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Case report

Jaw claudication is the only clinical predictor of giant-cell arteritis

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ABSTRACT

Giant-cell arteritis (GCA), commonly known as temporal arteritis, is a chronic granulomatous vasculitis that affects predominantly the extracranial branches of the carotid artery. Although an algorithm for diagnosing GCA that includes both biopsy and imaging examinations has been recently proposed, it harbors the possibility of false negatives. Hence, jaw claudication caused by ischemia of the masticatory muscles is one of the important clinical predictors for implementation of temporal artery biopsy (TAB). We describe a case of GCA in which jaw claudication was the only clinical predictor for implementation of TAB. A 78-year-old man was referred to our department with facial pain associated with mastication. He had been admitted 3 weeks previously to another department to investigate an unidentified fever. A blood test revealed an elevated C-reactive protein level and a high erythrocyte sedimentation rate (70 mm/h). Although ultrasonography and computed tomography angiography of the temporal region showed no findings specific for GCA, based on our examinations we determined that his facial pain arose from jaw claudication rather than temporomandibular disorder. Histopathological examination by temporal artery biopsy showed intima thickening with disruption of elastic lamina and inflammatory cell infiltration, and we consequently diagnosed GCA. Clinical symptoms immediately resolved after prescription of prednisolone 40 mg/day for 2 days. In this case of GCA the imaging examinations of the superficial temporal artery were false-negative, and jaw claudication was the most important predictor of GCA.

1. Introduction

Giant cell arteritis (GCA), also commonly known as temporal arteritis, is a systemic granulomatous vasculitis involving medium and large arteries, most predominantly the extracranial branches. While temporal artery biopsy (TAB) has long been considered the gold-standard investigation for the diagnosis of GCA [1–13], it is an invasive procedure that may carry a risk of facial nerve injury [2,14,15]. Although several imaging techniques as alternative diagnostic strategies have recently become available to investigate patients with suspected GCA [16–21], reduced sensitivity and specificity may hamper their effectiveness. Laboratory markers (e.g. CRP levels, ESR) and clinical features (e.g. abrupt-onset headache, temporal cutaneous hyperalgesia, jaw claudication, diplopia) are also useful as clinical predictors for the implementation of TAB [1–13]. Among these clinical predictors, jaw claudication is especially important as it can be discovered by a dentist [22].

We describe a case of GCA in which the imaging examinations of the superficial temporal artery were false-negative, whereby jaw claudication was the only predictor for implementation of Table

Abbreviations: GCA, giant cell arteritis; TAB, temporal artery biopsy; CT, computed tomography; CRP, C-reactive protein; ESR, erythrocyte sedimentation rate; TMD, temporomandibular disorders; STA, superficial temporal artery; FDG PET, fluorodeoxyglucose positron emission tomography; DC/TMD, Diagnostic Criteria for TMD; VAS, visual analog scale; ACR, American College of Rheumatology.

☆ AsianAOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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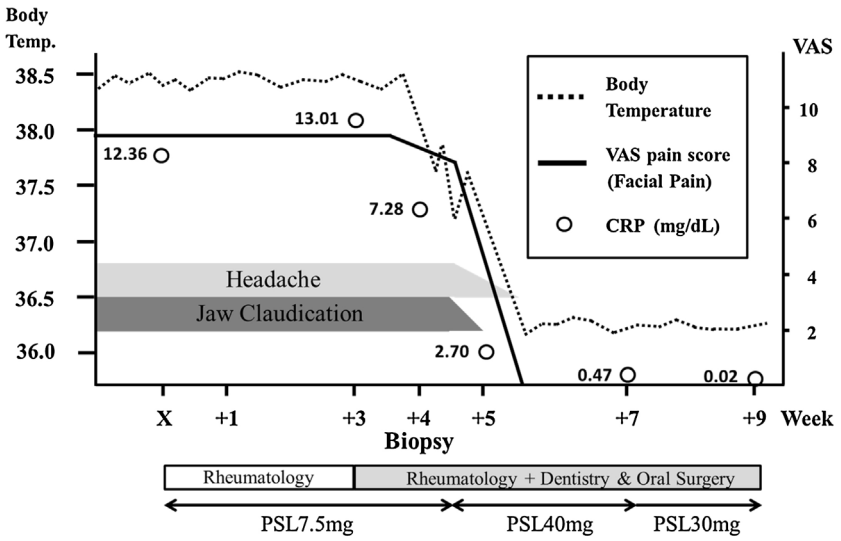

Fig. 1. X denotes the date of hospitalization. Temporal artery biopsy was performed 1 month after the first visit to our department (X+4). All of the clinical symptoms improved immediately after administration of prednisolone.

