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Case Report

Development of trismus in a case of temporal arteritis

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ABSTRACT

Giant cell arteritis, which most frequently occurs in individuals over 50 years of age, is a vasculitis of unknown etiology characterized by inflammation of the carotid arteries and their branches, particularly the temporal arteries. An 85-year-old woman visited our department with chief complaints of pain in the right temporomandibular joint and trismus. She had a history of gastric ulcer, but no notable family history. The left mandibular tooth #12 had been extracted at a local dental office. Subsequently, the patient developed pain in the area extending from the right temporomandibular joint to the occipital region and trismus. As these symptoms did not improve, she was referred to our department. Also, the causative tooth for oral pain was unknown, and so analgesic drugs were prescribed under a provisional diagnosis of suspected temporomandibular disorder. Two days later, the patient returned to us because of fever and worsening malaise. Marked elevation of white blood cell count (12,800/μL) and C-reactive protein level (33 mg/dL) led to emergency hospitalization. The cause of fever was unknown and the patient was therefore systematically examined in the internal medicine department, and giant cell arteritis was suspected. Based on biopsy results, giant cell arteritis was definitively diagnosed. After admission, administration of oral prednisolone was started, and symptoms resolved by the following day. The steroid was then gradually tapered, and no complications occurred. Once giant cell arteritis onset has occurred, some patients will visit the hospital with initial symptoms of trismus and masticatory muscle pain, and so these patients are sometimes first examined at the department of maxillofacial surgery. Despite the low incidence of this condition, severe complications, such as blindness or a dissecting arterial aneurysm, can occur if treatment is delayed, so this condition should always be borne in mind, and early diagnosis and treatment must be ensured.

1. Introduction

Giant cell arteritis is a disease of unknown etiology that occurs mainly in late-middle-aged and elderly individuals ≥50 years old and is primarily characterized by inflammation of the carotid arteries and their branches, particularly the temporal artery [1]. The main presenting symptoms are fever, fatigue, headache, loss of appetite, impaired visual acuity, and temporomandibular joint (TMJ) pain. Some reports have documented difficulty in differentiating this condition from TMJ disorders [2]. Here, we report a case of giant cell arteritis that we encountered that presented with trismus.

2. Case report

The patient was an 85-year-old woman who first presented to our center in December 2014 with chief complaints of right TMJ pain and trismus. Past medical history and family history were unremarkable. The patient had undergone extraction of tooth no. 12 from the left mandible 5 days before initial presentation. She subsequently developed pain extending from the right TMJ region to the occipital region and trismus. The symptoms did not improve, and she was then referred to our department for examination.

On general examination, her body temperature was 37.2 °C, blood pressure was 110/66 mmHg, and pulse rate was 89 beats per min. Two days later, her temperature rose to 38.2 °C, and she had poor appetite and generalized fatigue. Extraoral examination revealed that the pain was focused around the right temporal region, with spontaneous pain in the right TMJ region and trismus (maximum unassisted mouth opening: 13 mm). On intraoral examination, there were no signs of infection in the area around the tooth extraction site, and no other sources of dental infection or neoplastic lesions were observed (Fig. 1).

Blood tests revealed a total leucocyte count of 12,800/μL, C-reactive protein (CRP) level of 33.0 mg/dL, and erythrocyte sedimentation rate (ESR) of 111 mm (60 min), all indicative of severe inflammatory

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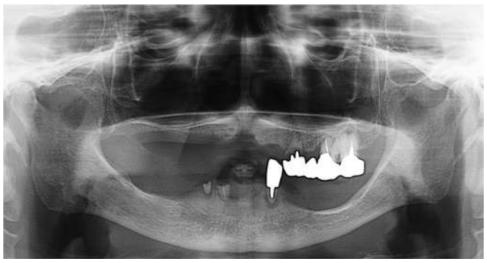


Fig. 1. Orthopantomography. No signs of intraoral infection are observed.

Table 1
Blood test findings during initial examination. Leucocyte count 12,800/μL, C-reactive protein (CRP) levels 33.0 mg/dL, and erythrocyte sedimentation rate (ESR) 111 mm (60 min), which all indicate severe inflammatory response.

