# Percutaneous balloon compression for trigeminal neuralgia due to the primitive trigeminal artery: A case report and review of the literature

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## Abstract

Trigeminal neuralgia (TN) is often caused by vascular compression of the trigeminal nerve cisternal segment. TN due to the primitive trigeminal artery formed by developmental variants of cerebral vessels is rare. We report here a 59-year-old male with TN whose preoperative MRI showed a primitive trigeminal artery that compressed the trigeminal nerve, and microvascular decompression (MVD) was performed to separate the vessel from the trigeminal nerve cisternal segment. The pain was relieved after MVD, but it recurred two months later. MRI reexamination showed that although MVD relieved the vascular compression of the trigeminal nerve cisternal segment, the nerve in the Meckel's cave was still compressed by PTA, which was thought to be the cause of TN recurrence. Due to the limitations of the surgical microscope view, completing the vascular decompression at the Meckel's cave is challenging. So percutaneous balloon compression was performed. After the procedure, the TN subsided. The patient remained pain free at one year follow-up. To our knowledge, this is the first case of primitive trigeminal artery-associated TN treated by percutaneous balloon compression. This case suggests that percutaneous balloon compression may be considered for TN caused by compression of the primitive trigeminal artery when MVD is difficult to perform.

Keywords: Trigeminal neuralgia, primitive trigeminal artery, microvascular decompression, percutaneous balloon compression

## INTRODUCTION

Trigeminal neuralgia (TN) is a paroxysmal, electric-shock pain that occurs within the distribution range of one or more branches of the trigeminal nerve.1 Vascular compression of the trigeminal nerve cistern segment is a common cause.<sup>2</sup> The offending vessels are mainly the superior cerebellar artery, the anterior inferior cerebellar artery, the vertebrobasilar trunk or peripheral veins.3 Primitive trigeminal artery (PTA) is a developmental variation of rare abnormal anastomosis between the carotid artery and basilar artery.<sup>4</sup> PTA causing TN is rare, and were usually treated by microvascular decompression (MVD).5-10 However, no cases of percutaneous balloon compression (PBC) treating PTA-associated TN have been reported. Here we

report a patient with PTA-related TN who was first treated by MVD but recurred shortly afterwards. He then underwent PBC with good results. We also reviewed the published literature of PTA associated with TN and their treatment.

## **CASE REPORT**

A 59-year-old male presented with an 8-year history of lightning-like paroxysmal pain in the right trigeminal V2 and V3 distribution, often induced by touch, drinking, and eating. He had no abnormal neurological symptoms other than facial pain. The patient was initially treated with carbamazepine orally but changed to oxcarbazepine due to allergy. The effect of oxcarbazepine on pain relief gradually decreased 4 months before admission.

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MRI examination showed that an artery originated from the cavernous sinus segment of the right internal carotid artery (ICA) that ran laterally, passing through the cavernous sinus into the right Meckel's cave (MC), and then turned anteromedially into the cistern at the entrance of MC. During this path, the right MC and part of the cistern trigeminal nerve were compressed (Figure 1a, b), and the artery was identified as PTA. After preoperative preparation, the patient underwent MVD through the right retrosigmoid approach. The artery in the cisternal segment of the trigeminal nerve was isolated during the operation, and the symptoms were relieved after the operation. However, TN recurred 2 months later. MRI reexamination showed that a Teflon piece placed during MVD had separated the trigeminal nerve cisternal segment from an artery originated from PTA, but the trigeminal nerve in the MC was still compressed by PTA, likely to result in TN recurrence (Figure 1c, d). Since the MC could not be visualized due to view limitation in either the microscope or neuroendoscope, decompression operation could not be performed. Therefore, he was treated with PBC.

