Author’s response to reviews

Title: A case report of revision occipital-cervical fusion after atlanto-axial instrumentation failure for neurofibromatosis type I

Authors:

Hayato Kinoshita (hayato@med.akita-u.ac.jp)
Naohisa Miyakoshi (miyakosh@doc.med.akita-u.ac.jp)
Takashi Kobayashi (takakoba826@gmail.com)
Toshiki Abe (tocci0302@gmail.com)
Kazuma Kikuchi (Kazuma.kiku@gmail.com)
Yoichi Shimada (seikei@doc.med.akita-u.ac.jp)

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Author’s response to reviews:

Hayley Henderson, Ph.D.
Associate Editor
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Re: Ms. No. BSUR-D-18-00346

Dear Prof. Henderson,

Thank you for your letter regarding our manuscript ‘A case report of revision occipital-cervical fusion after atlanto-axial instrumentation failure for neurofibromatosis type I’. We have responded to the reviewers’ comments in a point-by-point manner, and revised the manuscript accordingly. Please find enclosed the revised manuscript and our point-by-point responses to the reviewers’ comments. We would like to thank you and the reviewers for the time and effort taken to help us improve the quality of our manuscript.
We hope that the revised version of the manuscript is now acceptable for publication in BMC Surgery.

Sincerely yours,

Hayato Kinoshita, MD
Akita Kosei Medical Center
1-1-1 Nishibukuro, Iijima, Akita
011-0948, Japan
Tel: +81-18-880-3000; Fax: +81-18-880-3040
E-mail: hayato@med.akita-u.ac.jp

Point-by-Point Responses to Reviewer Comments (Ms. No. BSUR-D-18-00346)

Reviewer #1

Nesplon cables are not a standard way of fixation of such high degree of c1-2 subluxation, and the authors must acknowledge and present this case as a failure of that system rather than failure due to poor bone strength (of which there is no good evidence). Otherwise there is decent education value to this report.

Response: Thank you for this comment. As the reviewer indicated, nesplon cables may not be a standard method for fixation of c1-2 subluxation. Nevertheless, the utility of other polyethylene cables for C1-2 subluxation was reported in other studies. Thus, we have revised our manuscript to provide details on the potential benefits/limitations, as detailed below:

Discussion (lines 114-118): ‘Furthermore, Saito et al. demonstrated no surgical complications such as lamina fracture in 44 patients with atlanto-axial subluxation who received the modified Brooks technique using ultra-high molecular weight polyethylene cable [13]. However, there are no reports on the use of nesplon cable in NF1 patients. Of note, AAD recurred in our patient.'
Thus, further studies are required to confirm the utility of the nesplon cable in NF1 patients with AAD.

Reviewer #2

Could you please provide the x-rays and CT after index surgery. There are no x-rays which show the need for revision.

Response: Thank you for your comment. We apologize for not providing other CT images. However, we did not take any CT images after the first surgery, except for the one presented in Figure 3. Nevertheless, we have added the requested X-rays to Figure 3, and have revised our manuscript to provide the degrees of ADD, as detailed below:

Figure legend (lines 228-229): ‘Fig. 1 Preoperative imaging. a The cervical neural position showing expansion of the atlanto-dental interval. The upper atlantodental interval was 11.8 mm.’

Figure legend (lines 238-240): ‘Fig. 2 First operation. a Anterior–posterior radiography image at the first operation. b Sagittal radiography imaging at the first operation. The taping system and the iliac bone were used for fixation between C1 and C2. The upper atlantodental interval was 3.5 mm.’

Figure legend (lines 242-243): ‘Fig. 3 Recurrence of atlanto-axial dislocation. a Anterior–posterior radiography image. b Sagittal radiography imaging. The upper atlantodental interval was 8.5 mm.’

Figure legend (lines 247-249): ‘Fig. 4 Second operation. a The anterior–posterior radiography image at the second operation. b Sagittal radiography image at the second operation. A long fixation from the occipital bone to C4 was performed. The upper atlantodental interval was 2.8 mm.’

Reviewer#3:

GENERAL COMMENTS:

I have the pleasure to revise the case report about an occipital cervical fusion after atlanto-axial instrumentation failure for NF type 1.
Although the case is interesting and also well written, failure of atlanto-axial wiring for fixation is quite common. I think this manuscript has scientific value; authors should discuss the literature about failure wiring for atlanto-axial instability. Without this, the case did not add too much to the spinal literature.

Response: Thank you for your comment. We have added a discussion on the literature on failure wiring for atlanto-axial instability into the revised manuscript, as follows:

Discussion (lines 128-131): ‘It is also important to note that there are potential disadvantages when using cables for C1-2 fixation. For example, Yoshimoto et al. reported that C1-2 fixation in a hyperlordotic position caused subaxial kyphosis after surgery, and progression of kyphosis occasionally caused additional myelopathy [15].’

