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Surgical treatment of paediatric unilateral tinnitus due to cochleovestibular nerve compression by intrameatal anterior inferior cerebellar artery (AICA) loop

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Key words:

Cochleovestibular nerve, intrameatal anterior inferior cerebellar artery (AICA),
Tinnitus, Pediatrics, Surgery

Running title: surgical treatment of pediatric surgery

1 **Surgical treatment of paediatric unilateral tinnitus due to cochleovestibular**
2 **nerve compression by intrameatal anterior inferior cerebellar artery (AICA) loop**

3 **Abstract**

4 **Objectives:** Cochleovestibular nerve compression syndrome due to intrameatal
5 anterior inferior cerebellar artery (AICA) that causes tinnitus is an extremely rare
6 condition with special therapeutic challenges and implications. Here we present the
7 first surgically treated paediatric case of tinnitus showing microvascular conflict of
8 cochleovestibular nerve and intrameatal AICA loop.

9 **Methods:** A paediatric case of tinnitus is described and a surgical technique is
10 elaborated with video material. A Pubmed literature search was performed to identify
11 surgically treated paediatric cases of intrameatal compression of eighth nerve with
12 AICA loop.

13 **Results:** Tinnitus was completely resolved and hearing was intact immediately after
14 surgical decompression. The patient was free from further symptoms after a short
15 follow up of 12 weeks. Microsurgical decompression, including opening of IAC and
16 transposition of AICA, was an effective treatment option in this case.

17 **Conclusion:** Decompression of the cochleovestibular nerve including opening of IAC
18 and transposition of AICA appears to be an effective treatment option for cases with
19 radiologically confirmed intrameatal vascular compression of eighth nerve causing
20 unilateral pulsatile tinnitus.

21 **Key words:**

22 Cochleovestibular nerve, intrameatal anterior inferior cerebellar artery (AICA),
23 Tinnitus, Paediatrics, Surgery

24 **Introduction**

25 Continuous or pulsatile tinnitus may be due to variety of pathologies, such as
26 tumours of cerebellopontine angle and vascular pathologies, including dural AV
27 fistula (DAVF), intracranial aneurysms, atherosclerotic diseases of intracranial
28 vessels, and medical conditions with high cardiac output (such as hypertension,
29 anaemia or thyreotoxicosis) (3, 4, 7, 10). Vascular conflict with eighth cranial nerve is
30 another possible cause of tinnitus. Neurovascular conflicts of the trigeminal, facial,
31 and glossopharyngeal nerves are relatively common and have an established
32 therapeutic strategy. Tinnitus caused by cochleovestibular nerve compression
33 syndrome due to intrameatal anterior inferior cerebellar artery (AICA) is an extremely
34 rare condition. More than 500 cases of microvascular decompression surgery of
35 eighth nerve have been described to date in the literature. However, there are less
36 than 10 surgically treated cases of tinnitus due to intrameatal anterior inferior
37 cerebellar artery (4). None of these cases were paediatric. Surgical treatment of such
38 cases is challenging and the existing evidence is minimal due to few surgically
39 treated cases. Here, we present a paediatric case of microvascular conflict of
40 cochleovestibular nerve with intrameatal AICA treated surgically in our centre
41 (Helsinki live course). Opening of the internal auditory canal (IAC) and microsurgical
42 decompression and mobilization of AICA relieved tinnitus without any other focal
43 neurological deficit.

44 **Materials and methods**

45 **Case report**

46 A 15-year-old female presented with a history of right-sided intractable tinnitus of
47 over 5 years duration. On examination the patient was alert and oriented to all

48 qualities. Visual field was intact and pupil size was normal and reactive to light. The
49 patient showed normal hearing and facial nerve function. MRI scan showed an
50 approximately 1-cm long arterial loop of AICA coming into contact with the
51 cochleovestibular nerve (Fig 1 A, B, C, D). There was no other obvious pathology in
52 the cerebellopontine angle. Due to failure of conservative treatment (including
53 counselling) patient desired surgical treatment.

