Calcium pyrophosphate dihydrate crystal deposition disease and retro-odontoid pseudotumor rupture managed via posterior occipital cervical instrumented fusion: illustrative case

Armaan K. Malhotra, MD,1 Aayush R. Malhotra, MKin,2 Alexander P. Landry, MD,1 Arjun Balachandar, MD,3 William Guest, MD, PhD,4 Aditya Bharatha, MD,4 Thomas R. Marotta, MD,4 and Christopher D. Witiw, MD, MS1

1Division of Neurosurgery and 4Department of Interventional Neuroradiology, St. Michael’s Hospital, University of Toronto, Toronto, Ontario, Canada; 2Department of Kinesiology, University of British Columbia, Vancouver, British Columbia, Canada; and 3Department of Neurology, University of Toronto, Toronto, Ontario, Canada

BACKGROUND Craniocervical junction and subaxial cervical spinal manifestations of calcium pyrophosphate deposition disease are rarely encountered. The authors presented a severe case of retro-odontoid pseudotumor rupture causing rapid quadriparesis and an acute comatose state with subsequent radiographic and clinical improvement after posterior occipital cervical fusion.

OBSERVATIONS The authors surveyed the literature and outlined multiple described operative management strategies for compressive cervical and craniocervical junction calcium pyrophosphate deposition disease manifestations ranging from neck pain to paresthesia, weakness, myelopathy, quadriparesis, and cranial neuropathies. In this report, radiographic features of cervical and craniocervical junction calcium pyrophosphate deposition disease were explored. Several previously described surgical strategies were compiled, including patient characteristics and outcomes.

LESSONS With this case report, the authors presented for the first time an isolated posterior occipital cervical fusion for treatment of a compressive retro-odontoid pseudotumor with rupture into the brainstem. They demonstrated rapid clinical and radiographic resolution after stabilization of cranial cervical junction only 12 weeks postsurgery.

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KEYWORDS CPPD; crystal arthropathy; odontoid pseudotumor; brainstem; calcium pyrophosphate; occiput cervical fusion

Calcium pyrophosphate dihydrate (CPPD) deposition disease (pseudogout), in which calcium pyrophosphate crystals are identified from synovial fluid aspirate, was first described in 1962.1 CPPD may present acutely with inflammatory oligoarthritis lasting weeks to months, and typically it has a predilection for the knees and wrists. In its chronic form, it may present with polyarticular arthritis resembling osteoarthritis, with severe joint changes, intermittent inflammatory flares, and a tendency to involve glenohumeral joints, wrists, and metacarpophalangeal joints.2 A certain subtype of chronic CPPD may mimic rheumatoid arthritis, with a more persistent inflammatory arthropathy affecting small and large joints. The pathophysiology underlying CPPD involves calcium pyrophosphate crystal formation in articular cartilage, subsequent synovial inflammation, and resultant direct catabolism of joint tissues and cartilage degeneration.2

CPPD involvement in the cervical spine is a relatively uncommon entity; previous studies have demonstrated calcium pyrophosphate deposition in intervertebral disks and ligamentum flavum among affected patients.3,4 One series described 18 cases of crowded dens syndrome characterized by acute onset of febrile neck pain, elevated serum inflammatory markers, and computed tomography (CT) evidence of craniocervical ligament calcification in which calcium pyrophosphate deposition was identified in most cases.5 Craniocervical or subaxial cervical spinal CPPD has been rarely shown to cause acute neck pain, cervical myelopathy, and, in severe cases, quadriparesis.6–13 Nonsurgical therapy for cervical CPPD typically involves nonsteroidal antiinflammatory therapy or steroid therapy for acute flares and colchicine for longer term suppressive therapy.2,14 However, in the setting of acute neurological

ABBREVIATIONS CPPD = calcium pyrophosphate dihydrate; CT = computed tomography; MRI = magnetic resonance imaging.

