J Anesth (2010) 24:139–142
DOI 10.1007/s00540-009-0862-0

CLINICAL REPORT

A case of trigeminal neuralgia complicated by ipsilateral temporal arteritis

Yoko Kawaguchi · Masako Ebina · Tetsumi Sato · Yoh Ishiguro · Soroku Yagihashi · Kazuyoshi Hirota

Received: 6 August 2009 / Accepted: 22 October 2009 / Published online: 9 January 2010

Abstract Trigeminal neuralgia (TN) complicated with temporal arteritis (TA) is not a common disease, but it is a very important syndrome to consider for diagnosing facial pain in individuals older than 50 years. We therefore report on a rare case of TN with TA that occurred simultaneously on the same side with each symptom responding to specific treatment.

Keywords Trigeminal neuralgia · Temporal arteritis · Facial pain

Introduction

Trigeminal neuralgia (TN) is well defined by its characteristic symptoms and is one of the most common causes of facial pain in individuals older than 50 years. On the other hand, although temporal arteritis (TA) is an uncommon disease in Japan, it is likewise very important in the differential diagnosis for new unilateral facial pain in individuals older than 50 years. Both TN and TA need an early diagnosis and an appropriate therapy. We report a case of TN and TA that was simultaneously causing terrible pain in the same side of the patient's face.

Y. Kawaguchi (✉) · M. Ebina · T. Sato · K. Hirota
Department of Anesthesiology,
Hirosaki University Graduate School of Medicine,
5 Zaifu-cho, Hirosaki 036-8562, Japan
e-mail: my405maru@yahoo.co.jp

Y. Ishiguro
Department of Gastroenterology and Hematology,
Hirosaki University Graduate School of Medicine,
5 Zaifu-cho, Hirosaki 036-8562, Japan

S. Yagihashi
Department of Pathology and Molecular Medicine,
Hirosaki University Graduate School of Medicine,
5 Zaifu-cho, Hirosaki 036-8562, Japan

Case report

A 69-year-old man was referred to our department for evaluation of right unilateral facial pain. He had undergone an operation of right tympanoplasty for cholesteatoma otitis media a year previously. Six weeks before our initial physical examination, he experienced a paroxysmal attack of pain on the right side of his face that lasted several seconds when he was chewing. The paroxysm was provoked repetitively by chewing, talking, washing the face, and/or lightly touching the right side of his face. He consulted a dental clinic and started medication of carbamazepine 200 mg/day with a diagnosis of TN. Despite increasing carbamazepine to 400 mg/day, the pain gradually became more severe. During the 2 weeks before our evaluation, the paroxysm occurred spontaneously without any stimulation such as chewing or touching his face. He could not talk, chew, or sleep adequately when he visited our office.

The pain at first originated in the right temporal region. By the time the patient came to us, the area of pain had spread from the right forehead to the right posterior region of the neck, and the region near his right jaw joint was the most painful. Before investigating NRS or VAS, several paroxysmal attacks occurred during the medical interview, causing him to crouch and hold his head in his hands. Each attack continued for a half minute. There was no swelling, rubor, or exanthema in his face. There was no nuchal rigidity. We suspected TN caused by compression of V3. Exanthema was found on both forearms 10 days after

carbamazepine medication was started. At our initial examination, we thought carbamazepine had caused the exanthema and changed his treatment to clonazepam and naproxen.

Hematological values were as follows: white blood cell count 3920/μl, red blood cell count 507 × 10^3/μl, hemoglobin 15.5 g/dl, and platelets 12.9 × 10^4/μl. White blood cell count and platelets were slightly decreased. C-reactive protein (CRP) was positive at 1.2 mg/dl.

The day after our first examination, he came to our office again because the paroxysmal pain had become more severe and longer. Moreover, a pulsating pain had continued in the intervals between paroxysmal attacks. An otolaryngologist examined the patient and found evidence of chronic sinusitis but no other abnormal findings. Computed tomography showed no abnormalities. The strongest point of pain was in front of the right ear, and he had a slightly elevated temperature at 37.0°C; therefore, we suspected TA and started prednisolone 45 mg/day.