Table 1
Hematological findings on admission to our hospital. Bold* indicates abnormal levels.

Item	Data	Item	Data
WBC (X10^3 ul)	**11.8***	CRP (mg/dL)	**12.36***
RBC (X10^6 ul)	4.76	RF (IU/ml)	**91***
HGB (g/dl)	15.0	MMP-3 (ng/ml)	**179.8***
HCT (%)	46.8	Anti-CCP Ab (IU/ml)	<0.6
MCV (fl)	98	Anti-DNA Ab (IU/ml)	3
MCH (pg)	31.5	PRP	Negative
MCHC (g/dl)	32.1	HBsAg	Negative
PLT (X10^3 ul)	316	HBsAb	Negative
ESR (mm/hour)	**89***	HBCAb	Negative
CK (U/L)	<20	HIV	Negative

2. Case report

2.1. History before temporal artery biopsy

A 78-year-old man who complained of an unidentified fever, headache, and bilateral facial pain associated with mastication over the previous several months had been admitted to Keio University Hospital 3 weeks previously (Fig. 1). He was referred by the Department of Rheumatology to our department (Dentistry & Oral Surgery) for further examination of the bilateral facial pain. The patient's medical history was angina pectoris and prostatomegaly, and family history comprised adult Still's disease in his daughter. The fever often had spiked around 38.5 °C. On admission to the hospital a blood test had revealed an increased CRP level and a high ESR (70 mm/h) (Table 1). Rheumatoid arthritis had been excluded on the basis of hematological and physical findings. Although a whole-body survey with ^{18}F FDG PET (Fig. 2) and gallium scintigraphy (Fig. 3) to identify the source of the fever had found only prostatitis, his fever persisted after appropriate treatment for this condition. Adult Still's disease had also been suspected because of his family history, but clinical diagnostic examinations for this disorder proved negative. In addition, ultrasonography (Fig. 4A) and CT angiography (Fig. 4B, C) were performed to differentiate the GCA, but demonstrated no particular findings in the superficial temporal artery (STA). Because the characteristics of facial pain (described below) strongly indicated the existence of jaw claudication, TAB was performed 1 month after hospitalization (Fig. 1).

2.2. Characteristics of pain

Although the patient was unaware that the onset of headache and facial pain was associated with mastication, both symptoms were spreading from the bilateral temporal area to the bilateral buccal area. The patient described the pain as "pulsating". On a VAS scale from 0 to 10, pain intensity was graded 8–9. The pain began after initiation of mastication and continued for approximately 10 min before weakening. Any attempt to masticate, open the mouth, or grind the teeth aggravated the pain. The pain was not aggravated by physical activity, and no autonomic signs were apparent. The severe pain led to inability to masticate, and resulted in a body weight loss of 8 kg within 1 month.

2.3. Clinical findings on extra-oral and intra-oral examinations

Our clinical examinations for his facial pain associated with mastication revealed no unusual findings on intra-oral investigation. Although a dental panoramic radiograph revealed radiolucency at the apex of the first molar (Fig. 5), the teeth exhibited no percussion pain and mobility. The examinations to differentiate temporomandibular disorders (TMD) were based on the Diagnostic Criteria for TMD (DC/TMD) [23]. Unassisted opening without pain was 28 mm, even though the mandibular opening pattern was straight. Maximum unassisted opening and maximum assisted opening were limited to 35 mm and accompanied by pain at the bilateral temporal and masseter areas. Although the bilateral temporomandibular joints had tenderness on palpation at the lateral poles and periphery, the tenderness was a non-familiar pain. Meanwhile, the bilateral temporal muscle and masseter muscle were extremely sensitive to palpation, even though cutaneous haphalgesia in bilateral temporal area was unclear. Tenderness of these masticatory muscles was familiar to the patient's facial pain associated with mastication. All points on the masseter and temporalis that according to the DC/TMD should be palpated by the examiner had severe tenderness. We were unable to confirm the specific location of the source of pain on the masseter and temporalis. Although the patient sensitive to palpation, No funicular welling or pulsation of the STA was found on palpation of the bilateral temporalis muscle. The patient didn't have any optical symptom including blurred vision and diplopia.

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