A surgical case of C1 arch stenosis: A case report and review of literature

Masatoshi Yunoki
Department of Neurosurgery, Kagawa Rosai Hospital, Marugame City, Kagawa, Japan.
E-mail: *Masatoshi Yunoki - yunomasato@me.com

*Corresponding author:
Masatoshi Yunoki,
Department of Neurosurgery,
Kagawa Rosai Hospital,
Marugame City, Kagawa, Japan.
yunomasato@me.com

INTRODUCTION

Only 11 prior cases of symptomatic cervical stenosis due to C1 posterior arch compression have been reported in the literature. Here, we describe a case of cervical myelopathy attributable to C1 arch stenosis that was readily resolved with operative decompression.

CASE PRESENTATION

A 74-year-old male presented with a 6-month history of gait disturbance, and clumsy hands that had exacerbated over the past 2 months; there was no history of trauma. He exhibited a spastic gait, bilateral hyperactive deep tendon reflexes in the upper/lower extremities, and bilateral positive Hoffmann’s signs. Plain radiographs showed marked narrowing of the cervical spinal canal, while the axial and sagittal computed tomography (CT) scans revealed a hypoplastic but intact posterior arch of the atlas [Figure 1a-c]. At the atlas level, the retrodental space was only 9.0 mm. Notably, there was no atlantoaxial instability on flexion and extension X-rays [Figure 1c and d]. The sagittal diameter of the spinal canal ranged from 11.5 to 13.8 mm at the other cervical spine levels. The magnetic resonance imaging revealed focal dural compression due to the hypoplastic posterior C1 arch along with a high T2-weighted intrinsic cord signal [Figure 2a and b]. The patient underwent a C1 laminectomy and resulting in an uneventful postoperative course [Figure 3a-c].
C1 anomaly and stenosis

It is well known that congenital anomalies occasionally involve the arch of atlas. In 2018, Hyun et al. reported 5.6% of such congenital lesions at C1 among their 3273 subjects.\textsuperscript{[2]} C1 stenosis with an intact posterior arch requiring treatment is rare; only 11 such cases have been reported in the literature [Table 1].\textsuperscript{[1,3-5,7-9,11]}

Clinical data for 12 total cases

With this case added, a total of 12 prior cases of C1 congenital stenosis resulting in myelopathy all occurred in middle-aged Asian males.\textsuperscript{[1,2]} Interestingly, most also had spinal canal stenosis at subaxial levels [Table 1].

The threshold of a canal diameter

In the 12 patients cited, the average sagittal canal diameter at the level of atlas in symptomatic patients with C1 stenosis was $<10$ mm. On routine cervical MR, compression at C1 atlas level is readily established. Further, physicians dealing with such C1 disorders may readily establish the diagnosis on lateral cervical X-rays with careful observation of narrowed AP diameter based upon the spinolaminar line [Figure 4].\textsuperscript{[6]}

| Author                  | Age/Sex Concomitant diseases | AP diameter (C1/C4) | Symptom                  | Treatment                                      |
|-------------------------|------------------------------|---------------------|--------------------------|------------------------------------------------|
| Sawada et al. (1989)\textsuperscript{[9]} | 38/M                          | 7/12.5 mm           | Myelopathy               | C1 laminectomy                                 |
| Komatus et al. (1993)\textsuperscript{[3]}  | 56/M                          | 7/11.7 mm           | Myelopathy               | Laminectomy (C1) laminoplasty C2               |
| Tokiyoshi et al. (1994)\textsuperscript{[11]} | 55/M                          | 8/13.0 mm           | Myelopathy               | Laminectomy C1 FMD                             |
| Noguchi et al. (1998)\textsuperscript{[6]}  | 81/M                          | 10/12 mm            | Stiff shoulder           | LaminoplastyC1                                 |
| Phan et al. (1998)\textsuperscript{[8]}    | 80/M                          | 8/- mm              | Myelopathy               | Laminectomy C1), FMD                           |
| Hoshimaru et al. (1999)\textsuperscript{[1]} | 81/M                          | 10/- mm             | Myelopathy               | Laminectomy C1                                 |
| Nishikawa et al. (2001)\textsuperscript{[4]} | 82/M                          | 12/- mm             | Myelopathy               | Laminectomy C1 laminoplasty C3-7               |
| Tokiyoshi et al. (1994)\textsuperscript{[11]} | 55/M                          | 8/13.0 mm           | Myelopathy               | Laminectomy C1 FMD                             |
| Noguchi et al. (1998)\textsuperscript{[6]}  | 81/M                          | 10/12 mm            | Stiff shoulder           | LaminoplastyC1                                 |
| Phan et al. (1998)\textsuperscript{[8]}    | 80/M                          | 8/- mm              | Myelopathy               | Laminectomy C1), FMD                           |
| Hoshimaru et al. (1999)\textsuperscript{[1]} | 81/M                          | 10/- mm             | Myelopathy               | Laminectomy C1                                 |
| Nishikawa et al. (2001)\textsuperscript{[4]} | 82/M                          | 12/- mm             | Myelopathy               | Laminectomy C1 laminoplasty C3-7               |
| Pascual-Gallego et al. (2014)\textsuperscript{[7]} | 5/M Down syndrome | 76/M                          | 9/- mm | myelopathy | Laminectomy C1 |

