Report of four cases of crowned dens syndrome: Clinical presentation, CT findings and treatment

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Abstract. The clinical manifestations of crowned dens syndrome (CDS) include acute neck pain and neck stiffness accompanied by restricted cervical range of motion. CDS is frequently misdiagnosed as meningitis, epidural abscess, rheumatoid arthritis, rheumatoid polymyalgia, giant cell arteritis, cervical spondylosis or metastatic bone tumor, and the incidence of CDS appears to be underestimated. The present study reported on four cases of CDS diagnosed by CT. They included one male and three females, aged from 67 to 78 years, and their major symptoms were acute neck pain and restricted cervical range of motion. Serum C-reactive protein levels and erythrocyte sedimentation rate were significantly increased in all cases. Cervical CT scan revealed calcified deposits surrounding the odontoid process in all cases. Non-steroidal anti-inflammatory drugs (NSAIDs) markedly reduced the levels of inflammatory indicators and rapidly relieved the symptoms. CT scan is considered the gold standard for CDS diagnosis, which may demonstrate calcification around the odontoid process. The patients' symptoms may be improved by treatment with NSAIDs.

Introduction

Neck pain is a frequently encountered complaint in emergency and orthopedic departments. According to statistics, ~71% of individuals experience neck pain during their lifespan (1). Crowned dens syndrome (CDS) was first reported by Bouvet *et al* (2) in 1985, which is a rare cause of neck pain with restricted mobility and its incidence is 2% in patients with acute neck pain (3). Since this first case was reported, only 88 further cases were reported in the literature until March 2020 (1-50). Due to its rarity, clinicians at emergency departments and orthopedic surgeons are generally not sufficiently aware of the disease and numerous cases with matching symptoms are not properly diagnosed; thus, the incidence of CDS appears to be underestimated.

The clinical manifestations of CDS include acute neck pain, neck stiffness accompanied by restricted cervical range of motion, fever and/or high serum C-reactive protein (CRP) levels and erythrocyte sedimentation rate (ESR). There are reports on cases of nerve root compression and even rare cases of spinal cord compression (51,52); these cases have symptoms similar to cervical spondylotic radiculopathy and myelopathy, i.e., radicular pain in the upper extremities, difficulty in walking, paralysis of the extremities and even progressively aggravated quadriplegia. A CT scan reveals the presence of irregular high-density shadows at different sizes surrounding the top and lateral sides of the odontoid process, appearing as a crown surrounding the top of the dens.

The present study reported on four cases of CDS who rapidly recovered after treatment with non-steroidal anti-inflammatory drugs (NSAIDs) at Hubei 672 Orthopaedics Hospital of Integrated Chinese and Western Medicine (Wuhan, China).

Case report

Case 1. A 76-year-old female was admitted to the Department of Minimally Invasive Spinal Surgery in Hubei 672 Orthopaedics Hospital of Integrated Chinese and Western Medicine (Wuhan, China) in May 2018, presenting with neck pain with restricted cervical range of motion of unknown causes for 3 days. She complained of persistent pain but had no other type of discomfort, such as numbness or pain in the upper limbs or unstable walking. The patient denied a history of gout or rheumatoid arthritis. On admission, the patient's body temperature was 36.6˚C and the neck muscle (sternocleidomastoid) was stiff with an obviously restricted cervical range of motion with a pain Visual Analogue Scale (VAS) score of 8. The patient had no signs of neurological or spinal cord injuries. Laboratory examination results revealed the following abnormalities: White blood cells (WBC), 3.33x10^9/l [normal range (NR), 4-10x10^9/l]; high-sensitivity CRP

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(hs-CRP), 31.0 mg/l (NR, 0-10 mg/l); ESR, 49.0 mm/h (NR, 0-15 mm/h); calcium, 2.30 mmol/l (NR, 2.03-2.6 mmol/l); and magnesium, 1.00 mmol/l (NR, 0.67-1.04 mmol/l). Rheumatoid factor (RF), anti-streptolysin O (ASO), anti-cyclic citrullinated peptide antibody (anti-CCP antibody) and procalcitonin (PCT) levels were normal. After admission, CT scans revealed arc-shaped calcification of the transverse ligament (Fig. 1A) and vertical line-like calcification of the cruciate ligament of the atlas in the posterior area of the odontoid process (Fig. 1B). According to the patient's medical history, physical signs and auxiliary examination results, CDS was diagnosed. The patient was administered nimesulide (100 mg/tablet, orally, once in the morning and once in the evening). After 7 successive days of treatment, hs-CRP and ESR recovered to normal, neck pain and restricted cervical range of motion were obviously alleviated, and the pain VAS score was 1. Neck pain and restricted cervical range of motion did not recur during the 10-month follow-up. The follow-up CT images at 10 months are presented in Fig. 1C and D. There was no significant difference from the previous CT scan obtained.