[Full blood count]	[Biochemistry]	
WBC 12,800 /μl	TP 6.8 g/dl	AMY 53 U/l
RBC 400 × 104 /μl	ALB 2.8 g/dl	BUN 42.2 mg/dl
Hb 12.4 g/dl	TB1.0 mg/dl	Cre 1.38 mg/dl
Plt 23.4 × 104 /μl	AST73 U/l	UA 8.4 mg/dl
Neu 85.3 %	ALT40 U/l	TCH 169 mg/dl
PT activity 61.4%	LDH214 U/l	GLU 136 mg/dl
PT-INR 1.26	Na140 mmol/l	CRP 33.0 mg/dl
APTT 44.8seconds	K4.5 mmol/l	
FDP 12.2 μg/ml	Cl101mmol/l	
ESR (30 min) 50mm	Ca8.4 mg/dl	
ESR (60 min) 111mm		

response (Table 1). Based on these findings, we made an empirical clinical diagnosis of generalized inflammatory response of unknown cause.

Subsequently, we arranged for a review in the internal medicine department and obtained computed tomography (CT) scans of the neck, thorax, and abdomen, but found no lesions that could be an infective focus. Echocardiography revealed no findings suggestive of infective endocarditis. We also performed numerous serology tests for viral infection and these tests were all negative. The patient was considered to have an infection of unknown origin and was admitted for detailed investigation. After admission, the patient was started on amoxicillin at a dose of 250 mg/day, and ceftazidime at a dose of 2 g/day. Blood test findings on the fifth day of antibiotic administration showed no improvement of the inflammatory response, with persistence of severe headache. Subsequent palpation of the temporal region revealed funicular subcutaneous nodules along the tortuous course of the temporal artery. On suspicion of giant cell arteritis, we performed histopathological tests and started the patient on oral prednisolone (Predonine®, PSL) at a dose of 60 mg/day. The patient showed a favorable response to steroid therapy and the headache improved on the second day of PSL administration. By day 15 of hospitalization, leucocyte count had dropped to 9000/μL, CRP level was 0.3 mg/dL, and ESR was 27 mm (60 min), which all indicated marked improvement; trismus also disappeared (Table 2). The PSL dose was subsequently tapered, no recurrence was observed, and the patient was discharged.

Histopathology revealed vascular luminal stenosis associated with inflammatory hypertrophy of all layers of the vessel wall under low magnification. A histiocytic and neutrophilic cellular infiltrate was observed, as well as a granuloma comprising giant cells extending from the media to the adventitia (Fig. 2A, B). Based on these findings, we made a definitive diagnosis of giant cell arteritis.

3. Discussion

The concept of giant cell arteritis as a disease was first established by Horton et al. [3] in 1932. Giant cell arteritis is characteristically a granulating vasculitis that occurs in late-middle-aged and elderly patients and affects the areas supplied by the internal and external carotid arteries, but particularly the temporal artery. The cause remains unknown, but an abnormal cell-mediated inflammatory response has been postulated. The disorder is extremely rare in Japan with an onset frequency of approximately 0.65 individuals per 100,000 population. Onset most commonly occurs at a mean age of 71.5 ± 10.8, with a male to female ratio of 1:1.7; thus, it is most frequently observed in elderly female patients.

The main clinical symptoms are headache, fever, loss of appetite, impaired visual acuity, with redness, swelling, and impaired mandibular movement in the area supplied by the temporal artery. Symptoms caused by vasculitis are headache, as well as redness and induration in the area supplied by the temporal artery, while the symptoms mediated by inflammatory cytokines are fever and generalized fatigue. Symptoms due to ischemia of areas supplied by the injured vessels are impaired visual acuity, jaw claudication, and skin ulcers.