AP: Anteroposterior, FMD: Foramen magnum decompression
Treatment

For treating C1 stenosis, laminectomy or laminoplasty is typically effective.\textsuperscript{1,3-5,7-9,11} However, instability and the need for fusion should be carefully assessed at the atlantoaxial and craniocervical junctions.

If there is a large inferior facet angle and/or subaxial ankylosis, an anterior arch fracture may occur in up to 14.2% of cases follow a C1 laminectomy or laminoplasty initially be carefully considered.\textsuperscript{10}

CONCLUSION

Symptomatic myelopathy attributed to C1 stenosis may be readily reversed with a decompressive procedure.

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. Hoshimaru M, Hashimoto N. Myelopathy caused by developmental anomaly of the posterior arch of the atlas: Case report and review of literature. Spinal Surg 1999;13:53-8.
2. Hyun G, Allam E, Sander P, Hasiak C, Zhou Y. The prevalence of congenital C1 arch anomalies. Eur Spine J 2018;27:1266-71.
3. Komatsu Y, Shibata T, Yasuda S, Ono Y, Nose T. Atlas hypoplasia as a cause of high cervical myelopathy. Case report. J Neurosurg 1993;79:917-9.
4. Nishikawa K, Ludwig SC, Colón RJ, Fujimoto Y, Heller JG. Cervical myelopathy and congenital stenosis from hypoplasia of the atlas: Report of three cases and literature review. Spine (Phila Pa 1976) 2001;26:E80-6.
5. Noguchi A, Harada Y, Okabe S, Kohno T, Kamata K, Takahashi H. A surgical case of cervical canal stenosis caused by atlas hypoplasia in an elderly patient. No Shinkei Geka 1998;26:623-6.
6. Oshima Y, Kelly MP, Song KS, Park MS, Chuntarapas T, Vo KD, et al. Spinolaminar line test as a screening tool for C1 stenosis. Global Spine J 2016;6:370-4.
7. Pascual-Gallego M, Budke M, Villarejo F. Spinal stenosis at the level of atlas in a boy with Down syndrome. A case report and literature review. Neurocirugia (Astur) 2014;25:29-32.
8. Phan N, Marras C, Midha R, Rowed D. Cervical myelopathy caused by hypoplasia of the atlas: Two case reports and review of the literature. Neurosurgery 1998;43:629-33.
9. Sawada H, Akiyama I, Fukuyama H, Kameyama M, Koyama T. Marked canal stenosis at the level of the atlas. Neuroradiology 1989;31:346-8.
10. Shimizu T, Otsuki B, Fujibayashi S, Kumamoto S, Hijikata Y, Shimizu Y, et al. Incidence and risk factors of anterior arch fracture of the atlas following C1 laminectomy without fusion. Spine (Phila Pa 1976) 2018;43:667-74.
11. Tokiyoshi K, Nakagawa H, Kadota T. Spinal canal stenosis at the level of the atlas: Case report. Surg Neurol 1994;41:238-40.

How to cite this article: Yunoki M. A surgical case of C1 arch stenosis: A case report and review of literature. Surg Neurol Int 2021;12:71.