Case report

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Crowned dens syndrome as a rare cause of anterior neck pain after transurethral resection of the prostate: a case report

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We describe the case of a 79-year-old man who presented with progressive aggravation of severe axial neck pain and fever 3 days after transurethral resection of the prostate (TURP), despite maintaining neutral neck posture during surgery. Laboratory examination revealed markedly elevated C-reactive protein levels and erythrocyte sedimentation rates. Computed tomography revealed crown-like calcifications surrounding the odontoid process. We diagnosed crowned dens syndrome (CDS) as the cause of acute-onset neck pain after TURP. The patient was treated with nonsteroidal anti-inflammatory drugs for 5 days, and his symptoms resolved completely. CDS is a rare disease characterized by calcific deposits around the odontoid process with acute onset of severe neck pain and restricted motion. Evidence of inflammation on serological testing and fever are typical of CDS. However, the prevalence and pathophysiology of CDS remain unclear. We hypothesized that systemic inflammation after prostate surgery may have induced a local inflammatory response involving calcification around the odontoid process.

Keywords: Inflammation; Neck pain; Odontoid process; Transurethral resection of prostate

Introduction

Crowned dens syndrome (CDS) is a rare condition that presents as acute onset of severe axial neck pain with restricted cervical motion in patients who are elderly [1]. It is characterized by calcific deposits around the odontoid process, creating the appearance of a half-ringed crown [2]. Clinicians tend to conduct many laboratory and imaging tests and even invasive diagnostic procedures before diagnosing CDS. The exact prevalence and pathophysiology of CDS remain unclear [3-5]. CDS resulting in severe axial neck pain with restriction of cervical motion after transurethral resection of the prostate (TURP) has not been reported previously.

Case

Ethical statements: This study was approved by the Institutional Review Board (IRB) of Keimyung University Dongsan Hospital (IRB No: DSMC 2021-06-006). Written informed consent was obtained from the patient to participate in the study.

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A 79-year-old man underwent TURP for benign prostate hypertrophy in the urology department of a tertiary hospital. He presented with acute-onset, unbearable, and intractable axial neck pain, which was accompanied by fever, 3 days after surgery. A neutral neck posture was maintained during the TURP, and no neck pain was observed for 2 days after surgery. The patient worked as a driver but had retired 10 years ago. He denied experiencing recent trauma and did not have a history of musculoskeletal illness. His medical history included chronic kidney disease and hypertension. His neck pain had progressively worsened, with marked restriction of neck motion. The patient's body temperature, systolic/diastolic blood pressure, and pulse rate were 38.7°C, 160/90 mmHg, and 100 beats/min, respectively. On physical examination, his cervical motion was found to be restricted in all directions. His laboratory test results were as follows: white blood cell count, 7,360/μL (range, 4,000–10,000/μL); neutrophils, 63.4% (range, 39%–73%); C-reactive protein (CRP), 10.6 mg/dL (range, 0–0.5 mg/dL); erythrocyte sedimentation rate (ESR), 59 mm/hr (range, 0–15 mm/hr); procalcitonin, 0.1 μg/L (range, < 0.1 μg/L); rheumatoid factor, 53.7 IU/mL (range, 0–14 IU/mL); and anticyclic citrullinated peptide, < 8.0 U/mL (range, 0–17 U/mL). The patient did not have any neurological deficits but presented with severe neck pain and rigidity during Kernig test.

Cerebrospinal fluid was collected via lumbar puncture and examined for the differential diagnosis of central nervous system infections such as meningitis, but the findings were within the normal range. Although plain radiography revealed spondylosis, the atlantoaxial joint appeared normal, which could not explain the cause of the patient's condition. Computed tomography (CT) of the cervical spine revealed crown-shaped calcium deposits surrounding the odontoid process (Fig. 1). Degenerative osteoarthritis was excluded due to elevated ESR and CRP levels. By integrating the results of the examinations listed above, we diagnosed CDS as the cause of the acute neck pain after TURP. A short course of nonsteroidal anti- inflammatory medication (aceclofenac, 100 mg/day for 5 days) completely resolved the neck pain and fever. The results of a follow-up serological test performed 5 days after administration of aceclofenac showed no evidence of an inflammatory reaction suggestive of CDS.

Discussion

The calcifications in CDS have been postulated to be composed of calcium pyrophosphate dihydrate or hydroxyapatite [6]. Calcium pyrophosphate deposition disease affects 4% to 7% of the adult population in the United States and Europe [7]. A recent large cross-sectional study indicated a point prevalence of 5.2 per 1,000, an average patient age of 68 years, and 95% male prevalence in the United States [8]. Acute pain attacks often occur after trauma, acute illness, or during a postoperative period [7].

The diagnosis of CDS is based on a combination of clinical symptoms, biological inflammatory markers, radiologic findings, and therapeutic responses [9]. Patients with CDS typically present with acute attacks of severe axial pain and restriction of cervical motion [9]. The C1 and C2 segment includes the odontoid process and provides a large range of cervical rotation; therefore, it is presumed that periodontoid calcifications may provoke neck pain during rotation [10]. The acute inflammatory features of CDS are often accompanied by fever and increased levels of inflammatory markers such as ESR and CRP.