Case 2. A 70-year-old male was admitted to the Department of Minimally Invasive Spinal Surgery in Hubei 672 Orthopaedics Hospital of Integrated Chinese and Western Medicine in May 2019, presenting with neck pain with restricted cervical range of motion of unknown causes for 4 days. He complained of persistent pain but had no other types of discomfort, such as numbness or pain in the upper limbs or unstable walking. The patient had a history of gout. On admission, the patient's body temperature was 36.5˚C and his neck muscle (sternocleidomastoid) was stiff with an obviously restricted cervical range of motion with a pain VAS score of 8. He had no obvious signs of any neurological or spinal cord injuries. Laboratory examination results revealed the following: WBC, $8.17x10^9$/l; hs-CRP, 46.5 mg/l; ESR, 64 mm/h; calcium, 2.20 mmol/l; and magnesium, 0.92 mmol/l. RF, ASO, anti-CCP antibody and PCT levels were normal. After admission, CT scans revealed arc-shaped calcification of the apical ligament (Fig. 2A) and vertical line-like calcification of the cruciate ligament in the posterior area of the odontoid process (Fig. 1B). According to the patient's medical history, physical signs and auxiliary examination results, CDS was diagnosed. The patient was administered with lappaconitine hydrobromide (8 mg/ampoule, intravenous, once a day) and celecoxib (0.2 g/capsule, orally, once a day). After 3 successive days of treatment, hs-CRP and ESR recovered to normal, neck pain and restricted cervical range of motion were obviously relieved and the pain VAS score was 2. Neck pain and restricted cervical range of motion did not occur during the 3-month follow-up. The follow-up CT images at 3 months are presented in Fig. 2C and D. There was no significant difference from the previous CT scan obtained.

Case 3. A 73-year-old female was admitted to the Department of Minimally Invasive Spinal Surgery in Hubei 672 Orthopaedics Hospital of Integrated Chinese and Western Medicine in May 2019, due to neck pain of unknown causes for 10 days. The patient complained of persistent neck pain but had no discomfort, such as numbness or pain in the upper limbs or unstable walking. The patient had a history of gout. On admission, the patient's body temperature was 36.3˚C, and the neck muscle (sternocleidomastoid) was stiff with an obviously restricted cervical range of motion and a pain VAS score of 6. The patient had no signs of neurological or spinal cord injuries. Laboratory examination results indicated the following: WBC, $9.95x10^9$/l; hs-CRP, 19.6 mg/l; ESR, 34 mm/h; calcium, 2.31 mmol/l; and magnesium, 0.7 mmol/l. RF, ASO, anti-CCP antibody and PCT levels were normal. After admission, CT scans revealed arc-shaped calcification of the apical ligament in the anterior area of the odontoid process (Fig. 3A) and vertical line-like calcification of the cruciate ligament in the posterior area of the odontoid process (Fig. 1B). According to the patient's medical history, physical signs and auxiliary examination results, CDS was diagnosed. The patient was administered celecoxib (0.2 g/capsule, orally, once a day). After 7 successive days of treatment, hs-CRP and ESR recovered to normal, neck pain was obviously alleviated and the pain VAS score was 1. The neck pain did not recur during the 3-month follow-up. The follow-up CT images at 3 months are presented in Fig. 3C and D. Calcification in the anterior area of the odontoid process was more marked in the last follow-up.