Because PTA entered the MC through cavernous sinus, avoiding PTA injury during puncture was vital to the success of PBC. 3D Slicer was used

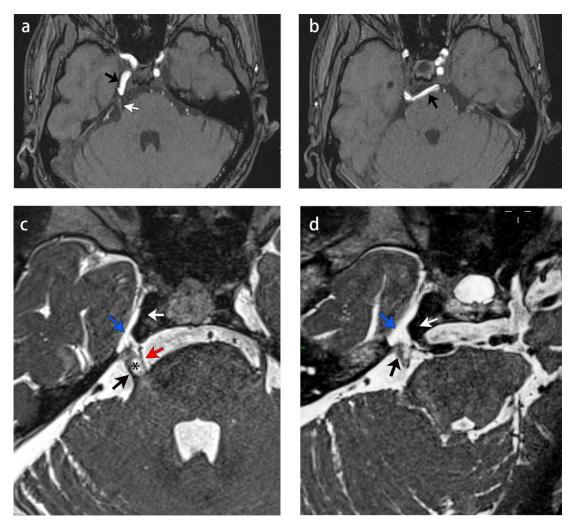


Figure 1. Axial TOF-3D sequence of MRI examination before MVD (a, b) and axial T2-SPACE sequence of MRI reexamination before PBC (c, d) PTA (black arrow) originates from the cavernous sinus segment of the right ICA and passes through the MC, compressing the right MC and part of the cistern trigeminal nerve (white arrow) (a). PTA turns anteromedially into the cistern at the entrance of the MC (b). A Teflon piece (asterisk) has separated the cistern trigeminal nerve (black arrow) from the small artery (red arrow) from PTA (c). The dilated PTA (white arrow) trunk in the MC (blue arrow) also compresses the trigeminal nerve (d).

to simulate the direction and depth of puncture before the operation (Figure 2a). The appropriate insertion depth was 8.7cm when just entering the foramen ovale. During PBC, the depth of the puncture needle was controlled under the guidance of X-ray according to the preoperative simulation. When the needle was in place, a balloon catheter was put in. After injecting 0.7ml of contrast agent, X-ray imaging showed that the balloon expanded and presented a typical "pear" (Figure 2b). With 5 minutes of pressure, the balloon was removed. The patient's pain disappeared immediately after PBC with mild facial numbness. The pain-free state remained at one year follow-up.

## DISCUSSUION

PTA is a very rare anastomosis between the

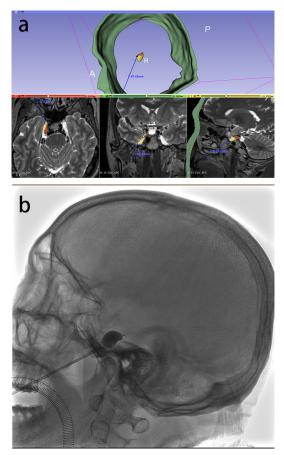


Figure 2. Preoperative puncture simulation of PBC and the left intraoperative X-ray of the patient during PBC 3D Slicer is used to simulate the direction and depth of puncture during PBC. When the insertion depth to the MC (yellow part) is about 8.7cm, the PTA (red part) will not be damaged (a). The balloon showed a typical "pear" (b).

ICA and the basilar artery (BA), which can be seen in 0.1%~1% of all cerebrovascular angiography.<sup>4</sup> At the 4mm embryo stage, the paired longitudinal neural arteries are supplied by the carotid arteries via four important arterial anastomoses, namely, the trigeminal artery, the auricular artery, the sublingual artery, and the anterior interatlas artery. Subsequently, an anastomosis forms between the distal ICA and corresponding longitudinal neural artery, which becomes the posterior communicating artery. Meanwhile, the presegmental arteries and proatlantal intersegmental artery regress and obliterate.4,5 Incomplete arterial degeneration or abnormal development of the ipsilateral posterior communication artery may result in continuous carotid basilar anastomosis, such as PTA.11

PTA that originates from the posterolateral part of the cavernous sinus of ICA and runs underneath the abducens nerve and continue caudally between the trigeminal and abducens nerves to join the distal BA is classified as the lateral type. PTA that originates from the posterior medial part of the cavernous sinus of ICA and runs through the dorsum sellae and perforate the dura mater near the clivus is classified as the medial type.<sup>4</sup> For the lateral type is closer to the trigeminal nerve, oculomotor nerve, trochlear nerve and abducent nerve anatomically, it is more likely to compress them, causing TN, oculomotor nerve paralysis, trochlear nerve paralysis and abducent nerve paralysis.<sup>12</sup> In this case, the patient had a lateral type of PTA, which passed through the MC and compressed the trigeminal nerve during its course (Figure 1), thus resulting in TN.