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compromise secondary to compressive subaxial or craniocervical junction CPPD, operative intervention has been described in different reports through transoral resection of the anterior arch of C1 and odontoid either alone or in combination with posterior cervical decompression and fusion (Table 1).

Herein, we present for the first time a case of cervical CPPD with associated retro-odontoid pseudotumor rupture into the brainstem leading to acute cranial neuropathy, quadriparesis, and respiratory failure. We describe a technique for posterolateral CT-guided needle biopsy of a retro-odontoid cyst, followed by treatment of C1–2 CPPD through posterior occipital cervical fusion alone without ventral decompression. The clinical course along with subsequent radiographic resolution of the compressive effects are highlighted. A comprehensive literature review was performed to examine cases in which operative intervention for craniocervical and subaxial cervical CPPD was undertaken.

Illustrative Case

A 68-year-old man with a history of chronic pain, remote left hip arthroplasty, depression, and atrial fibrillation presented to the emergency department with a history of several preceding months of bilateral hand numbness and acute onset of paresthesia in both arms and legs. These symptoms were followed by diplopia, gait instability, and speech difficulty. Clinically, he was awake and alert; his pertinent physical examination findings included bilateral sixth nerve palsy, preserved vertical eye movements, dysarthria, and hoarse speech. His face was symmetric, there was normal facial sensation, and tongue protrusion was midline. His distal power was reduced in the bilateral upper extremities, with evidence of intrinsic muscle wasting, diffuse hyperreflexia, and an upward left plantar reflex. Shortly after admission, he was unable to maintain adequate oxygenation and developed quadriparesis with only antigravity strength in the myotomes of his left side. His level of consciousness declined, necessitating intubation. He was transferred to our neurosurgical center for further investigation and management.

On arrival at the neurosurgical center, he had no responses to central or peripheral pain, and his pupils remained reactive to light. CT revealed a lytic lesion at C2 with evidence of cortical erosion through the posterior odontoid wall (Fig. 1). Magnetic resonance imaging (MRI) demonstrated an epidural cystic structure extending superiorly into the retro-odontoid and retroclival spaces that appeared to contact the posterior odontoid wall (Fig. 1). Enhancement of the lesion was 6.7 cm. The retroclival epidural cystic structure was in continuity with a cystic cavitation within the pons measuring 2.2 × 2.2 × 2.4 cm, raising the possibility that the epidural lesion ruptured through the dural membrane and entered the pons. A thin border at the posterior aspect of the pontine cystic lesion separated it from the posterior aspect of the pontine cystic lesion. The C2 lesion demonstrated rim enhancement following gadolinium administration, and both the retro-odontoid and pontine regions exhibited increased susceptibility in keeping with hemorrhagic contents. Abscess was believed unlikely given complete absence of diffusion restriction. There were severe spondylotic changes between the occiput and C2, calcification of the apical andalar ligaments, facet degeneration between the articulations of occuput and C1–2, and autofusion at C2, C3, and C4. The radiographic differential diagnosis included a synovial-based process, such as calcium pyrophosphate deposition disease or pigmented villonodular synovitis, versus a chronic infection or craniocervical neoplasm, such as chordoma.

After interdisciplinary case review, CT-guided biopsy was performed by interventional neuroradiology. The patient was placed prone, and contrast-enhanced CT was performed to identify the vertebral arteries. A needle trajectory was planned to target the lytic lesion at C2 via a posterolateral approach, passing between the laminae of C2 and C3 to enter the epidural space. An 18-gauge 9-cm spinal needle was advanced to the lateral epidural space; needle tip position was confirmed on imaging and epidural location was demonstrated through injection of a small volume of air. Next, the needle was advanced into the epidural space along the pedicle and carefully stepped along the posterior cortex of the C2 vertebral body until its tip was visualized within the lytic lesion in the dens (Fig. 2). Aspiration yielded partially coagulated chronic appearing sanguineous material. Fluid microbiology was negative for any growth, and cytology was negative for malignant cells. Microscopy confirmed positivity for intracellular birefringent crystals with rare calcium pyrophosphate crystals, thus confirming a diagnosis of retro-odontoid pseudotumor secondary to CPPD. Subsequent shoulder radiographs showed severe bilateral glenohumeral articular degenerative findings, chondrocalcinosis, and ossific bodies in the axillary recess, also in keeping with advanced CPPD arthropathy. The patient was maintained on dexamethasone therapy and over the following week demonstrated gradual improvement in neurological function. He remained intubated, his eyes opened spontaneously, and he obeyed commands in four extremities. He had 1/5 power in right-sided myotomes and 3/5 power in left-sided myotomes through upper and lower extremities.