Case 4. A 78-year-old female was admitted to the Department of Minimally Invasive Spinal Surgery in Hubei 672 Orthopaedics Hospital of Integrated Chinese and Western Medicine in June 2019, due to neck pain of unknown causes for 9 days. The patient complained of persistent pain but had no other type of discomfort, such as numbness or pain in the upper limbs or unstable walking. The patient had a history of hyperlipidemia. On admission, the patient's body temperature was 36.6˚C and the neck muscle (sternocleidomastoid) was stiff with an obviously restricted cervical range of motion and a pain VAS score of 9. The patient had no signs of neurological or spinal cord injuries. Laboratory examination results revealed the following: WBC, $7.52x10^9$/l; hs-CRP, 52.25 mg/l; ESR, 64 mm/h; calcium, 2.22 mmol/l; and magnesium, 0.95 mmol/l. RF, ASO, anti-CCP antibody and PCT levels were normal. After admission, CT scans revealed arc-shaped calcification of the transverse ligament (Fig. 4A) and vertical line-like calcification of the cruciate ligament of the atlas in the posterior area of the odontoid process (Fig. 4B). According to the patient's medical history, physical signs and auxiliary examination results, CDS was diagnosed. The patient was administered celecoxib (0.2 g/capsule, by mouth, once a day). After 5 successive days of treatment, hs-CRP and ESR recovered to normal, neck pain was obviously alleviated and the pain VAS score was 2. The neck pain did not recur during the 3-month follow-up. However, the patient refused to undergo CT examination again during the follow-up period.

Discussion

In 1985, Bouvet *et al* (2) first reported on CDS. They indicated that CDS mainly occurred in older individuals, i.e., at least 65% of patients with CDS were aged ≥70 years, with a male-to-female ratio of 3:5. Therefore, the majority of patients with CDS were female older adults. In the present study, four cases were aged ≥70 years and three out of four cases were female.

To date, the specific causes of CDS have remained elusive. CDS may be a pseudo atlantoaxial joint disorder caused by deposits of calcium pyrophosphate crystals (5,53,54). Calcium