He was admitted to our hospital 2 days after the second examination because the pain had become more severe and prolonged, especially at night, and he could not sleep, and neither open his mouth nor eat. We consulted a physician who was a specialist in collagen diseases. His examination identified a pulsating induration and tenderness of the right superficial temporal artery. The white blood cell count, platelets, and CRP had improved; however, TA was strongly suspected, and prednisolone medication was continued at 20 mg/day. Simultaneously, baclofen 15 mg/day was started for TN.

The frequency and strength of the paroxysmal attacks became less, and the pain disappeared 6 days after the medication was started.

Magnetic resonance angiography (MRA) (Fig. 1) showed that the right superior cerebellar artery compressed the upper medial border of the right proximal trigeminal nerve and a petrosal vein was in contact with the lateral part of the right trigeminal nerve root. These findings supported the diagnosis of TN. Biopsy of the right superficial temporal artery proved TA based on the presence of active acute angitis (Fig. 2) and inflammatory angitis (Fig. 3).

In spite of being treated with baclofen and prednisolone, a month later, an electric shock-like paroxysm recurred when he crunched a cherry seed. Because paroxysms were provoked by chewing, grimacing, and washing his face, he had a surgical microvascular decompression performed for TN. After the operation, he was completely free from pain.

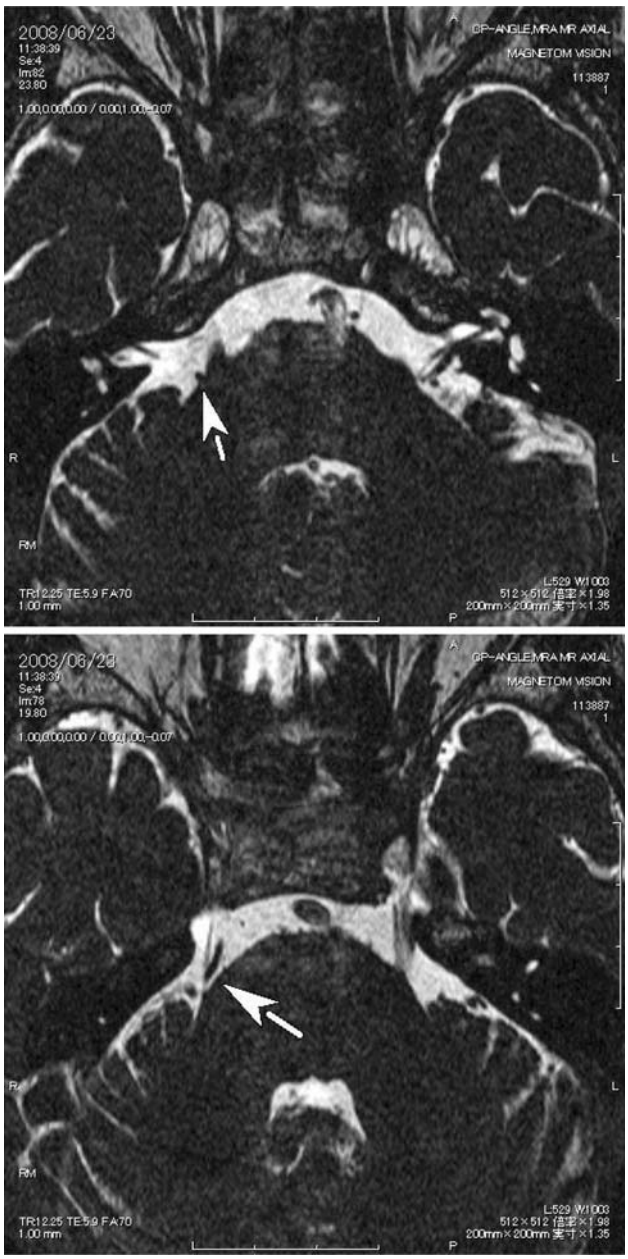

Fig. 1 Magnetic resonance angiography (MRA) shows that the right superior cerebellar artery (*arrow*) compresses the upper medial border of the right proximal trigeminal nerve (*upper panel*); a petrosal vein (*arrow*) is in contact with the lateral part of the right trigeminal nerve root (*lower panel*)