Given his significant improvement in level of consciousness with steroid therapy, definitive surgical management was discussed. A ventral decompression with odontoidectomy was believed to pose significant morbidity given the operative risks and his current clinical condition. A transsphenoidal transclival skull base approach to the brainstem was also considered although it was believed to pose high operative risk. Moreover, concerns regarding radiographic findings suggestive of a dorsal membrane breach were also considered in the context of the associated risk of postoperative infection and cerebrospinal fluid leak. We felt that this retro-odontoid pseudotumor was likely the result of instability at the craniocervical junction and secondary joint synovium hypertrophy and crystal deposition. With this rationale, a posterior occipital-cervical fusion would achieve the goal of restoring craniocervical junction stability and facilitating calcium pyrophosphate deposition resolution with fusion of motion segments. The patient therefore received surgery for posterior occiput–C3 instrumented fusion with occipital plate, C2 pedicle screws, and C3 lateral mass screws (Fig. 3A and B). After a period in the intensive care unit, the patient was extubated within 1 week and started on inpatient physiotherapy. At his 4-week postsurgical follow-up, the strength in his upper and lower extremities had improved substantially although he remained nonambulatory and continued to experience difficulty feeding himself. MRI performed at the 12-week follow-up examination demonstrated marked and rapid resolution of the retro-odontoid pseudotumor with no ongoing compressive myelopathy and improvement in the brainstem cystic lesion (Fig. 3C).