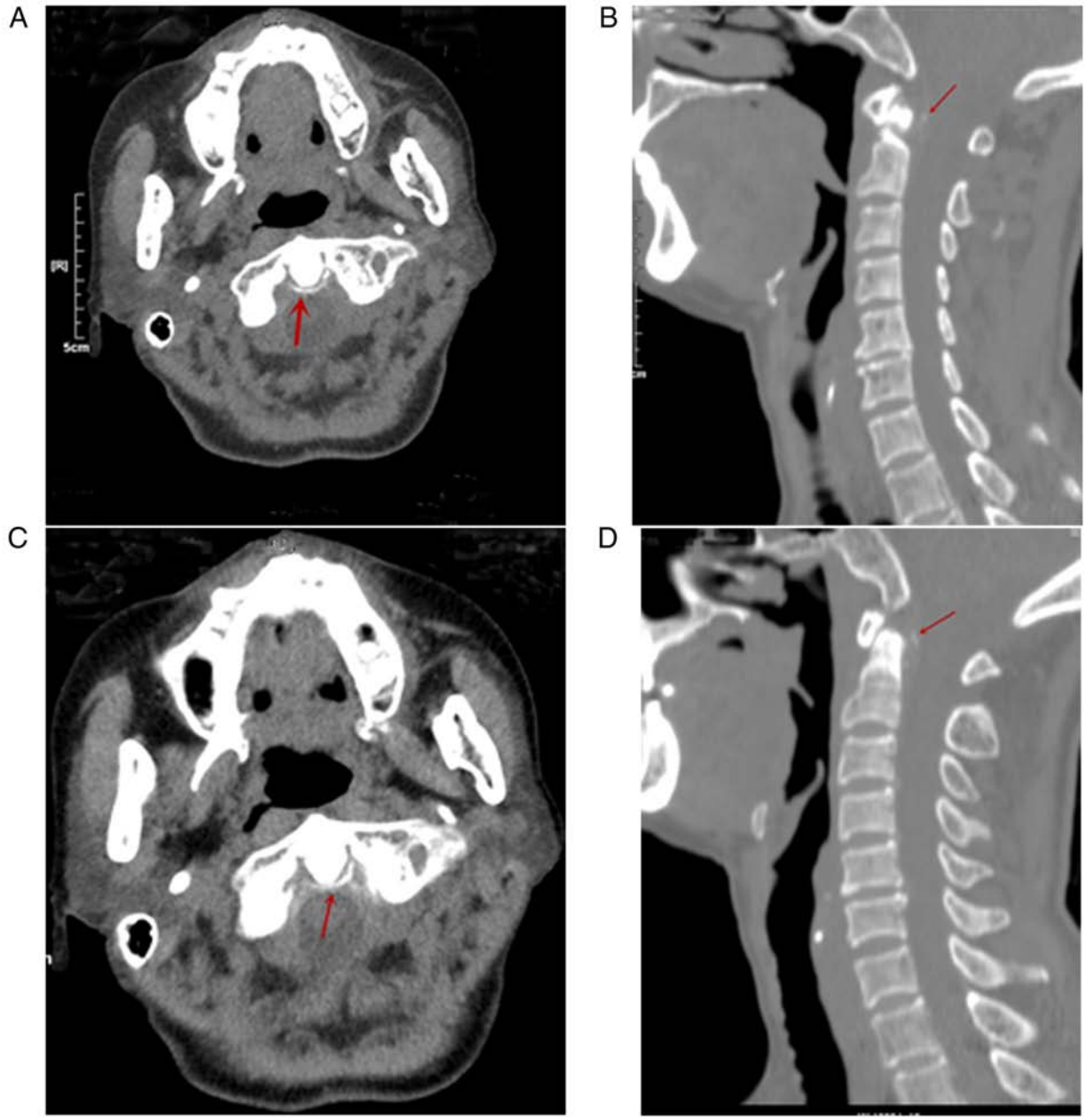

Figure 1. CT scans of a 76-year-old female. (A and B) CT scans at baseline revealing (A) an arc-shaped calcification of the transverse ligament and (B) vertical line-like calcification of the cruciate ligament of the atlas in the posterior area of the odontoid process. (C and D) Follow-up CT images at 10 months display (C) the arc-shaped calcification of the transverse ligament and (D) vertical line-like calcification of the cruciate ligament of the atlas in the posterior area of the odontoid process. Red arrows indicate the sites of calcium deposition.

pyrophosphate dihydrate crystal deposition disease occurs mostly in articular cartilage and ligaments. It is asymptomatic in half of the patients and it manifests as a joint inflammation similar to gout, which is referred to as pseudogout in certain patients. In contrast to the gout frequently occurring in older adult males, pseudogout is more common in older adult females. Pseudogout frequently affects the knees, hands, shoulder joints, elbow joints and feet, and it occasionally occurs in the cervical, thoracic and lumbar spine (55). Pseudogout occurring around the cervical odontoid process may cause neck pain, which usually manifests as acute or subacute moderate to severe neck pain, even restricted cervical range of motion and occipital pain (25). Certain patients have a fever, but neurological examination results are usually normal. All of the four cases reported in the present study had only acute severe neck pain and a restricted cervical range of motion, with no obvious fever or abnormal neurologic symptoms.

CT plain scan focusing on the atlantoaxial joint is considered the gold standard for CDS diagnosis. CT scans indicate calcification of the transverse, alar and apical ligaments around the odontoid process, which may occur anywhere around the odontoid process, but it most frequently occurs in the posterior and posterolateral area. In the radiological classification of CDS proposed by Goto *et al* (56), calcification may be present posterior (50%), posterolateral (27.5%), circular (12.5%), anterior (5%), lateral (5%) to the odontoid process. In the present study, calcification occurred at the posterior side of the odontoid process in all of our cases. Regarding laboratory