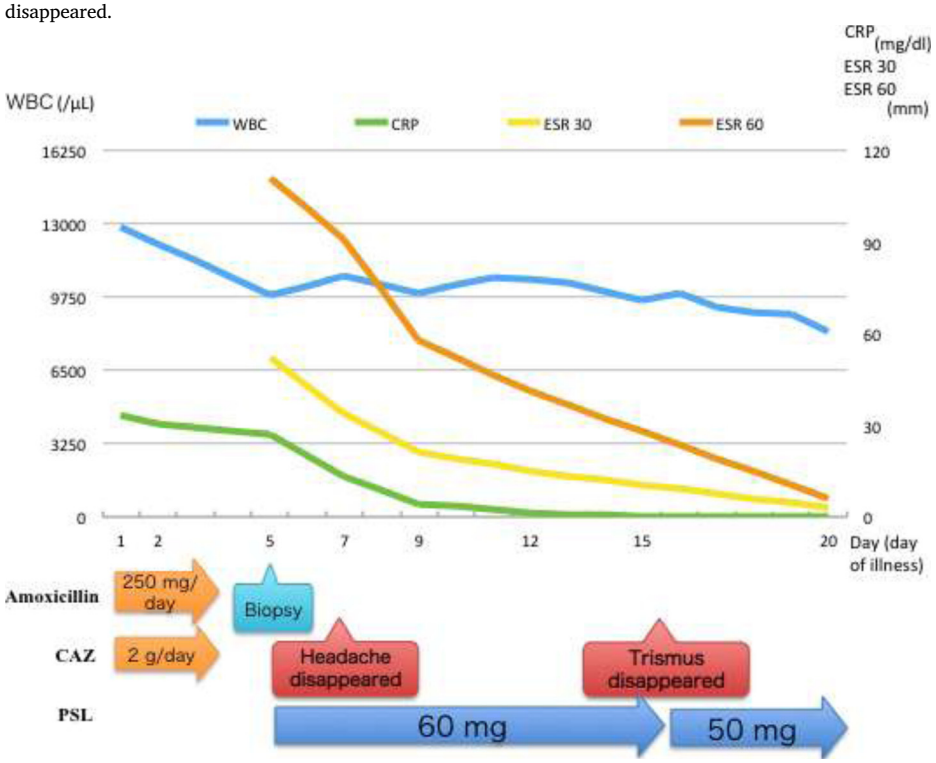
The trismus observed in our patient is a characteristic symptom of giant cell arteritis resulting from ischemia of the muscles of mastication due to the vasculitis, leading to myalgia and dyskinesia. This finding is known as jaw claudication, and strongly resembles a TMJ disorder, so careful differentiation is required. In this case, we had the fortuitous opportunity to perform the initial examination in our department.

To our knowledge, there have been 17 cases of giant cell arteritis [1,5,6]. that presented with trismus between 1990 and 2013 in Japan, and of those, 2 cases were examined at the department of dentistry and maxillofacial surgery [1,7]. The frequency of jaw claudication among cases of giant cell arteritis is reported to be between 8% and 50% in Europe and the US compared with 15% in Japan[8]. According to Kanno et al. [1], despite the differences in frequency due to genetic background, the lower incidence in Japanese reports could be attributed to the fact that it is simply overlooked because jaw claudication is not recognized as a symptom.

In addition, if the disorder progresses, patients could develop ischemia of the retina or optic nerve due to vascular lesions affecting the ophthalmic and posterior ciliary arteries, which in turn could cause decreased visual acuity and diplopia. This may be associated with the risk of complete blindness if the disease further progresses. Recovery is difficult once ocular symptoms have developed, so early diagnosis and treatment are preferable. We consulted an ophthalmologist about our patient as soon as we suspected giant cell arteritis and confirmed that there were no abnormalities. However, reports indicate that impaired visual acuity is observed in approximately half of all patients presenting with trismus in Europe and the US [9] as in the present case. The mean morbidity period from the time of giant cell arteritis onset until confirmed diagnosis is 8 weeks, although reports indicate that some cases take up to 6 months before the correct diagnosis is made [1]. We administered steroid treatment to our patient 7 days after she noticed headache, so she suffered no complications and could be discharged, although we believe that there is a risk of blindness if treatment is delayed.

The diagnostic criteria for giant cell arteritis are based on the American College of Rheumatology criteria (1990) [10] (Table 3). Our patient met all five of the diagnostic criteria. Some patients do not present with the classical histological profile and a temporal artery biopsy may be considered. This is because the histological profile differs based on biopsy site and timing, disease stage, and effects of treatment. The presence of inflammatory cell infiltrates does not necessarily lead to the identification of giant cells, and lesions may sometimes be non-continuous, so the best practice is to harvest a portion of the artery that is ≥2.5 cm long and investigate numerous sections [11]. Recently, ultrasound, magnetic resonance imaging (MRI), CT, and positron emission tomography scans have all be used to assess giant cell arteritis and recent studies have helped to clarify their utility. vascular imaging is increasingly being used in the diagnostic algorithm for giant cell arteritis. Results from recent vascular ultrasound and high-resolution cranial MRI studies have led some groups to suggest foregoing temporal

Table 2
Clinical course after admission. The patient showed a favorable response to steroids; headache improved on the second day of PSL administration. By day 15 of hospitalization, leucocyte count was 9000/μL, CRP level was 0.3 mg/dL, and ESR was 27 mm (60 min) which all indicated marked improvement. Her trismus also disappeared.



artery biopsy (TAB) in selected patients [12].

Steroid therapy is an effective treatment modality. Patients are started on PSL doses of 40–60 mg/day, which are then gradually reduced by 5 mg/week. Normally, when the dose drops below 15 mg/day, it is reduced by 2.5 mg/month based on the symptoms and test values, then a maintenance dose of 5 mg/month is administered for a period of 6–12 months.

