Retro-Odontoid Mass in High Elderly Patients
- Genesis, Therapeutic Strategies and Post-Operative Care -

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ABSTRACT

It has been controversial whether surgical treatment for retro-odontoid mass including disc hernia is indicated. We surgically treated 8 patients with retro-odontoid mass. We discuss clinical characteristics, therapeutic strategies, the genesis based on histological findings, and problems of post-operative care in this lesion.

Age distribution of the patients was from 77 to 86 years old (mean: 81.0 years). Male/female ratio was 6/2. All patients showed neurological deficits due to myelopathy. Pre-operative magnetic resonance (MR) imaging showed an isointensity mass at the retro-odontoid space. Only marginal enhancement was seen in 2, and heterogenous enhancement in 6 patients.

All patients underwent surgery via postero-lateral transdural approach for removal of the extradural retro-odontoid mass. All patients showed neurological improvement, however, severe neurological deficits still remained in 2 patients who had been in bed-ridden pre-operatively. The surgical specimens consisted of degenerative cartilaginous tissue and reactive fibrovascular tissue.

As the retro-odontoid mass contains various pathological lesions, histological confirmation is necessary. Among reported surgical approaches, the postero-lateral transdural approach is less invasive, especially for high elderly patients. A source of cartilaginous tissue is probably derived from the herniated disc penetrated posterior longitudinal ligament at C2/3, which has migrated upward to the retro-odontoid space. Not only such migration, but also some proliferative change might come to form the retro-odontoid mass.

Retro-odontoid mass is characteristic in very elderly patients. Elderly patient-specific problems such as disuse atrophy due to less movement and painful arthritis deformans in the lower extremities prevent successful rehabilitation. Earlier planning to advance rehabilitation in cooperation with neurosurgeons, orthopedists, and rehabilitation physicians is necessary for very elderly patients' quality of life.

Key words: aging, disc hernia, myelopathy, odontoid process, surgical approach

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Introduction

It is well known that higher cervical degenerative diseases such as cervical spondylosis and cervical disc hernia are encountered more frequently in elderly patients compared with younger patients [11,21,22,27]. In elderly patients, spondylotic changes occurred in mid- and lower cervical levels and flexibility at these levels has become limited. Because of such reason, higher cervical spines are more loaded for the motion of the neck in elderly patients than in younger patients. Cervical flexion/extension study clearly demonstrates such evidence [22].

One of characteristic cervical lesions in very elderly patients is retro-odontoid mass. In a clinical entity of retro-odontoid mass, retro-odontoid disc hernia is included [19,21,22,24]. There have still been a few reports regarding retro-odontoid disc hernia [19,21,22,24]. Pathogenesis of such lesion, histological findings, and strategies for the treatment have been previously reported [21,22]. We have surgically treated retro-odontoid mass in 8 patients. Because the ages of all patients are very high, specific post-operative care is mandatory. We reported surgical strategies and post-operative care for these patients with retro-odontoid mass, and discuss the clinical implications of this lesion.

Clinical Materials and Methods

Clinical Materials

We have surgically treated 8 patients with retro-odontoid mass. Age distribution of the patients was from 77 to 86 years old (mean ± standard deviation: 81.0 ± 3.5 years). Male / female ratio was 6 / 2. Pre-operative neurological state was as follows: 3 patients; ambulatory and independent in their daily lives, although they had some clumsiness in bilateral hands and some weakness in lower extremities, 3 patients; ambulatory with a cane and some assistance was necessary in their daily lives, 2 patients; bed-ridden and total care was necessary in their daily lives. Table 1 shows the clinical summary of the patients.

Magnetic resonance (MR) imaging showed large retro-odontoid mass and spinal cord was severely compressed at C1 / C2 levels. The patterns of MR images were classified into two types; a low-isointensity mass and the margin of the mass was only enhanced (2 patients) (Fig. 1), and an isointensity mass and the mass was heterogeneously enhanced (6 patients) (Fig. 2 left, center). Pre-operative cervical dynamic study showed atlanto-axial subluxation in only one patient.