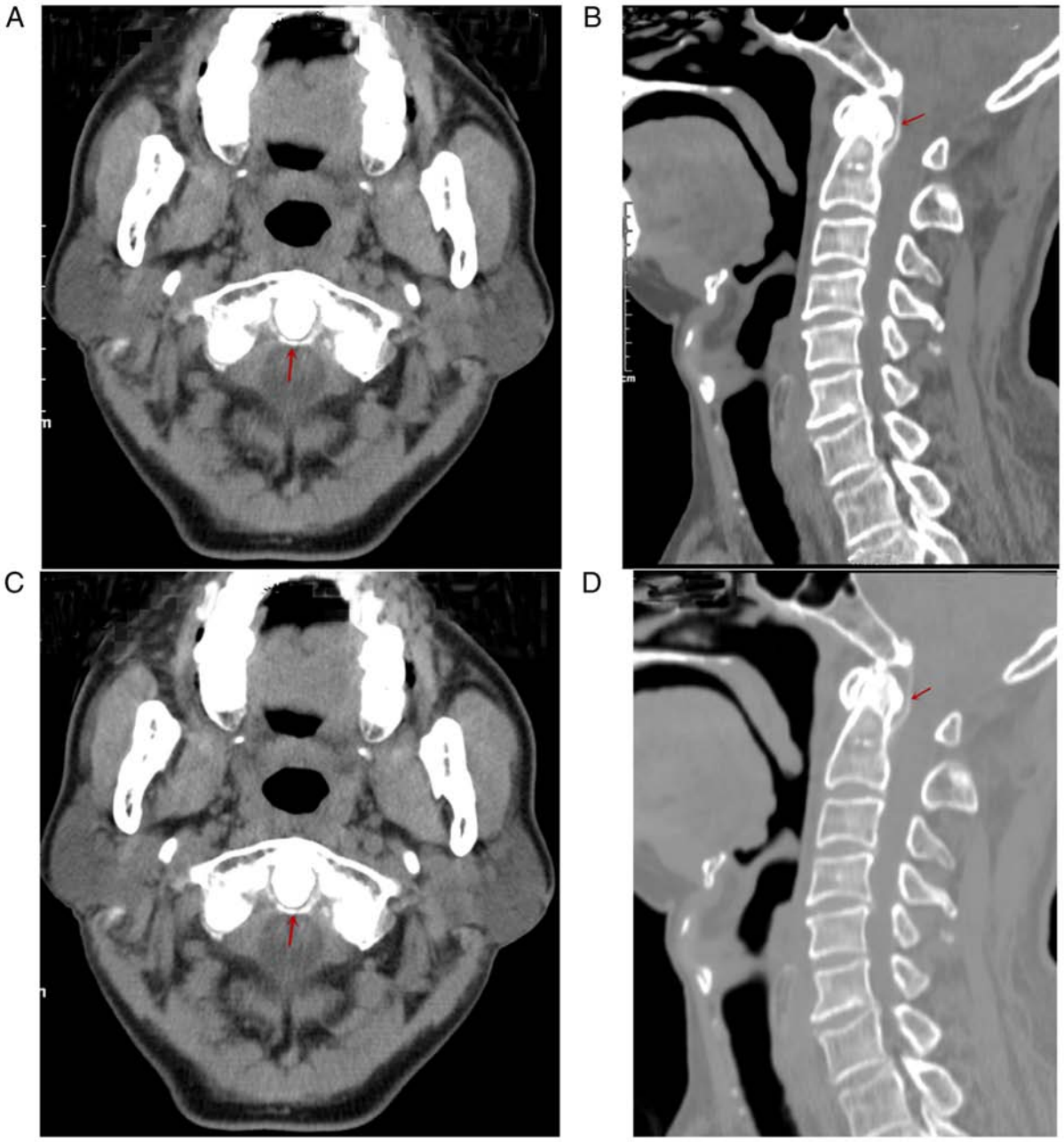

Figure 2. CT scans of a 70-year-old male. (A and B) CT scans at baseline revealing (A) arc-shaped calcification of the apical ligament and (B) vertical line-like calcification of the cruciate ligament in the posterior area of the odontoid process. (C and D) Follow-up CT images at 3 months displayed (C) the arc-shaped calcification of the apical ligament and (D) the vertical line-like calcification of the cruciate ligament in the posterior area of the odontoid process. Red arrows indicate the sites of calcium deposition.

parameters, CRP and ESR are frequently markedly elevated and WBC are normal or slightly increased (53,30,43). In the four cases of the present study, CRP and ESR were obviously elevated, but the increase in WBC was not obvious, and it was decreased in one case.