Discussion

Observations

Craniocervical junction manifestations of inflammatory arthritides remain an important diagnostic consideration for neurosurgeons. The most common radiographic presentation of craniocervical junction CPPD is as a periodontoid “pseudotumor,” a partially calcified
| Authors & Year                  | Patients (n) | Presentation | Lesion Location       | Surgical Intervention                                                                 | Outcome                                                                 |
|--------------------------------|--------------|--------------|-----------------------|----------------------------------------------------------------------------------------|-------------------------------------------------------------------------|
| Nagashima et al., 1984<sup>17</sup>* | 3            | Neck pain (n = 2), paresthesias (n = 1), tetraparesis (n = 1) | Subaxial spine                  | Laminectomy w/o fusion (n = 3), additional facetectomy (n = 2)                             | Neurological improvement (n = 3); 1 patient died of postoperative pneumonia |
| Berghausen et al., 1985<sup>18</sup> | 1            | Paresthesias, weakness, ataxia                          | Subaxial spine                  | Laminectomy w/o fusion (C2–4)                                                            | Neurological improvement                                               |
| Kawano et al., 1988<sup>19</sup>* | 3            | Paresthesias & ataxia (n = 3), quadripareisis (n = 3)    | Subaxial spine                  | Laminectomy w/o fusion                                                                  | Neurological improvement (n = 3)                                       |
| Ciricillo & Weinstein, 1989<sup>12</sup> | 1            | Paresthesias, weakness, incoordination                    | Craniocervical junction         | Transoral resection of C1 arch, C2 body, & dens                                          | Neurological improvement                                               |
| Gomez & Chou, 1989<sup>20</sup>   | 1            | Neck pain, weakness, ataxia                             | Subaxial spine                  | Laminectomy (C3–5) w/ interfacet wire fusion (C2–6)                                     | Neurological improvement                                               |
| Kingdom et al., 1995<sup>28</sup>  | 1            | N/A                                                     | Craniocervical junction (C1–2)  | Transoral transpalatopharyngeal resection                                               | N/A                                                                    |
| Norris & Hope, 1995<sup>21</sup>* | 1            | Quadripareisis                                          | Subaxial spine                  | Laminectomy w/o fusion                                                                  | Neurological improvement                                               |
| Shaffrey et al., 1995<sup>26</sup> | 1            | Neck pain                                              | Craniocervical junction (posterior C1–2) | En bloc posterior resection of lesion, C1–3 laminectomy w/o fusion                  | Clinical improvement                                                  |
| Zünkeler et al., 1996<sup>29</sup> | 7            | Neck pain (n = 3), paresthesias (n = 6), ataxia (n = 5), fine motor problems (n = 7) | Craniocervical junction         | Transoral transpharyngeal resection including C1 arch & dens (n = 7), posterior occiput to C2 fusion (n = 6) | Neurological improvement (n = 7)                                       |
| Hasegawa et al., 2000<sup>34</sup> | 1            | Paresthesias, ataxia, incoordination                    | Craniocervical junction         | Posterolateral resection including C1 hemilaminectomy & partial C2 hemilaminectomy     | Neurological improvement                                               |
| Yamagami et al., 2000<sup>24</sup> | 1            | Hypesthesia, arm weakness, ataxia                       | Subaxial spine                  | Laminectomy (C4–6) w/o fusion                                                          | Neurological improvement                                               |
| Griesdale et al., 2004<sup>30</sup> | 1            | Numbness, hand weakness                                 | Craniocervical junction         | Transoral resection & posterior C1–2 fusion w/ transarticular screw & interlaminar autograft | Neurological improvement                                               |
| Muthukumar & Karuppuswamy, 2003<sup>23</sup> | 2            | Quadripareisis (n = 2)                                  | Subaxial spine                  | Laminectomy w/o fusion                                                                  | Neurological improvement                                               |
| Lin et al., 2006<sup>33</sup>     | 1            | Weakness, paresthesias                                  | Craniocervical junction (retro-odontoid) & subaxial spine (ligamentum flavum) | Laminectomy (C3–6) w/o fusion, including removal of ligamentum flavum up to posterior arch of atlas | Neurological improvement                                               |
| Doita et al., 2007<sup>10</sup>   | 1            | Weakness, paresthesias                                  | Craniocervical junction         | Posterolateral resection w/ C1 laminectomy & partial C2 hemilaminectomy                 | Neurological improvement                                               |
| Sethi et al., 2007<sup>35</sup>   | 1            | Quadripareisis                                          | Craniocervical junction (w/ intradural extension, vertebral encasement) | Posterolateral resection w/ C1 & C2 laminectomies, no fusion                            | Postoperative quadriplegia, ventilator dependence leading to death from pneumonia & sepsis |

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soft-tissue mass centered behind the odontoid process in direct contact with the transverse ligament that may compress the cervical spinal cord with progressive growth. Although no clear imaging features can distinguish nonrheumatoid (such as CPPD) from rheumatoid causes of periodontoid pseudotumor, the typical radiological manifestations of craniocervical junction CPPD include calcific deposits in the annulus fibrosis and transverse ligament of the dens as well as in the supraspinous, interspinous, and longitudinal ligaments of the surrounding cervical spine. CT of the cervical spine may show mineralization within the pseudotumor, which may be associated with crowded dens syndrome, although this may also be present in hydroxypatite deposition and appear almost identical to CPPD. MRI of the cervical spine usually shows low T1 signal and heterogeneous T2 signal within the pseudotumor. Associated arthropathy may also be seen, characterized by narrowing of the intervertebral discs, osteophytes, bony sclerosis, and bony erosions with subchondral cysts. Finally, increased motion at the craniocervical junction may often be present on flexion-extension radiographs in a manner similar to rheumatoid arthritis, with suspected shared mechanical instability pathophysiology underlying further joint space disease. Other differential diagnoses in this region include bony neoplasms, such as chordoma or sarcoma, chronic infectious processes, and, in our case, primary intraparenchymal brain neoplasms given the significant brainstem lesion.