Methods

Surgical procedures

All patients underwent surgery. In prone position, the midline was incised and the laminae of C1 and C2 were exposed, and bilaterally laminectomy. The
Fig. 2: MR imaging showing another type of retro-odontoid mass, in which the content is almost homogenously isointensity (left, pre-operative pre-contrast MR imaging) and heterogenously enhanced (center). Post-operative MR imaging showing the removal of the retro-odontoid mass and sufficient decompression of the upper spinal cord (right).

Fig. 3: A picture showing retro-odontoid mass covered by the dura. After incising the dentate ligament, retro-odontoid mass can be easily exposed via postero-lateral transdural approach without any retraction of the spinal cord (upper), and sufficient removal of the retro-odontoid mass through the anterior dural incision was achievable (bottom).

Fig 4: Pre- and post-operative MR imagings of a patient with atlanto-axial subluxation (instability index: 24.2 %). MR imaging shows homogenously isointensity retro-odontoid mass (upper left) with heterogenous enhancement (upper right). Post-operative MR imaging shows sufficient decompression of the upper spinal cord (lower left). The patient underwent surgery of occipitocervical fixation with instrumentation (lower right).
lesion was located almost at midline but slightly deviated to each side. At that side, the laminae of C1 and C2 were drilled away more laterally. The dura was incised, and dentate ligaments were cut. As the spinal cord was compressed backward, the approach to the lesion was easily achievable via posterolaterally without any retraction of the spinal cord or sacrifice of nerve roots. The lesion was completely covered by the dura (Fig. 3 upper) and the mass was removed after incision of the dura (Fig. 3 bottom). The content of the mass was composed of partly gelatinous component and mostly cartilaginous and fibrous tissue. In some cases, ultrasound aspirator was necessary to remove the content of the mass. After sufficient removal of the mass and decompression of the spinal cord, anterior dural incision was sealed with Surgicel® and fibrin glue, and dural incision of the posterior surface was watertight closed. As one patient showed atlanto-axial subluxation pre-operatively (instability index [1]: 24.2%), posterior fixation between occipital bone and C3 with instrumentation was performed (Fig. 4 lower right). No internal fixation was necessary in another 7 patients. All patients wore a Philadelphia neck collar for 2 weeks post-operatively, thereafter removed. Rehabilitation was started a few days after surgery.

Results

Post-operative findings of MR images

Post-operative follow-up period was 1.5 - 5.2 years (mean: 3.8 ± 1.4 years). Post-operative MR imaging showed that the retro-odontoid mass was removed enough to obtain sufficient decompression of the upper spinal cord in all patients (Fig. 2 left, Fig. 4 lower left). Post-operative dynamic cervical pictures revealed no instability in upper cervical spines in 7 patients. The patient who underwent posterior fixation with instrumentation showed no instability in upper cervical spine.

Post-operative neurological findings

All patients showed some neurological improvement after surgery. Three patients, who had been ambulatory by themselves but had had clumsiness in hands and weakness in lower extremities, could be totally independent in their lives. They complained of numbness in distal parts of all extremities. Two patients out of 3, who had been ambulatory with a cane and had needed some assistance in their daily lives, could be ambulatory without a cane and independent, but another one patient still needed a cane in walking and some assistance in his daily life. Two patients, who had been in bedridden before surgery, could use a wheel chair and stand up with assistance after surgery. One patient could use a spoon for her eating by herself. However, they still needed almost total care for their daily lives.

Histological examinations of surgical specimens

In all patients whose pre-operative MR imaging
showed either type described above, the surgical specimens consisted of degenerative cartilaginous tissue (Fig. 5 upper) and reactive fibrous tissue with some vascular proliferation (Fig. 5 bottom).