Up to 2022, there have been 14 reports of the treatment of PTA-associated TN consisting of 21 patients. (Table 1) In 3 patients, the compression was caused by the superior cerebellar artery, the anterior inferior cerebellar artery or the posterior inferior cerebellar artery from PTA without PTA itself being involved. PTA was identified as the responsible vessel in the remaining 18 patients (85.7%). Among the 18 patients, 14 (77.8%) underwent MVD to isolate the vessel compressing the cisternal segment of the trigeminal nerve, 3 underwent interventional embolization due to arteriovenous fistula or aneurysm, one was treated by gamma knife after rejecting MVD. The pain of 14 patients underwent MVD was relieved immediately after the surgery, and there was no recurrence during the follow-up, indicating good effect from MVD. The relevant information of patients is shown in Table 1. According to reports published to-date, MVD has been used to treat

| Literature                              | Number of<br>case(s) | Age and sex                           | Affected side<br>and range of<br>pain | History of<br>symptoms<br>(years) | Offending vessels | Therapy        | Outcome of therapy  |
|---|----------------------|---------------------------------------|---------------------------------------|-----------------------------------|-------------------|----------------|---|
|   |                      | 75, female                            | left, V2, V3                          | 3                                 | PTA               | MVD            |   |
|   |                      | 77, female                            | left, V2                              | 20                                | PTA               | MVD            |   |
|   |                      | 53, female                            | left, V2                              | 4                                 | PTA, SCA, AICA    | MVD            |   |
| Monito of all 10005                     | 0                    | 54, female                            | right, V2, V3                         | 5                                 | PTA, SCA          | MVD            | At a mean follow-up of 28   |
| IVIUIIIA EI UI. 1707                    | 0                    | 74, female                            | right, V2, V3                         | 9                                 | PTA, SCA, AICA    | MVD            | months (12-42 months), 8  |
|   |                      | 56, female                            | left, V2                              | 9                                 | PTA               | MVD            | patients remained painless  |
|   |                      | 69, female                            | left, V1, V2                          | 1                                 | PTA               | MVD            |   |
|   |                      | 67, female                            | left, V2, V3                          | б                                 | PTA               | MVD            |   |
| Tamura <i>et al</i> . 2003 <sup>3</sup> | 1                    | 51, female                            | left, V2                              | 10                                | PTA               | MVD            | Immediate painless after MVD  |
| Yamada <i>et al.</i> 2006 <sup>18</sup> | 1                    | 31, male                              | left, V2                              | 3                                 | SCA, AICA         | MVD            | Immediate painless after MVD  |
| Kawahara et al. 20118                   | 1                    | 86, female                            | right, V2                             | 10                                | PTA               | MVD            | Immediate painless after MVD  |
| Kato <i>et al</i> . 2011 <sup>19</sup>  | 1                    | 62, female                            | right, V1                             | ND                                | SCA               | MVD            | Remained painless after 14<br>months of follow-up   |
| Lee <i>et al</i> .2011 <sup>20</sup>    | 1                    | 61, female                            | right, V2, V3                         | 7                                 | PICA              | MVD            | Remained painless after 9   |
|   | -                    |                                       |                                       |                                   |                   | E              |   |
| Kono et al. 2013 <sup>21</sup>          | I                    | os, male                              | lett, ND                              | DN                                | PIA               | IE             | Kemained painless arter 12<br>months of follow-up   |
| 97 100 1 Q                              | -                    | · · · · · · · · · · · · · · · · · · · |                                       | 0                                 | 1 T.A.            |                | $\mathbf{D}$ = $\mathbf{D}$ |
| Park et al. 2014°                       | -                    | oo, male                              | lett, ND                              | ע                                 | PIA               | MVD            | Kemained painless after 12<br>months of follow-up   |
| Ladner et al. 2014 <sup>22</sup>        | 1                    | 66, female                            | left, V1, V2                          | ND                                | PTA               | IE             | Remained painless after 6   |
|   |                      |                                       |                                       |                                   |                   |                | dn-moning 10 sunnom   |
| Chen <i>et al.</i> 2015 <sup>23</sup>   | -                    | 69, female                            | left, V1, V2                          | ND                                | PTA               | IE             | Remained painless after 12<br>months of follow-up   |
| Fu <i>et al</i> . 2015 <sup>7</sup>     | 1                    | 63, female                            | left, V1, V2                          | 5                                 | PTA               | Gamma<br>knife | Remained painless after 6<br>months of follow-up  |
| Miki <i>et al.</i> 2019 <sup>9</sup>    | 1                    | 65, male                              | left, V2                              | 10                                | PTA, SCA          | MVD            | Remained painless after 4 months of follow-up   |
| Sano <i>et al.</i> 2020 <sup>10</sup>   | 1                    | 65, male                              | left, V2                              | 7                                 | PTA               | MVD            | Remained painless after 12<br>months of follow-up   |
| Sadashiva, 2021 <sup>24</sup>           | 1                    | 55, male                              | right, ND                             | 15                                | PTA, SCA          | MVD            | Remained painless after 6<br>months of follow-up  |