Discussion

The annual incidence of TN is 4–13 per 100,000 people [1, 2]. TN is one of the most frequently seen neuralgias in the elderly, and consultations in the pain clinic are common. Most idiopathic cases occur over age 50. The incidence increases gradually with age [3]. Most cases of TN are caused by compression of the trigeminal nerve root, usually within a few millimeters of entry into the pons (the root entry zone) [4]. Compression by an aberrant loop of artery or vein is thought to account for 80–90% of cases. The pain usually lasts from one to several seconds. It has been

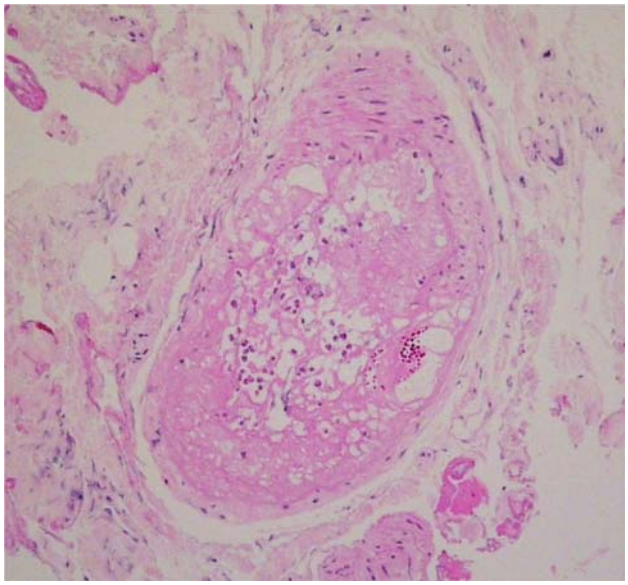

Fig. 2 Pathological sample of the right superficial temporal artery shows invasion of neutrophils into the inner and medial membranes and fibrinoid necrosis with partial vacuolation. Hematoxylin and eosin stain

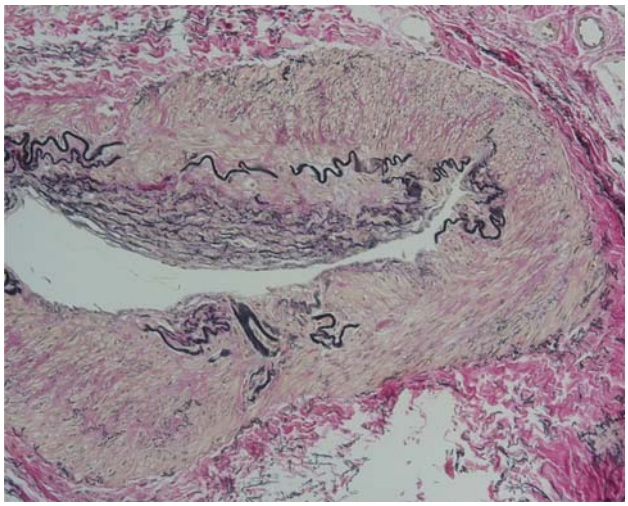

Fig. 3 Pathological sample of the right superficial temporal artery shows the thickness of the inner and medial membranes, a narrowed lumen, and split inner elastic lamella. Elastica van Gieson stain

described as electric, shocklike, or stabbing, and occurs repetitively within an interval of a few minutes. The pain is provoked by chewing, talking, brushing the teeth, washing the face, grimacing, and/or lightly touching the trigger point of the affected nerve. In contrast to some other facial pain syndromes, TN typically does not awaken patients at night. The symptoms in this case were only a few seconds of paroxysm provoked by chewing, which was consistent with TN.

However, it was difficult to conclude it was only typical TN at our first examination, because the area of pain was too wide, the period of paroxysm was too long, and there were sudden attacks at night and when resting. In medication therapy, a number of randomized, controlled trials have established the effectiveness of carbamazepine for TN, with a response rate of more than 70% [5].

On the other hand, the annual incidence of TA is about 690 cases, and 1.45 per 100,000 people in Japan [6]. It is very uncommon compared with the incidence in Europe and America [7]. In addition to geography and race, age is a risk factor for TA; TA almost never occurs before the age of 50. The cause of TA is unknown, but some reports suggest a viral infection as a previous symptom. At least 70% of patients with TA manifest a new type of headache, and the level and type of pain can vary. The classic TA headache is located in the temporal regions, and it may also occur in frontal or occipital areas or be generalized. The headaches may become progressively worse, especially at night, but can also wax and wane, sometimes subsiding temporarily before treatment is started. Tenderness or thickening of the temporal or other cranial arteries can also occur [8].