It may be difficult to differentially diagnose CDS from other diseases that present with similar clinical symptoms, such as meningitis, spondylodiscitis, epidural abscess, osteomyelitis, polymyalgia rheumatica, giant cell arteritis, metastatic bone disease, and tumors

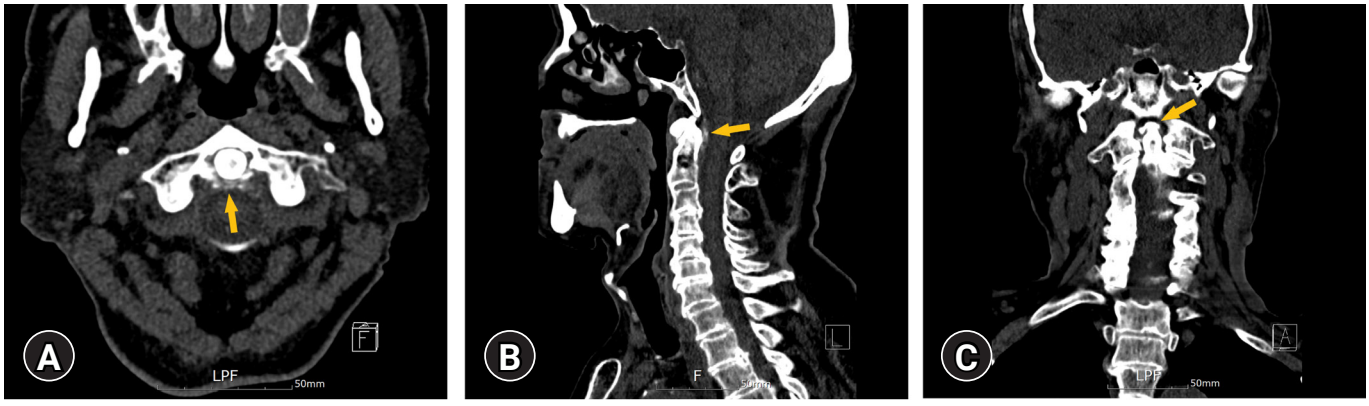

Fig. 1. (A) Axial, (B) sagittal, and (C) coronal computed tomography scans at the C1 and C2 level demonstrate crown-shaped calcium deposits (arrows) at the posterolateral side of the odontoid process.

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[2,3]. CT is the gold standard imaging method for identifying periodontoid calcific deposits with a half-ringed appearance [6]. Magnetic resonance imaging (MRI) can reveal inflammatory changes; however, MRI is less effective in showing calcifications than CT imaging [1]. Single-photon emission CT (SPECT) can assist with differential diagnosis in cases with subacute or chronic features, such as degenerative pain [1]. The appropriate treatment for CDS remains debatable; however, its prognosis is generally good. In many cases, a short regimen of anti-inflammatory agents, including nonsteroidal anti-inflammatory drugs, corticosteroids, colchicine, or combination therapy, can alleviate symptoms [11].

The exact prevalence, cause, and pathophysiology of CDS remain unclear. CDS predominantly affects women over 60 years of age [3,4]. Possible risk factors include recent surgery, invasive procedures, trauma, serious illness, and electrolyte abnormalities due to the use of diuretics or from dehydration [4,5]. Some authors have hypothesized reasonable theories regarding the pathogenesis of CDS related to inflammatory reactions [4,6]. CDS may involve phagocytosis of calcifications by inflammatory cells, which can evoke inflammatory responses in the joint [4]. The periodontoid calcifications may mechanically damage nearby structures, which can cause an inflammatory reaction [6]. The production of prostaglandins induced by the calcification may also lead to clinical symptoms of CDS [1].

While recent surgery is known to be a risk factor for CDS, no study has reported CDS development after TURP [1,12]. In previous studies, the occurrence of CDS after surgery included drainage of the subdural hematoma and endoscopic submucosal dissection for gastric cancer [5,13]. We assume that systemic inflammation after TURP may have been the main etiology of CDS in this case. Systemic inflammation due to surgery can trigger CDS, inducing a local inflammatory response in periodontoid calcifications [4]. These inflammatory reactions can cause symptoms attributed to CDS, such as axial neck pain, restricted neck motion, and fever in patients who are elderly. More cases are needed to determine whether these factors are associated with the onset of CDS. Our patient presented with pain 3 days after surgery and denied experiencing any symptoms between the immediate postoperative period and 2 days after surgery. The patient had no specific findings related to his urological surgery; thus, his symptom relief may not have been related to his postoperative recovery.

To the best of our knowledge, this is the first case report of CDS causing severe axial neck pain and fever after TURP. CDS should be considered in the differential diagnosis of patients with unexplained fever and acute onset of axial neck pain after TURP in order to perform appropriate management without unnecessary and aggressive examinations, inappropriate management, or long-term hospitalization. Although the patient was diagnosed with post-TURP-related CDS, he refused additional evaluations, such as MRI and SPECT, and the absence of such procedures is a limitation of this study. Clinicians should consider the possibility of CDS in patients with calcifications around the odontoid process even if they are asymptomatic at the time of examination after TURP. Appropriate recognition, accurate diagnosis, and prompt management are essential to minimize the impact of this condition and prevent postoperative neck pain in patients who are elderly.

Notes

Conflicts of interest

No potential conflict of interest relevant to this article was reported.

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Author contributions

Conceptualization: MGJ, BSP, ESS; Data curation: MGJ, ESS; Resources: ESS; Software: MGJ; Investigation: BSP; Funding acquisition, JHC; Writing-original draft: MGJ, BSP; Writing-review & editing: JHC.

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