**TABLE 1. Summary of operative management and outcome for reported craniocervical and subaxial cervical spine calcium pyrophosphate deposition disease**

| Authors & Year | Patients (n) | Presentation | Lesion Location | Surgical Intervention | Outcome |
|----------------|-------------|--------------|-----------------|----------------------|---------|
| Fenoy et al., 2008 | 21 | Neck pain (85%), paresthesias (61%), lower cranial neuropathy (29%) | Craniocervical junction | Transoral transpalatopharyngeal resection (n = 19), concomitant posterior occipital cervical fusion (n = 16), posterolateral C2 laminar exploration for laterally oriented CPPD lesion (n = 2) | Improved at follow-up (n = 17), unchanged at final follow-up (n = 1), lost to follow-up (n = 3) |
| Ali et al., 2011 | 1 | Neck pain, quadriplegia | Craniocervical junction | Posterior decompression & fusion | No recovery of quadriplegia, died of cardiac arrest |
| Kobayashi et al., 2016 | 1 | Neck pain | Subaxial spine | Laminectomy (C6) w/ removal of ligamentum flavum at C5–6 | Complete relief of neck pain |
| Manhas et al., 2016 | 1 | Quadriplegia | Craniocervical junction w/ significant intracranial extension | Far lateral craniotomy w/ C1 laminectomy, lesion resection | Neurological improvement |
| Ng et al., 2016 | 1 | Neck pain, incoordination, ataxia | Craniocervical junction (posterior C1–2) | Laminectomy (C2–6) w/ instrumented fusion | Follow-up not documented |
| Madhavan et al., 2018 | 3 | Neck pain (n = 3), incoordination (n = 1), ataxia (n = 1), weakness (n = 1) | Craniocervical junction | Suboccipital craniectomy w/ C1/partial C2 laminectomies, transdural lesion resection (n = 3), occipitocervical fusion (n = 2) | Neurological improvement (n = 3) |
| Chang et al., 2020 | 2 | Neck pain, radicular arm pain, & ataxia (n = 2) | Subaxial spine | Laminectomy w/ removal of calcified lesions (n = 2), posterior instrumented fusion (n = 1) | Neurological improvement (n = 2) |
| Liao et al., 2021 | 1 | Neck pain, numbness, weakness | Subaxial spine | Laminectomy (C4) w/ ligamentum resection, no fusion | Neurological improvement |