54 **Literature review**

55 A literature search for surgically treated cases of paediatric tinnitus due to intrameatal
56 compression of eighth nerve was performed using Pubmed, Ovid Medline and
57 Scopus database. We used the PRISMA checklist as a guide to achieve accepted
58 standards for reporting systematic reviews. The first author performed all steps and
59 discussed with senior author. We used the keywords “tinnitus” in combination with
60 “microvascular decompression” and “intrameatal AICA”. A total of four articles with
61 nine case reports were found on surgically treated decompression of intrameatal loop
62 of AICA. Paediatric cases were not found.

63

64 **Results**

65 **Surgical decompression and operative findings**

66 Surgery was performed via a retrosigmoid approach using OPMI PENTERO 900
67 microscope (Carl-Zeiss, Germany). Dura was opened in a curved fashion. The basal
68 cistern was opened to release CSF. The AICA loop entering in the internal auditory
69 canal (IAC) was identified and dissected from arachnoid. The IAC was drilled and the
70 AICA loop and eighth nerve was exposed. Microvascular decompression was
71 performed and the AICA loop was mobilized and fixed with dura using a 10-0 suture
72 (Fig. 2. A, B, C, D, and video 1). The IAC was closed using fibrin glue and muscle
73 fascia. Dura was closed in a water-tight fashion.

74

75 **Post-operative outcome:**

76 Tinnitus was completely relieved immediately after the surgery without any new focal
77 neurological deficit. No other complications occurred. The patient was discharged
78 from the hospital after 4 days. The patient was completely symptom free after a
79 follow up of 12 weeks.

80 **Discussion**

81 Microvascular decompression surgery is a standard treatment for trigeminal
82 neuralgia, glossopharyngeal neuralgia, and hemifacial spasm (1, 6, 8). However,
83 microvascular decompression with ontological symptoms (including tinnitus, vertigo,
84 and hearing loss) is debatable as in multiple cases the arterial conflict with eighth
85 nerve is a normal variant. However, in the case of pulsatile tinnitus there is an 80-fold
86 greater chance of microvascular conflict than in non-pulsatile tinnitus, thus showing a

87 relatively strong association (2, 5). Microvascular decompression in cases of tinnitus
88 is less successful in comparison to trigeminal neuralgia, glossopharyngeal neuralgia,
89 or hemifacial spasm. However, the cases described in literature are a mixture of
90 various neurovascular conflicts with the eighth nerve. A meta-analysis of over 500
91 surgically treated cases of cochleovestibular nerve decompression revealed only nine
92 cases of tinnitus caused by intrameatal loop (4). Interestingly, tinnitus was
93 successfully treated in all nine cases with a decompression and mobilization of AICA
94 loop (4). To the best of our knowledge, none of these cases were paediatric. We
95 demonstrated a successfully treated paediatric case of tinnitus caused by the
96 intrameatal loop of AICA. Most paediatric cases of tinnitus are treated conservatively.
97 However, our patient with a pulsatile unilateral tinnitus failed over several years all
98 types of non-surgical treatment options. Although there are only a few cases of
99 surgically treated tinnitus due to intrameatal AICA loop, surgical treatment resolved
100 tinnitus in all cases as in our case. Hence, surgery could be a valid option for
101 selected cases with unilateral pulsatile tinnitus and radiologically confirmed
102 intrameatal vascular conflict (4, 9, 11). Proper patient selection is however crucial.

103

104

105

106 **Conclusion**

107 Decompression of the cochleovestibular nerve including opening of IAC and
108 transposition of AICA seems to be an effective treatment option for cases with
109 radiologically confirmed intrameatal vascular compression of eighth nerve causing
110 unilateral pulsatile tinnitus.

