

## A Case of Intermediate Nerve Neuralgia with a Successful Outcome with Neurectomy

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### Abstract

Intermediate nerve neuralgia (INN), a relatively rare disease, is caused by vascular compression of the intermediate nerve. The main symptoms include pain in the external auditory canal, which may be accompanied by lacrimation and taste disturbance. Recently, surgery has been performed using microvascular decompression. We report a case of a 30-year-old female patient who presented with paroxysmal electric shock pain in the external auditory canal lesion. The MRI showed no vessel compression on the trigeminal nerve but the vessels ran between the facial nerve and the vestibulocochlear nerve, suggesting intermediate nerve compression. Surgery was performed with a diagnosis of INN. Intraoperative findings showed that the intermediate nerve was transected as the displacement of the responsible blood vessel would cause stretching of the internal auditory artery exiting the vessel compressing the intermediate nerve, resulting in hearing loss. Paroxysmal electric pain in the external auditory canal disappeared without the appearance of new postoperative neurological deficits.

Three-dimensional magnetic resonance cisternogram/magnetic resonance angiogram fusion images were helpful in surgical decision-making. In some cases, vascular displacement may be unsafe and difficult, and intermediate nerve transection should be considered in the procedure.

**Keywords:** Intermediate Nerve Neuralgia; Neurectomy; Microvascular Decompression; Magnetic Resonance Imaging

### Abbreviations

INN: Intermediate Nerve Neuralgia; TN: Trigeminal Neuralgia; HFS: Hemifacial Spasm; MRI: Magnetic Resonance Imaging; MRA: Magnetic Resonance Angiogram; MRC: Magnetic Resonance Cisternogram; MVD: Microvascular Decompression

### Introduction

Intermediate nerve neuralgia (INN) is caused by vascular compression of the nerve. There have been reports in the recent years on the usefulness of microvascular decompression (MVD) as a reasonable approach for INN [1-4]. However, in some cases, nerve decompression by replacing offending vessels may be difficult in

the presence of a perforating branch to the brain stem. In addition, ischemia may occur because of stretching of the inner ear arteries due to vascular displacement, which may lead to hearing loss [1]. We report a case in which a neurectomy was done to achieve a good result because sufficient nerve decompression could not be performed using the vascular displacement technique. Therefore, neurectomy was used as a treatment option for INN.

### Case Presentation

We present a case of a 30-year-old woman who began experiencing paroxysmal electric pain in the right external auditory canal without any specific trigger a year ago. The pain radiated to the

area of the posterior auricular region and the third branch of the trigeminal nerve. The patient was diagnosed with trigeminal neuralgia by her family physician. Her condition was controlled with pregabalin, but she was referred to our department because the effect of pregabalin reduced.

Preoperative three-dimensional magnetic resonance cisternogram/magnetic resonance angiogram (3D-MRC/MRA) fusion image (Figure 1) showed no apparent compressed vessels around the trigeminal nerve (data not shown). The meatal loop of the anterior inferior cerebellar artery (AICA) running between the facial nerve and the vestibulocochlear nerve was identified (Figure 1). The AICA loop compressed the intermediate nerve in the cisternal portion, thus diagnosed as INN. The intermediate nerve was not identified on MRI. Informed consent was obtained from the patient to perform an MVD or neurectomy of the intermediate nerve.

Surgery was performed using a retrosigmoidal approach in the lateral recumbent position. Intraoperative auditory brainstem response was continuously monitored. A 6-cm linear skin incision was done in the retroauricular region, and craniotomy was performed until the sigmoid sinus was partially exposed. The dura mater was opened, cerebrospinal fluid was aspirated, the cerebellum was gently retracted, and the arachnoid membrane around the lower cranial nerves was incised. Then, the surgical field was transferred to the VII/VIII complex, and the entire length of the nerves was identified. A thin nerve, an intermediate nerve, ran between the facial nerve and the vestibulocochlear nerve (Figure 2A). The AICA running between the facial nerve and the vestibulocochlear nerve from the cranial side formed a loop and compressed the intermediate nerve in the cisternal portion (Figure 2A). This was consistent with the preoperative findings on 3D-MRC/MRA fusion imaging (Figure 1). First, the responsible vessel, the AICA, was dissected, mobility was obtained, and vascular displacement was attempted. However, sufficient displacement could not be achieved because of concerns on stretching the internal auditory artery, and decompression to the intermediate nerve could not be obtained. The decision to perform the neurectomy of the intermediate nerve was made (Figure 2B).