N/A = not applicable.
* Isolated subaxial ligamentum flavum CPPD.
† Review article.
The optimal surgical strategy for craniocervical and subaxial spinal CPPD is not clear. Results from our literature review on surgical management strategies and outcomes of subaxial and craniocervical junction CPPD are summarized in Table 1 and included 24 articles with relevant cases. No identified cases used isolated posterior fusion for craniocevical junction CPPD, nor did any cases describe such profound retro-odontoid pseudotumor rupture into the brainstem, as in this report. Ten articles reported on CPPD in the subaxial cervical spine (n = 16 patients), most commonly involving the ligamentum flavum. Symptoms in this group included neck pain, radiculopathy, and myelopathy. All cases underwent focal posterior decompression, and one underwent fusion with neurological improvement, with the exception of one postoperative mortality secondary to pneumonia.8,17–25 Two articles (n = 2 patients) reported posterior craniocevical junction involvement, and both patients were treated with posterior decompression (fusion in one case); one patient improved, and no follow-up was available for the other.7,26 The remaining articles identified described retro-odontoid CPPD pseudotumor management through different surgical techniques across a total of 40 patients. Neck pain, paresthesia, ataxia, motor weakness/quadriparesis, myelopathy, and lower cranial neuropathies were described in this subgroup. Thirty-one patients received transoral resections, and a total of 23 were stabilized with concomitant posterior fusion including either occipital cervico-2 fusion (n = 22) or C1–2 fusion (n = 1).12,27–30 No mortalities were reported in this group; most patients improved, four patients did not have follow-up information, and one patient was unchanged at follow-up. Posterior suboccipital craniectomy and C1–2 partial laminectomies with transdural pseudotumor resection with or without occipital cervical fusion was performed in three patients with improvement in all cases.31 Posterior craniocevical junction decompression alone for ligamentum flavum calcium deposition was performed in two patients; one had neurological improvement and the other passed away secondary to cardiac arrest.32,33 Four patients received posterolateral resection (C1–2 laminectomies), with posterior fusion in one case.10,27,34 Of this group, there was improvement in two cases, and none worsened postoperatively. This excluded the two patients with significant intracranial extension. Two patients had significant intracranial extension most similar to our case. Both were treated with posterolateral resection without fusion, and one patient had postoperative quadriplegia and ventilator dependence and CPPD are summarized in Table 1 and included 24 articles with relevant cases. No identified cases used isolated posterior fusion for craniocevical junction CPPD, nor did any cases describe such profound retro-odontoid pseudotumor rupture into the brainstem, as in this report. Ten articles reported on CPPD in the subaxial cervical spine (n = 16 patients), most commonly involving the ligamentum flavum. Symptoms in this group included neck pain, radiculopathy, and myelopathy. 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This excluded the two patients with significant intracranial extension. Two patients had significant intracranial extension most similar to our case. Both were treated with posterolateral resection without fusion, and one patient had postoperative quadriplegia and ventilator dependence and
arthritides, should be considered with high cervical lesions. When possible, efforts should be made to confirm diagnoses with a direct biopsy if imaging is not clear to determine optimal next-management strategies. We demonstrate for the first time an instance of complete resolution of craniovertebral junction compressive myelopathy secondary to retro-odontoid and retroclival CPPD with posterior occipital cervical fusion alone. This approach may be considered when a patient’s clinical circumstances suggest that the morbidity profile of a ventral approach is unfavorable. Furthermore, we provide a summary of management strategies used in available literature that demonstrate good efficacy and result in improvement in neurological status in most cases of CPPD. Overall limitations include lack of long-term follow-up in available literature, especially when comparing long-term craniovertebral junction stability in cases receiving fusion versus no fusion. This remains an area to be addressed in future studies.

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Lessons

It is challenging to generate overall surgical management recommendations given the heterogeneity of included case imaging and patient features. A broad differential diagnosis, including inflammatory

died due to respiratory sepsis whereas the other experienced neurological improvement.35,36 Nonsurgical management for degenerative retro-odontoid pseudotumor has also been described using rigid cervical collar immobilization with good clinical and radiographic results although this strategy certainly requires close surveillance and may be considered in patients with high operative risk or significant frailty.37
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**Disclosures**

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

**Author Contributions**

Conception and design: AK Malhotra, Balachandar, Guest, Witiw. Acquisition of data: all authors. Analysis and interpretation of data: AK Malhotra, AR Malhotra, Balachandar, Bharatha, Marotta. Drafting the article: AK Malhotra, AR Malhotra, Balachandar, Bharatha, Marotta. Guest, Balachandar, Bharatha, Marotta. Approved the final version of the manuscript on behalf of all authors: AK Malhotra. Administrative/technical/material support: Bharatha, Marotta, Witiw. Study supervision: Witiw.

**Correspondence**

Aarmaan K. Malhotra: St. Michael’s Hospital, Toronto, ON, Canada. ak.malhotra@mail.utoronto.ca.