We started our patient on 60 mg of oral PSL, and there was remarkable improvement of headache and generalized fatigue 2 days later. After 10 days of oral administration, on day 15 of hospitalization, leucocyte count was 9000/μL, CRP level was 0.3 mg/dL, and ESR was 27 mm (60 min), all indicative of marked improvement, and trismus disappeared. There were no subsequent complications, PSL dose was tapered, and the patient was discharged.

PSL therapy is the standard of care for giant cell arteritis, although methotrexate is used in some cases and anti-IL-6 therapy is now approved for the treatment of this condition [13]. Sammel et al. reported their current use of tocilizumab (TCZ) for patients who cannot tolerate long-term corticosteroids or those in whom corticosteroid therapy with methotrexate and/or leflunomide-based treatment failed. They also state that it offers an effective treatment option beyond corticosteroids. Furthermore, underpinning these developments have been steady

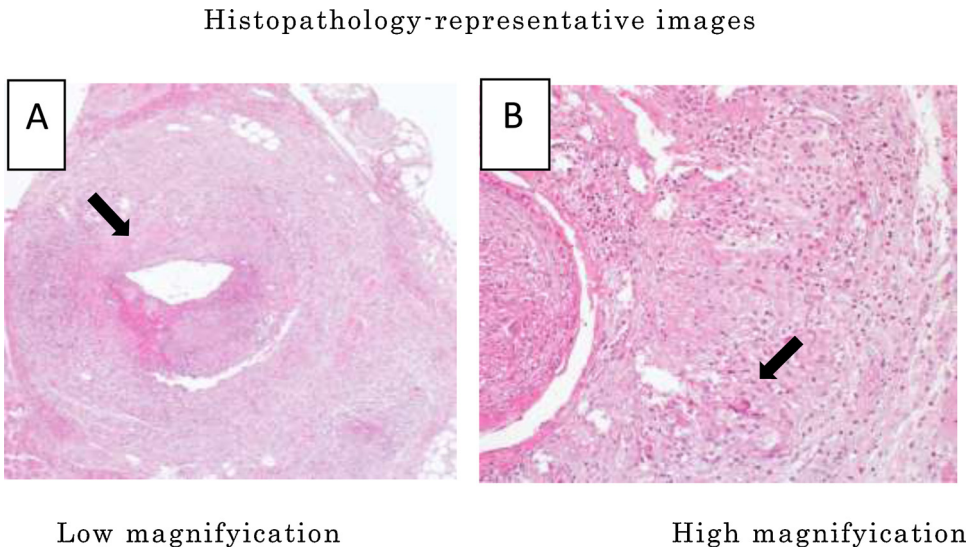


Fig. 2. Histopathology image. A Vascular luminal stenosis associated with inflammatory hypertrophy of all layers of the vessel wall is observed under low magnification. B Histiocytic and neutrophilic cellular infiltrate is observed, as well as a granuloma containing giant cells extending from the media to the adventitia.

Table 3
The American College of Rheumatology diagnostic criteria for giant cell arteritis. Our patient met all five of the diagnostic criteria.

Criterion	Definition
1. Age at disease onset ≥ 50years	Development of symptoms or findings beginning at age 50 or older
2. New headache	New onset of new type of localized pain in the head
3. Claudication of jaw, tongue, or on deglutition	Development of worsening of fatigue discomfort in muscles of mastication, tongue, or swallowing muscles while eating
4. Temporal artery abnormality	Temporal artery tenderness to palpation or decreased pulsation, unrelated to artery or other cranial arteries
5. Scalp tenderness or nodules	Development of tender areas of or nodules over the scalp, away from the temporal artery or other cranial arteries
6. Abnormal artery biopsy	Biopsy specimen with artery showing vasculitis characterized by a predominance of mononuclear cell infiltration or granulomatous inflammation, usually with multinucleated giant cells

improvements in our understanding of the immunopathology of this vision-threatening disease with the potential to develop better biomarkers and therapeutics in the years to come [12]. In 2017, TCZ became the first medication to be granted US Food and Drug Administration (FDA) approval specifically for the treatment for giant cell arteritis.

Once giant cell arteritis onset has occurred, some patients will visit the hospital with initial symptoms of trismus and masticatory muscle pain, and these patients are often first examined at the department of maxillofacial surgery. Despite the low incidence of this condition, severe complications, such as blindness or a dissecting arterial aneurysm, may occur if treatment is delayed, so this condition should always be borne in mind, and early diagnosis and treatment must be ensured.

Disclosures

There were no conflicts of interest related to this publication.

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