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PTA-associated TN in a few cases, while PBC has not been reported to be used previously in such cases, so the efficacy of PBC remains uncertain.

In our case, the patient experienced pain recurrence just two months after MVD. In MVD, the trigeminal nerve should be inspected from the brainstem to the MC to confirm the compressing vessels.13 We carefully inspected the cistern segment of the trigeminal nerve and successfully separated the nerve from the small artery originated from PTA, which relieved part of the compression and temporarily ameliorated the patient's symptoms. Nevertheless, the compression of the trigeminal nerve from PTA is not limited to the cisternal segment under certain conditions. Bondt et al. pointed out that in patients with PTA-related TN, the compression may lead to TN even if it is outside the cistern segment of the trigeminal nerve, such as when PTA is close to the medial part of the MC, especially a dilated PTA<sup>11</sup>, which is similar to the present case. As shown in Figure 1c, d, not only the small artery from PTA compressed the cistern segment of the trigeminal nerve, the dilated PTA trunk in the MC also oppressed the trigeminal nerve. On account of visualization limitation, the compression in the MC cannot be resolved surgically. As the vascular compression of the trigeminal nerve was not completely relieved, the effect of MVD surgery was only temporary.

For this patient, we chose PBC rather than MVD as the treatment for recurrence of TN. The purpose of MVD is to relieve the vascular compression of the cistern segment of the trigeminal nerve, but the compression due to the PTA trunk in the MC could not be resolved by MVD. Previous studies recommend PBC as a treatment option for failed MVD.<sup>14,15</sup> Therefore, we decided to treat the patient with PBC.

According to the literature review, there were no previous reports of PBC treatment in patients with PTA-related TN, so we had no previous reported experience as reference. We thus paid particular attention to two major considerations before performing PBC. First, the puncture depth of PBC should be shallower than usual. PTA occupied a part of the MC and severely squeezed its space (Figure 1c, d), requiring constant attention to the insertion depth during puncture. In order to prevent PTA frpm being damaged, the puncture was simulated by 3D Slicer before PBC, and the insertion depth was successfully estimated. Meanwhile, during the operation, the position of the puncture needle was controlled to just enter the foramen ovale through the

guidance of X-ray to avoid PTA injury, which was the key to the success of PBC. Second, the duration of balloon compression during PBC was given careful consideration. Mullan et al. found that there seems to be no difference in the efficiency between 1minute and 5-7minutes of the duration of balloon compression.<sup>16</sup> But for patients with recurrent TN or a long history of TN, a longer compression time is usually required to ensure the efficacy of PBC.<sup>17</sup> Our patient had recurrent TN with 8-year history of pain, so we applied a 5 minutes of balloon compression. The pain disappeared immediately after PBC. Based on the one year follow-up, the PBC achieved a satisfactory outcome, with only mild facial numbness and no recurrence of pain.

In conclusion, we report here the first case of PTA-associated TN treated by PBC. Although it was reported in the past that PTA-associated TN had achieved good results by MVD, our patient relapsed shortly after receiving MVD because of the compression to the trigeminal nerve in the MC from the PTA trunk. We conclude that for PTA-associated TN, if MVD is not successful, PBC can be considered as alternative treatment.

## DISCLOSURE

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