111

112

113 Figure legends

114 Figure 1: MRI scan showing microvascular conflict between AICA loop and
115 vestibulocochlear nerve in IAC (A, B, C, D).

116 Figure 2: Intraoperative images showing different steps of neurovascular
117 decompression (A, B, C, and D). Upper arrow shows AICA loop; lower arrow shows
118 the VIII nerve (D) after opening internal auditory canal.

119 Video 1: Decompression surgery demonstrating different steps of surgery and
120 surgical anatomy.

121

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124 base fellowship at the Department of Neurosurgery in Helsinki University Hospital.

125 Conflict of interest

126 The authors certify that they have no affiliations.

127 Ethical approval

128 All procedures performed in this study involving human
129 participants were in accordance with the ethical standards of the research committee
130 of the University of Helsinki and with the 1964 Helsinki declaration and its later
131 amendments or comparable ethical standards.

132 Informed consent

133 The parents of the child provided informed consent.

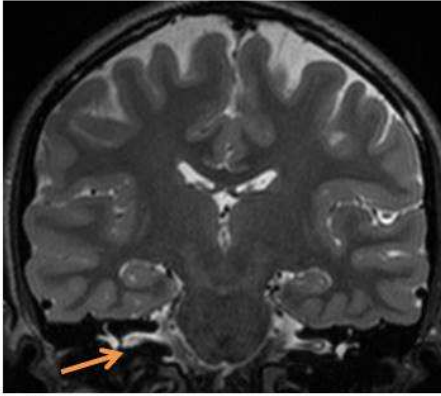
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136 References:

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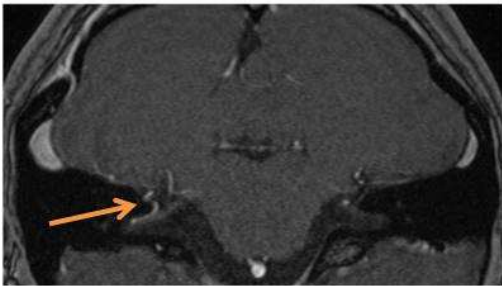
- 138
139 1. Capelle HH, Brandis A, Tschan CA, Krauss JK: Treatment of recurrent trigeminal neuralgia due
140 to Teflon granuloma. **J Headache Pain** 11:339-344, 2010.
- 141 2. Chadha NK, Weiner GM: Vascular loops causing otological symptoms: a systematic review
142 and meta-analysis. **Clin Otolaryngol** 33:5-11, 2008.
- 143 3. Daneshi A, Hadizadeh H, Mahmoudian S, Sahebjam S, Jalesi A: Pulsatile tinnitus and carotid
144 artery atherosclerosis. **Int Tinnitus J** 10:161-164, 2004.
- 145 4. Esposito G, Messina R, Carai A, Colafati GS, Savioli A, Randi F, De Benedictis A, Cossu S,
146 Fontanella MM, Marras CE: Cochleovestibular Nerve Compression Syndrome Caused by
147 Intrameatal Anterior Inferior Cerebellar Artery Loop: Synthesis of Best Evidence for Clinical
148 Decisions. **World Neurosurg** 96:556-561, 2016.
- 149 5. Gultekin S, Celik H, Akpek S, Oner Y, Gumus T, Tokgoz N: Vascular loops at the
150 cerebellopontine angle: is there a correlation with tinnitus? **AJNR Am J Neuroradiol** 29:1746-
151 1749, 2008.
- 152 6. Jannetta PJ: Neurovascular compression in cranial nerve and systemic disease. **Ann Surg**
153 192:518-525, 1980.
- 154 7. Kim DK, Shin YS, Lee JH, Park SN: Pulsatile tinnitus as the sole manifestation of an internal
155 carotid artery aneurysm successfully treated by coil embolization. **Clin Exp Otorhinolaryngol**
156 5:170-172, 2012.
- 157 8. Laha RK, Jannetta PJ: Glossopharyngeal neuralgia. **J Neurosurg** 47:316-320, 1977.
- 158 9. Pirayesh Islamian A, Lutjens G, Krauss JK: Microvascular decompression of the eighth cranial
159 nerve for unilateral pulsatile tinnitus. **Clin Neurol Neurosurg** 117:102-106, 2014.
- 160 10. Sila CA, Furlan AJ, Little JR: Pulsatile tinnitus. **Stroke** 18:252-256, 1987.
- 161 11. Zhang L, Yu Y, Yuan Y, Xu J, Xu X, Zhang J: Microvascular decompression of cochleovestibular
162 nerve in patients with tinnitus and vertigo. **Neurol India** 60:495-497, 2012.
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164