**Discussion**

**Genesis of retro-odontoid mass**

The retro-odontoid mass contains various pathological lesions. The surgical specimens obtained from our all patients consisted of degenerative cartilaginous tissue and reactive fibrous tissue with some vascular proliferation. Obviously, no disc component exists between C1 and C2, or retro-odontoid space. A source of retro-odontoid cartilaginous tissue is probably derived from herniated disc between C2 and C3. Herniated disc between C2 and C3 might penetrate posterior longitudinal ligament and migrate upward. Those phenomena have been already reported before [18]. From these viewpoints, retro-odontoid mass in our series can be considered as "retro-odontoid herniated disc". Retro-odontoid herniated disc is very characteristic specifically in very elderly patients [21,22]. In these patients, mid- and lower cervical spines are less mobile because of spondylotic changes, and most load in neck movement is shifted from C5/6 to the levels of C2/3 and C3/4 [22]. The incidence of cervical disc hernia is highest at most loaded level during cervical movement [9]. Mechanical forces at higher cervical segments might be a cause to push the herniated disc at C2/3 upward to retro-odontoid space in very elderly patients. To support our speculation, we have already reported an elderly patient who had herniated disc at C2/3 [22]. Furthermore, MR imaging of one case also shows disc hernia between C2 and C3, which continuously extends to retro-odontoid mass (Fig. 6). This MR imaging of the case also supports our speculation.

As Fig. 2 and Fig. 4 show, the disc volume between C2 and C3 are not always decreased even in our patients with retro-odontoid mass. This is a contradictory result to support our speculation that retro-odontoid herniated disc might migrate from C2/3. However, histological examinations show some proliferative change such as reactive fibrovascular tissue. From these evidences, retro-odontoid mass in our series might be formed not only by migrated herniated disc, but also by reactive proliferation due to overloaded neck movement at C2. Based on histological findings and cervical dynamic study, this is a most likely mechanism in the formation of retro-odontoid mass.
Basic concept of therapeutic strategies
Since MR image has been routinely used for cervical lesions, retro-odontoid masses have unexceptionally become identified. Various pathology are contained in retro-odontoid mass, such as inflammatory, metabolic, traumatic, and neoplastic lesions. As inflammatory lesion, rheumatoid arthritis is most common to induce abnormal proliferation of the synovial soft tissue (pannis) [25,28]. In metabolic lesion, deposition of calcium pyrophosphate dehydrate in cartilage is well known as pseudogout or chondrocalcinosis [7,16]. With relation of os odontoidem, retro-odontoid soft tissue mass has been reported [6,12,15]. By traumatic origin, periodontoid hypertrophic cicatrix has been reported as a pseudotumor [17,23,26]. Atlanto-axial subluxation also forms retro-odontoid soft tissue mass [14,26]. In neoplastic lesion, chordoma [4], chondroma [20] or osteoblastoma [13] have been reported.

In most of pseudotumor, retro-odontoid soft tissue mass by atalanto-axial subluxation, or pannus by rheumatoid arthritis, a conservative treatment using a neck collar is adopted as the first choice for the treatment. However, the mass significantly compresses the spinal cord and neurological deficits of the patients do not relieve by a conservative treatment, surgical managements should be considered as the next step. Posterior decompression by laminectomies and posterior fixation by transarticular fixation or by a neck collar without removal of the lesion itself have been reported as the first choice for the surgical treatment [15,17,23,28]. If the lesion is small and the degree of the compression of the upper spinal cord is not severe, such treatments might be justified. However, when the compression of the spinal cord at C2 by the lesion and patients' neurological condition are severe, simple decompression by laminectomies and posterior fixation are not sufficient enough. As described, tumorous lesions also contain in retro-odontoid mass. In that sense, surgical removal of the lesion itself and histological confirmation of the lesion are necessary. Then, following appropriate treatments can be planned.

Surgical strategies
Various surgical approaches have been reported for extradural lesions in the cranio-vertebral junction or retro-odontoid space, such as transoral [3,5,8,10], transcondylar [2], far-lateral [19] or postero-lateral transdural approaches [21,22]. There are advantages and disadvantages in each approach. As described, retro-odontoid mass is most characteristic in very elderly patients. Among these approaches, postero-lateral transdural approach is significantly less invasive. In this approach, removal of the extradural retro-odontoid mass is easily achievable by just simple laminectomies of C1 and C2. In our series, no mortality and morbidity were encountered. Because upper cervical cord is compressed backward, the lesion can be easily identified and removed through enough surgical space via postero-lateral transdural approach. Furthermore, sufficient decompression of upper spinal cord can be obtained by wide laminectomies.

