Case Report
Regression of retro-odontoid pseudotumor following botulinum injection into cervical muscles
Masahiro Kawanishi, Hidekazu Tanaka, Yutaka Ito, Makoto Yamada, Kunio Yokoyama, Naokado Ikeda, Akira Sugie
Department of Neurosurgery, Takeda General Hospital, Kyoto City, Kyoto, Japan.
E-mail (*Corresponding author): Masahiro Kawanishi - mkawanis@takedahp.or.jp; Hidekazu Tanaka - bozu.hidekazu@nifty.com; Yutaka Ito - yutaka_727@yahoo.co.jp; Makoto Yamada - dsamako@mac.com; Kunio Yokoyama - kunio.yokoyama@ompu.ac.jp; Naokado Ikeda - naokado.ikeda@ompu.ac.jp; Akira Sugie - akira.sugie@ompu.ac.jp

ABSTRACT
Background: Posterior fixation of C1/2 has become more commonly performed to treat retro-odontoid pseudotumor (ROP). Here, we report a 60-year-old female with cervical dystonia (CD), whose ROP regressed and whose quadriplegia improved after a series of cervical intramuscular botulinum injections.

Case Description: A 60-year-old female with 30 years of CD newly presented with a progressive quadripareisis. When the MRI showed ROP compression the cervical spinal cord, she refused surgery and underwent multiple cervical muscular botulinum injections over the next 2 years. Following these injections, the patient's quadriplegia improved as the ROP regressed on subsequent MR studies.

Conclusion: Over a 2-year period, multiple cervical botulinum injections caused regression of a retro-odontoid cervical pseudotumor improvement in the patient's quadriplegia.

Keywords: Botulinum, Regression, Cervical dystonia, Retro-odontoid pseudotumor

INTRODUCTION
Retro-odontoid pseudotumor (ROP) is a nonneoplastic mass that forms behind the dentate ligament located dorsal to the axial vertebrae. Posterior fusion is the typical treatment of choice for ROP-related atlantoaxial instability (AAI). Here, a 60-year-old female with 30 years' duration of cervical dystonia (CD) newly developed a progressive quadriplegia attributed to ROP that markedly improved following repeated botulinum injections over a 2-year period.

CASE REPORT
Clinical presentation
A 60-year-old female, with CD for 30 years' duration, newly presented over the past 2 years with a progressive quadriplegia. On neurological examination, she was quadriplegic, exhibiting right more than left clumsiness of the hands and a spastic lower extremity paresis.
MR examination and botulinum injections

The cervical MR showed a low-intensity mass behind the odontoid process that compressed the cervical spinal cord and resulted in a high intrinsic cord signal [Figure 1]. The first choice for treatment was to inject botulinum into the cervical muscles; trapezius muscles, suboccipital muscles on the left, and sternocleidomastoid muscle on the right. As the patient refused, she underwent over the next 2 years, serial intramuscular botulinum injections. Over this interval, her quadriparesis and MR findings improved proportional to the extent of ROP regression. Two years later, the MR confirmed the reduction in size of the ROP mass that correlated with the regression of cord compression and the high intrinsic cord signal [Figure 2].

DISCUSSION

Acquired CD usually occurs between the ages of 20 and 60. It often leads to progressive cervical spinal degeneration most typically involving the C5/6 level. Interestingly, it rarely occurs above C2, and when it is present, which is seen in conjunction with AAI and myelopathy. Neeraja et al. reported that long-standing CD can lead to cord compression at the craniovertebral junction and result in myelopathy [Table1].[3] Watanabe et al., further, noted that a patient with cervical myelopathy due to ROP due to torticollis required an occipital atlantoaxial fusion that reduced ROP and improved symptoms.[5]

Posterior surgery for ROP

In general, posterior fixation of C1/2 has generally is typically required to treat ROP with AAI.[1,2] For the treatment of CD, botulinum is pre-injected into the cervical muscles to stop excessive cervical motion and is usually followed by a C1/C2 fusion.[4] Here, as the patient refused a C1/2 fusion surgery, it was elected, over a 2-year period, to perform successive intramuscular injections of botulinum into the cervical muscular to reduce the size of the ROP thus contributing to a reduction of cord compression/high cord signal, and improvement in the patient's myelopathic/quadriparietic deficit.

CONCLUSION

In a 60-year-old female, repeated intramuscular cervical botulinum injections over a 2-year period caused regression of ROP. This results in improvement of her quadriparietis in proportion to progressive resolution of her cord compression and high cord signal.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Goel A, Darji H, Shah A, Prasad A, Hawaldar A. Retro-odontoid and retro-C2 body pseudotumor, pannus, and/or

Table 1: Pertinent literature findings.

| Author   | Important findings                                           |
|----------|-------------------------------------------------------------|
| Neeraja  | Cervical dystonia leads to cord compression at the           |
|          | craniovertebral junction                                    |
| Traynelis| Botulinum is pre-injected into the cervical muscles followed |
|          | by a C1/C2 fusion for cervical dystonia                    |
| Watanabe | Occipital atlantoaxial fusion reduced retro-odontoid         |
|          | pseudotumor due to torticollis                             |
Kawanishi, et al.: Regression of retro-odontoid pseudotumor following botulinum injection

cyst. A study based on analysis of 63 cases. World Neurosurg 2021;151:e170-7.
2. Nakazawa T, Inoue G, Imura T, Miyagi M, Saito W, Shirasawa E, et al. Regression of retro-odontoid pseudotumor using external orthosis without atlantoaxial fusion: A case report. JBJS Case Connect 2019;9:e0329.
3. Neeraja K, Prasad S, Surisetti BK, Holla VV, Sharma D, Kamble N, et al. Cervical myeloradiculopathy and atlantoaxial instability in cervical dystonia. World Neurosurg 2021;146:e1287-92.
4. Traynelis VC, Ryken T, Rodnitzky RL, Menezes AH. Botulinum toxin enhancement of postoperative immobilization in patients with cervical dystonia. Technical note. J Neurosurg 1992;77:808-9.
5. Watanabe M, Iwashina T, Sakai D, Yamamoto Y, Mochida J. Cervical myelopathy with retroodontoid pseudotumor caused by atlantoaxial rotatory fixation and senile tremor. Tokai J Exp Clin Med 2009;34:39-41.

How to cite this article: Kawanishi M, Tanaka H, Ito Y, Yamada M, Yokoyama K, Ikeda N, et al. Regression of retro-odontoid pseudotumor following botulinum injection into cervical muscles. Surg Neurol Int 2022;13:321.