In this case, the spread of the area of pain and the onset of headaches at night and during rest suggested that TA occurred independently during the treatment of TN. It made the symptoms more complicated. Characteristic laboratory abnormalities seen in most patients with TA include a high erythrocyte sedimentation rate (ESR), elevated CRP levels, and anemia. Autoantibody is usually negative. In this case, the white blood cell count and platelets were slightly decreased, CRP was positive at 1.2 mg/dl, autoantibodies were negative, and other laboratory findings were in the normal range. The white blood cell count, platelets, and CRP returned to normal immediately after discontinuation of carbamazepine and administration of prednisolone. After that, the red blood cell count decreased and recovered. This finding suggests that carbamazepine contributed to bone marrow suppression, and the time lag between recovery of the various blood cells was caused by differences in their respective lifetimes.

We thought the symptoms of this case must have more than one cause. TN was strongly suspected at the first examination, but carbamazepine was not effective and the pain was not typical for TN. Additionally, consideration was complicated by the presence of chronic sinusitis and a history of ear surgery on the same side for cholesteatoma otitis media. While continuing to treat TN, we quickly contacted an otolaryngologist and a specialist in collagen diseases. TA was promptly diagnosed, and it was treated with systemic glucocorticoids to avoid visual loss. Fortunately, in this case, visual symptoms never occurred. The

pain from TA disappeared after the beginning of prednisolone medication and did not recur after decreasing prednisolone. However, the symptoms of TN were sustained. The residual pain resulted from typical TN because the patient had no symptoms at night or in rest. The pain was limited to areas supplied by the trigeminal nerve (V3), and the paroxysm was not prolonged. In addition to clinical symptoms, MRA and histological findings demonstrated that TN and TA occurred concomitantly in this patient.

Conclusion

Trigeminal neuralgia and TA are not common diseases but should be considered in cases of facial pain in individuals older than 50 years. We therefore reported on a rare case of TN and TA that occurred simultaneously on the same side with each syndrome responding to specific treatment.

References

1. Katusic S, Williams DB, Beard CM, Bergstralh EJ, Kurland LT. Epidemiology and clinical features of idiopathic trigeminal neuralgia and glossopharyngeal neuralgia: similarities and differences, Rochester, Minnesota, 1945–1984. Neuroepidemiology. 1991; 10:276.
2. MacDonald BK, Cockerell OC, Sander JW, Shorvon SD. The incidence and lifetime prevalence of neurological disorders in a prospective community-based study in the UK. Brain. 2000; 123:665–76.
3. Childs AM, Meaney JF, Ferrie CD, Holland PC. Neurovascular compression of the trigeminal, glossopharyngeal nerve: three case reports. Arch Dis Child. 2000;82:311.
4. Love S, Coakham HB. Trigeminal neuralgia: pathology and pathogenesis. Brain. 2001;124:2347–60.
5. Wiffen P, Collins S, McQuay H, Carroll D, Jadad A, Moore A (2005) Anticonvulsant drugs for acute and chronic pain. Cochrane Database Syst Rev CD001133.
6. Kobayashi S, Yano T, Matsumoto Y, Numano F, Nakajima N, Yasuda K, et al. Clinical and epidemiologic analysis of giant cell (temporal) arteritis from a nationwide survey in 1998 in Japan: the first government-supported nationwide survey. Arthritis Rheum. 2003;49:594–8.
7. Lawrence RC, Helmick CG, Arnett FC, Deyo RA, Felson DT, Giannini EH, et al. Estimates of the prevalence of arthritis and selected musculoskeletal disorders in the United States. Arthritis Rheum. 1998;41:778–99.
8. Hunder GG, Bloch DA, Michel BA, Stevens MB, Arend WP, Calabrese LH, et al. The American College of Rheumatology 1990 criteria for the classification of giant cell arteritis. Arthritis Rheum. 1990;33:1122–8.