes are not appropriate candidates for CDA. Leung et al recommended copious irrigation of the operative site with normal saline during milling and again before closure.¹⁹ Additionally, it has been reported that radiation therapy and prophylactic use of non-steroidal anti-inflammatory drugs can decrease the incidence rates of HO after arthroplasty.³² Moreover, Yang et al proposed that maximizing the implant-endplate interface can help to reduce high-grade HO and preserve motion.²³ We believe that residual bone and blood supply are the key factors in the formation of SF after CDA. In our case, we routinely irrigated the wound with copious saline to remove residual bone dust introduced during endplate milling, and used bone wax to seal the milled endplate stump and block the blood supply before closing the wound to prevent SF formation (Figure 5). Before that, only 1 case of SF occurred, and after the routine protocol was in place,



Figure 5 The bone wax (arrows) was used to seal the milled endplate stump.

SF never happened again. This measure has never been reported before; we think it is a very effective way to prevent SF after CDA, and it should be used widely in the clinical setting.

In our case, the occurrence of SF after CDA did not significantly affect the patient's clinical outcome, except for limited range of motion at the index level, which was consistent with the conclusions of previously published literature.^{14,18,31,34} However, SF formation may increase the risk of ASD if fusion occurs at the surgical level.²³ A second surgery is only indicated when conservative treatment for cervical spondylosis due to ASD fails.²⁶ Our patient was satisfied with his clinical outcome, except for the occasional stiffness of his neck. So we just advised him to follow-up regularly. His symptom has not worsened since then, and no specific radiological evidence of ASD in this patient was found at his last follow-up.

Conclusion

SF after CDA is a rare condition that can be attributed to patient- or prosthesis-related causes, and its risk factors are diverse. In our case, SF after CDA did not affect the patient's clinical outcome, and no special treatment was required for it. Practitioners should be aware of this rare complication and advise patients of the risks before performing CDA.

Abbreviations

CDA, cervical disc arthroplasty; ASD, adjacent segment disease; ACDF, anterior cervical discectomy and fusion; SF, spontaneous fusion; HO, heterotopic ossification.

Ethics and Consent Statement

This study was approved by the ethics committee of Xi'an Honghui Hospital and the patient has provided written informed consent for publication of the case details and accompanying images.

Acknowledgments

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Disclosure

The authors report no conflicts of interest in this work.

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