REQUESTED REVISIONS:

Adds little value to the current literature. Some concomitant literature review about failure C12 fusion with wiring techniques would be interesting.

Response: As the reviewer indicated, we have added a discussion on failure of C12 fusion with wiring techniques, as follows:

Discussion and Conclusion (lines 119-131): ‘Ito et al. compared the outcomes of 48 patients receiving posterior wiring using the McGraw or Brooks techniques with 28 patients receiving transarticular screw fixations using the Magarl technique [14], and found that nine of the 48 wiring patients did not obtain bone union, four of whom showed recurrence of atlantoaxial subluxation, while all patients receiving the Magarl technique obtained bone union. The authors suggested that even if posterior wiring was performed in AAD patients, sufficient postoperative rest periods and external fixation (e.g., Halo) would provide the best bone union and prevent recurrence of atlantoaxial subluxation. Our case wore a Philadelphia brace continuously after the first operation, but AAD recurrence occurred. Thus, it may have been useful to perform transarticular screw fixation (e.g., using the Magarl technique) at the first surgery, taking care to look for abnormal vertebral arteries. It is also important to note that there are potential disadvantages when using cables for C1-2 fixation. For example, Yoshimoto et al. reported that C1-2 fixation in a hyperlordotic position caused subaxial kyphosis after surgery, and progression of kyphosis occasionally caused additional myelopathy [15].’
ADDITIONAL REQUESTS/SUGGESTIONS:

I would expand the introduction - discussing the types of spinal deformity in NF, as well the incidence of cervical instability in this group of patients. A comprehensive literature is necessary.

Considering case presentation - the intra-articular spacers were peek cages? titanium? used off label from cervical spacers? please clarify. Discuss the nuances of cervical fixation in NF - the bone anomalies, vertebral artery abnormal course, etc.

Response: Thank you for your comment. The incidence of cervical instability in NF 1 patients is very low, with only seven cases previously reported (lines 47-50). We have added text on the types of spinal deformity in NF into the revised manuscript, as follows:

Background (lines 45-47): ‘In a study of 102 patients with neurofibromatosis, patients with dystrophic changes (defined as dystrophic type) exhibited a rapid progression of spinal deformities, while conservative treatments were ineffective [2].’

Discussion and Conclusion (lines 138-143): ‘The only obvious dystrophic change in our case was expansion of the left C2/3 foramina, with no evidence of other dystrophic changes such as rib penciling, scalloping of the vertebral body, costotransverse spindling, or increased width between each pedicle. Furthermore, there was no evidence of the rapid progressive spinal deformities reported in dystrophic type neurofibromatosis [2]. Thus, we suggest that the fracture of the C1 lamina in our case was not caused by rapid spinal deformity.’

We have also added details of the intra-articular spacers used, as follows:

Case Presentation (lines 79-80): ‘We then introduced two intra-articular titanium spacers (KiSCO, Kobe, Japan) for fixed bilateral atlanto-axial joints.’

Further, we have added a discussion on the nuances of cervical fixation in NF, as follows:

Case Presentation (lines 77-79): ‘We reoperated using a long posterior fusion. Because of the existing abnormal vertebral artery inside of the C1 lamina, we gently removed scar tissue using a subperiosteal approach, and revealed the C1/2 facet under direct vision.’

Case Presentation (lines 86-87): ‘To avoid stress concentration and refractures, sublaminar taping was performed at C2, C3, and C4 using nesplon cables.’
Reviewer#4:

Title and discussion is well written Case presentation need more to present Justify lines need English correction work more on reference blood loss need to write in mls not grms

Response: Thank you for your comment. We have revised the text on blood loss, as follows:

Case Presentation (line 71-72): ‘Furthermore, we tied two nesplon cables to the sublamina of C1 and C2 according to the Brooks technique.’

Case Presentation (line 73): ‘The operative time was 1 h 35 min, and bleeding was <50 ml.’

Case Presentation (lines 88-89): ‘The operative time was 3 h 56 min, and bleeding was 425 ml.’

We have also revised our manuscript to clarify the details of the second operation, as follows:

Case Presentation (lines 77-79): ‘We reoperated using a long posterior fusion. Because of the existing abnormal vertebral artery inside of the C1 lamina, we gently removed scar tissue using a subperiosteal approach, and revealed the C1/2 facet under direct vision.’

Case Presentation (lines 86-87): ‘To avoid stress concentration and refractures, sublaminar taping was performed at C2, C3, and C4 using nesplon cables.’