CDS should be differentiated from meningitis, epidural abscess, rheumatoid arthritis, rheumatoid polymyalgia, giant cell arteritis, cervical spondylosis or metastatic bone tumor (1,43,44,53). All of the above diseases may manifest as neck pain, fever and restricted cervical range of motion. Neck pain in CDS radiates from the bilateral suboccipital area to the neck part, with no specific tender point or obvious neck rotation limitation. It may be clearly determined from CT scans of the atlantoaxial joint. This avoids unnecessary invasive treatments (such as lumbar puncture, tissue biopsy), inappropriate medication (such as antibiotics, antiviral drugs) and long-term hospitalization. CDS should also be differentiated from atlantoaxial synovial cysts, which represent a rare disease entity and may also cause neck pain. The development of atlantoaxial synovial cysts has been linked to spinal instability and trauma. Imaging with CT and MRI scans is crucial for the diagnosis and characterization of synovial cysts (56,57).

In general, patients with CDS have a good prognosis and their symptoms usually resolve within a few weeks. However, the current treatment of CDS remains controversial. NSAIDs are usually recommended. In most cases reported in the

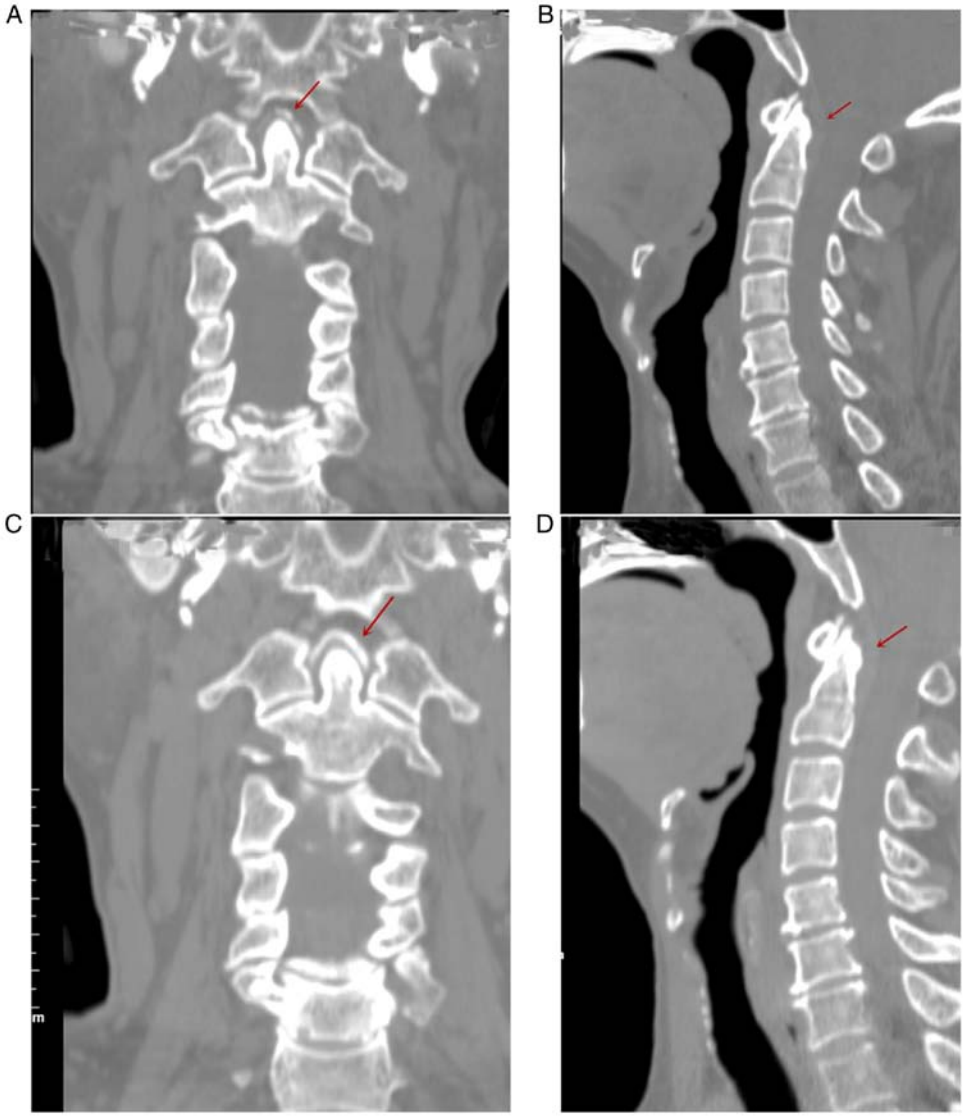

Figure 3. CT scans of a 73-year-old female. (A and B) CT scans at baseline revealed (A) an arc-shaped calcification of the apical ligament in the anterior area of the odontoid process and (B) vertical line-like calcification of the cruciate ligament in the posterior area of the odontoid process. (C and D) Follow-up CT images at 3 months displayed the (C) arc-shaped calcification of the apical ligament in the anterior area of the odontoid process and (D) vertical line-like calcification of the cruciate ligament in the posterior area of the odontoid process. Red arrows indicate the sites of calcium deposition.

