

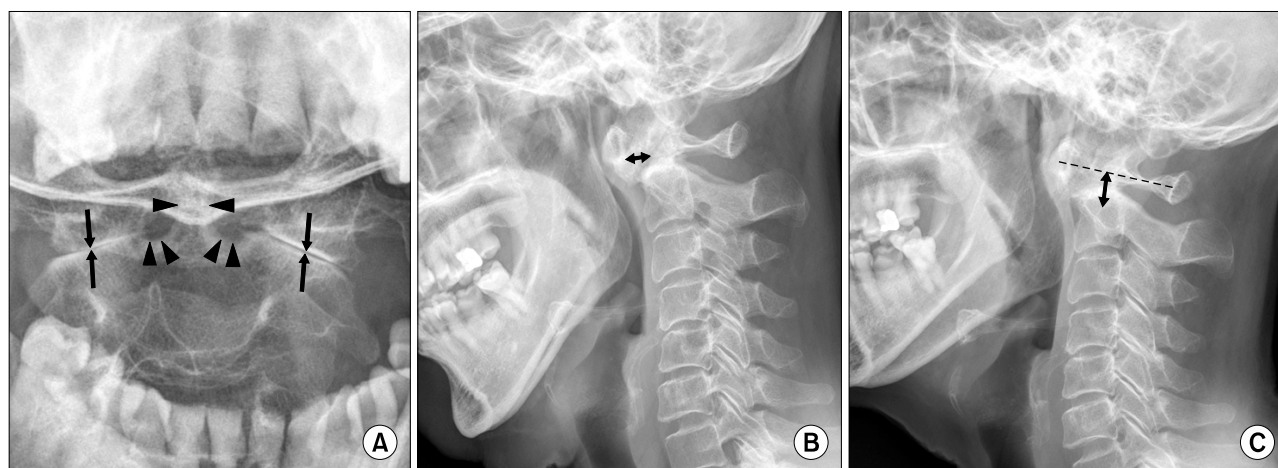
# Progressive Occipitocervical Subluxation Despite Biologic Agent Therapy in a Patient with Advanced Rheumatoid Arthritis

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Rheumatoid arthritis (RA) is a chronic systemic inflammatory disease that can affect the cervical (c-) spine in up to 80% of the patients [1,2]. Although c-spine involvement in RA is often asymptomatic, uncontrolled high disease activity eventually becomes a major risk factor for future spinal instability [3]. Adequate treatment with conventional or biologic disease-modifying anti-rheumatic drugs (DMARDs) can minimize cervical instability, but it does not halt the progression of pre-existing joint damage [3]. Furthermore, progressive atlantoaxial instability can induce intractable neck pain and can cause neurological deficits [4]. We hereby report a

case of a 68-year-old woman who presented with occipital headache for 10 weeks. She was diagnosed with sero-positive RA in 2003. She was first treated conservatively according to her own wish. She shortly developed pain in her knees, and eventually received total joint replacement arthroplasty for her left (2006) and right (2013) knees. A preoperative c-spine evaluation performed in 2013 revealed bony erosions at both C1 and C2 facet joints and anterior atlantoaxial subluxation (Figure 1). The Ranawat C1~C2 index, which is used to evaluate vertical subluxation, was 12 mm, and it was determined by assessing the length between the center of the C2 pedicle and a line



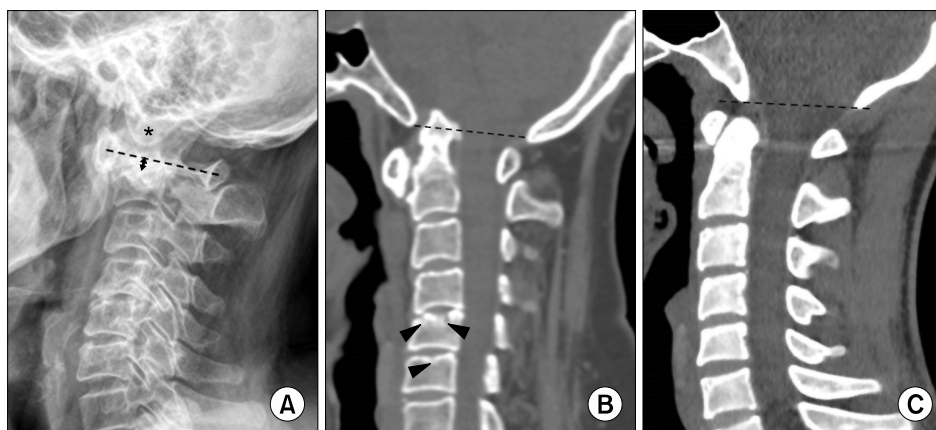
**Figure 1.** (A) There are multiple erosions (arrowheads) at the base and neck of the odontoid process of axis (open mouth view). Note the symmetrical joint space narrowing (arrows) of lateral atlantoaxial joints, which is a typical finding in rheumatoid arthritis. (B) Anterior subluxation of the atlantoaxial joint is evident at flexion. The atlantodental interval is 7 mm (double-headed arrow). (C) The Ranawat C1~C2 index (double-headed arrow) is 12 mm, just below the normal range.

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**Figure 2.** (A) Three years later, the Ranawat C1~C2 index (double-headed arrow) is markedly decreased, suggesting basilar impression. Note the odontoid process (asterisk) is protruded beyond the horizontal axis of atlas (dashed line). (B) The reformatted sagittal computed tomography (CT) scan shows that the odontoid process is protruded above the level of foramen magnum (McRae line; dashed line). There are multiple erosions (arrows) in the C4, C5 discovertebral area. (C) A reformatted sagittal CT scan of a 25-year-old healthy female. The odontoid process is under the McRae line (dashed line).

connecting the midpoint of the anterior and posterior arches of C1 (normal value; male >15 mm, female >13 mm) [5]. This index is useful because it does not require visualization of the tip of the odontoid process nor the hard palate (as in the McGregor index). She finally began receiving conventional DMARDs in April 2014 after developing intractable joint pain in her hands, shoulders, and knees. Her disease activity score (DAS)28 soared up to 5.8. Intravenous tocilizumab therapy without methotrexate was initiated in May 2015. After the third infusion, her joint symptoms were alleviated and DAS28 was substantially reduced to 2.0. Nevertheless, the image obtained in August 2016 showed vertical atlantoaxial instability which had worsened than before; she developed severe occipital headache. The Ranawat C1~C2 index was significantly reduced to 4.3 mm (Figure 2A). Bony erosions in the discovertebral area of her c-spine were also prominent (Figure 2B). In summary, this case presents a patient with advanced RA who showed progressive occipitocervical subluxation, regardless of one-year long treatment with biologics.

## CONFLICT OF INTEREST

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

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