Pre-operative cervical dynamic study is necessary. If a patient has any instability at atalanto-axial joint, wide laminectomies might cause worsening of instability. In that case, occipito-cervical fusion using instrument should be performed. Alternatively, C1/2 fusion with transarticular screws can be also selected.

Problems in post-operative care
As already described, retro-odontoid mass is characteristically encountered in very elderly patients. In the present series, the mean age of the patients was 81.0 years. The oldest patient was 86 years old. Most difficult problems of post-operative care for these elderly patients are muscle atrophy and weakness in lower extremities due to less exercise before surgery and painful arthrosis deformans in lower extremities joints by aging process. Even after successful surgery, these factors prevent sufficient advance of rehabilitation. For this purpose, cooperative cares with neurosurgeons, orthopedists, and rehabilitation physicians are necessary for the improvement of their quality of life.

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Reviewer's comment: 高橋 宏（都立神経病院 脳神経外科）

本論文はretroodontoid disc herniaの治験例に関する英文の報告であり、興味深く読ませて頂いた。私自身の同じような環椎椎間板破壊例の手術経験では、最終的にRAあるいはchondromaという症例ばかりであり、retroodontoid disc herniaという診断例は経験していない。本論文に報告された例では、術前後外側診断に関連する記載が明確ではないため、これらの検討は術前あるいは術後の理診断等で済んでいるものと考えて読ませて頂いた。手術適応に関しては筆者等が引用している文献にもあるように、後方固定でmassが縮小する例も多いものと推測される。従って適応、手術法およびその評価は慎重さが必要である。手術アプローチについて、後方からの硬膜内経由による進入法が記載されている。術側に主座を有する環椎椎間板破壊に、我々は高齢者を含め、全て側方からの進入法で行っているが、従来に関しては後方進入と有意な差は考えている。また環椎椎間板側方進入であれば、硬膜を開けないでmassに到達出来ることが利点である。術前後の転位像のMRあるいはCTが無いので、本論文の引用例についての判断は難しいが、massが大きい程外側へ張り出し方が強いため側方からの硬膜外アプローチが容易となることを経験している。硬膜外の処理のみで手術が済めば、長期的に見ても機能性等による後遺症発生を未然に防ぐことが可能であると考える。何例か後方進入、硬膜内経由による椎板開窓ヘルニア摘出術を経験しているが、硬膜下スペースで術側にアプローチし、斜面の可及的温存を心がけるとともに、注意して前方の硬膜修復を施行している。これらのことは環椎椎間板によるヘルニア摘出手術に際して特に重要であることを改めて強調したい。

Reviewer's comment: 小柳 泉（札幌医科大学 脳神経外科）

歯突起後方への腫瘍を形成した高齢者8例の手術治療を報告し、その病名と治療方針について考察した論文で、手術治療には、後方からのC1、C2の椎弓切除後に、硬膜内に入り、後側方からの前方の硬膜を切開してヘルニアを摘出しています。著者らは摘出腫瘍の病理組織検査を詳細に行い、変性した軟骨組織と血管構造の増生を含んだ反応性の線維組織であることから、C2/3から脱出椎間板と頸椎の動きに伴う反応性組織の増殖が成分であると考えています。近年、歯突起後方の腫瘍形成は、チウマチ病変や腫瘍以外にも多くの病態が報告されています。本報告は、高齢者で特にみられる上位頸椎の病変について、放射線学的および病理学的詳細に検討しており、重要な知見を含んだ論文といえます。脱出椎間板組織が、このような症例の全てに関わっているのか、環椎椎間板と歯突起間の関節の変性や反応性肥厚などの程度関与しているのか等、病態を解明するための課題は残されており、今後も検討が必要と思われます。