The postoperative course was uneventful, with no new neurological deficits. The INN was resolved, and the patient was discharged 7 days after surgery.

**Figure 1:** Preoperative 3DMRC/MRA fusion image.

AICA meatal loop (arrowhead) has entered between the facial nerve (arrow) and vestibulocochlear nerve (double arrows), compressing both nerves.

**Figure 2:** Surgical view

A: Before intermediate neurectomy.

The intermediate nerve (arrowhead) runs between the facial nerve (arrow) and the vestibulocochlear nerve (double arrows) and is compressed by the AICA.

B: After intermediate neurectomy

The intermediate nerve is disconnected (double arrowheads).

## Discussion

It is a relatively rare disorder in which INN is the chief complaint of paroxysmal electric pain in the external auditory canal, which radiates to the posterior auricular region and presents with lacri-

mation and taste disturbances [5]. Since the trigeminal and glossopharyngeal nerves are involved in the sensation of the external auditory canal, the diagnosis of INN can be difficult [1]. We previously have reviewed 12 of 16 cases (75%) in which compression occurred at the cisternal lesion rather than at the root exit zone of the nerve [1]. In addition, 75% of the cases in our review showed that the responsible vessel was the AICA [1]. The treatment methods described were nerve transection and MVD, in which the responsible blood vessel is displaced and the intermediate nerve is decompressed. Both have been shown to have good results [1-4,6]. In the present case, the meatal loop of the AICA running between the facial nerve and the vestibulocochlear nerve compressed the intermediate nerve in the cistern.

The intermediate nerve is reported to be challenging in the operative field because it is located dorsal to the vestibulocochlear nerve [7]. There are concerns that the vestibulocochlear nerve may be strained because of the visibility of the intermediate nerve, resulting in hearing loss as a complication [7]. It has been reported that complications occur more frequently in cases of MVD for INN in which the intermediate nerve is visualized than in those in which it is not. Furthermore, the authors noted that there is no need to confirm intermediate nerve intraoperatively forcibly [7]. In this case, the intermediate nerve was easily visible because the AICA loop from the dorsal side compressed it. Meanwhile, whenever it is necessary to sever the intermediate nerve, the entire nerve needs to be visible; however, care must be taken to avoid the appearance of complications.

Since INN is due to vascular compression, decompression by vascular displacement is the first choice. Recent reports of surgery for INN mainly include MVD [1-4]. In cases such as the present case, in which ischemia is present in the responsible vessel to the brainstem or in the region of the internal auditory artery and there is a possibility of brainstem infarction or hearing loss, severing the nerve without displacing the vessel may be an option.

Dry eyes and taste disorders are expected owing to intermediate nerve transection. Peris-Celda M., *et al.* found no apparent complications in nine cases of intermediate nerve transection [8]. A case report of a patient who underwent MVD for facial spasm and had the intermediate nerve severed showed no complications [9]. In this case, there were no apparent postoperative complications.

3D-MRC/MRA fusion images are used for preoperative diagnosis of trigeminal neuralgia and hemifacial spasm, evaluation of the anatomical relationship of the nerves and responsible vessels, and identification of any nerve deformities [10]. 3D-MRC/MRA fusion imaging was conducted in the present case, but no apparent neurovascular conflict was found around the trigeminal or glossopharyngeal nerve. Only compression in the cisternal portion of the CN VII/VIII complex by the AICA was observed. The intermediate nerve was not identified. The findings were informative for surgical decision-making. The intermediate nerve is thin and difficult to detect using the currently available technology. Therefore, further research is needed to conduct more detailed and accurate neuroimaging diagnoses.

The problem is how to identify the intermediate nerve. In a previous report on the transaction of the intermediate nerve, the nerve was identified and transected based on the anatomical findings of the intermediate nerve. In our case, the intermediate nerve was identified as a thin nerve running between the facial and vestibulocochlear nerves from the brain stem and was transected. We obtained a good result, but intraoperative monitoring of the intermediate nerve is an important issue for the future.

## Conclusion

We reported an INN case where neurectomy was performed with excellent results.

Although MVD seems reasonable in surgery for INN, neurectomy should be considered since it may be difficult to displace the offending vessels.

## Conflict of Interest

The authors declare that they have no conflict of interest.

## Ethics Approval

The Ethical Committee of the International University of Health and Welfare approved all procedures used in this research.

## Submission Statement

This original manuscript has not been submitted elsewhere in part or whole.

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