MRI Scan showing AICA loop compressing VIII nerve in IAC

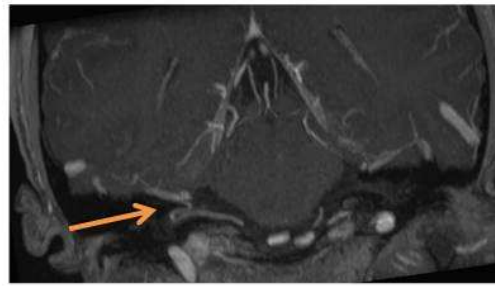
A



B



C



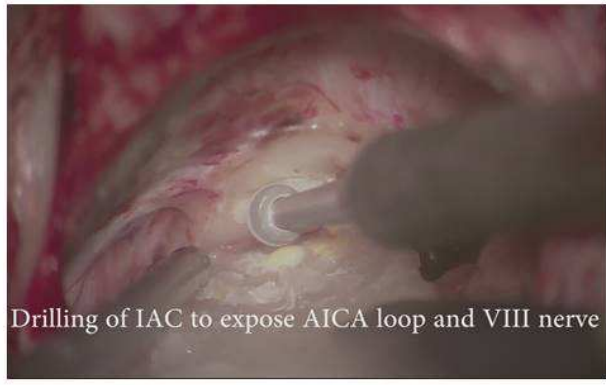
D

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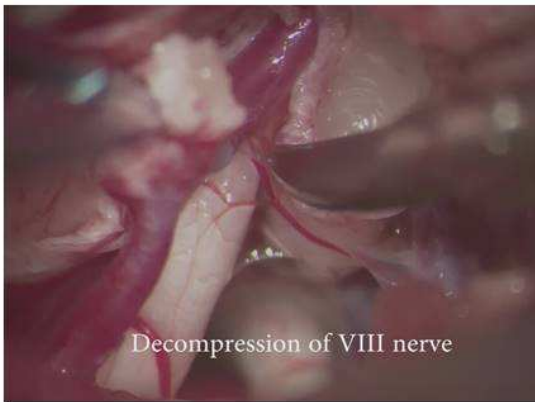
Dissection of AICA at dural attachment

A



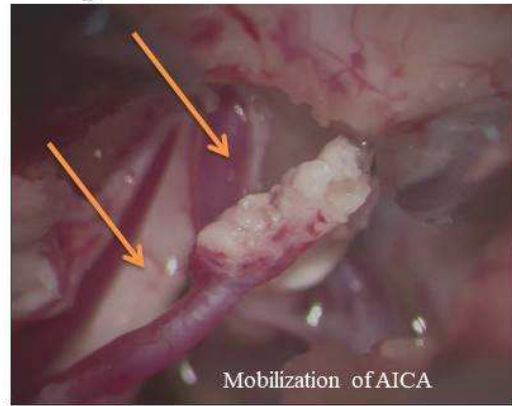
Drilling of IAC to expose AICA loop and VIII nerve

B



Decompression of VIII nerve

C



Mobilization of AICA

D

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Abbreviations:

Cochleovestibular nerve, intrameatal anterior inferior cerebellar artery (AICA),

Tinnitus, Pediatrics, Surgery

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