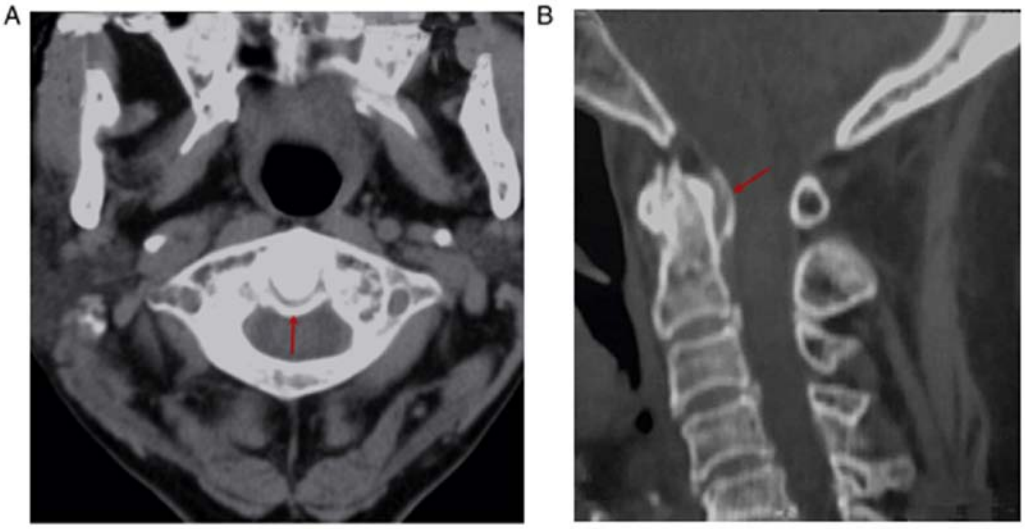

Figure 4. CT scans of a 78-year-old female. CT scans revealing (A) an arc-shaped calcification of the transverse ligament and (B) vertical line-like calcification of the cruciate ligament of the atlas in the posterior area of the odontoid process. Red arrows indicate the sites of calcium deposition.

literature, oral NSAIDs alone may improve symptoms within a few days. Although severe neurological complications are rare, extensive deposits may result in myelopathy or cervical stenosis, for which surgical decompression may be necessary (14). Surgical decompression and stabilization may alleviate the compression on the cervical spinal cord, but the potential for neurological recovery remains to be further elucidated (23). In the cases of the present study, the symptoms rapidly resolved after oral administration of NSAIDs. In certain refractory cases, colchicine or a small amount of corticosteroids may be administered, but since CDS occurs mainly in older individuals, steroid therapy should be considered with caution to avoid any fatal side effects (30,43,44,53,58).

In summary, due to the rare and non-specific manifestations of CDS, its diagnosis is frequently missed, which delays its treatment and CDS is easy to treat. Therefore, when patients have acute neck pain accompanied by a restricted cervical range of motion, as well as fever, particularly in older individuals, CDS should be considered.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Authors' contributions

JT and JL made substantial contributions to the study conception and design, the acquisition of data and the analysis and interpretation of data. CW, XL, YL, QL, WX and TZ contributed to drafting the manuscript and critically revising the manuscript for important intellectual content. JT prepared the manuscript. All authors read and approved the final manuscript.

Ethics approval and consent to participate

The present study was approved by the Ethics Committee of Hubei 672 Orthopaedics Hospital of Integrated Chinese and Western Medicine (Wuhan, China; permit no. HB6720121) and was in conformity with the guidelines of the National Institute of Health.

Patient consent for publication

The four patients provided written informed consent for the publication of their data.

Competing interests

The authors declare that they have no competing interests.

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