

# A Rare Case of Bilateral Temporal Arteritis in a 40-year-old Patient

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## Abstract

**Introduction:** Temporal arteritis (TA) is a chronic inflammatory vasculitis involving medium and large arteries, which mostly occurs after the age of 50 and involves one side of the body.

**Case Presentation:** We present a 40-year-old male patient with simultaneous bilateral temporal arteritis along with a headache, mild fever, palpation of temporal arteries and anemia who responded to 1 mg/kg oral daily prednisone for three months. This patient was referred to the Pars Hospital in Tehran, Iran on March 2016.

**Conclusions:** Temporal arteritis should be kept in mind as an important curable differential diagnosis, not only in ages above 50, but also in lower ages and should be evaluated in both sides.

**Keywords:** Bilateral Temporal Arteritis, Giant Cell Arteritis, Young Patient

## 1. Introduction

Temporal arteritis (TA), also known as giant cell arteritis (GCA), is a chronic granulomatous inflammatory disease and the most common systemic necrotizing vasculitis that involves large and medium arteries (1). It typically occurs in people over the age of 50 (2) and its prevalence increases with age, however, its prevalence varies amongst different nations and is reported to be more rare in Asians than any other nation (3, 4).

It has several differential diagnoses due to the wide range of signs and symptoms. The most common symptoms include temporal artery tenderness in 66% of patients and a new onset of severe headaches in one-third of patients. It may also be complicated with serious symptoms such as, cardiovascular events, cerebrovascular ischemia and permanent or transient visual loss (1, 5). Therefore, high clinical suspicion is required for on-time diagnosis and treatment of patients to prevent severe complications, such as visual loss (2, 6).

Five criterias have been identified and diagnosed, based on three of the following five, 93.5% has sensitivity and 91.2% has specificity: age over 50 years, newly onset localized headache, tenderness or decreased pulse of the temporal artery, elevated erythrocyte sedimentation rate (ESR)  $\geq$  50 mm / hour and necrotizing arteritis in the

biopsy specimen of the artery. As it usually involves one side of the body, very few cases of bilateral temporal arteries have been reported (7), and few cases have been reported at an age earlier than 50 (8). Here, we described a rare case of bilateral temporal arteritis, presented earlier than 50 years of age.

## 2. Case Presentation

A 40-year-old man was referred to the neurology clinic of Pars hospital (private hospital in Tehran and not a referral hospital), affiliated to the Iran University of medical sciences, Tehran, Iran, in March 2016. This patient had a three-week history of double-sided temporal headaches that was resistant to usual treatment. On admission, he had a mild fever (38.2°C) with no specific medical condition or positive past medical history. He had normal vital signs and a BMI of 23 kg/m<sup>2</sup>. The temporal arteries of both sides were inflated and palpable. Funduscopic examination was normal and no visual problem was detected. There were no jaw or tongue claudication. Erythrocyte sedimentation rate (ESR) and C - reactive protein (CRP) levels were elevated (Table 1).

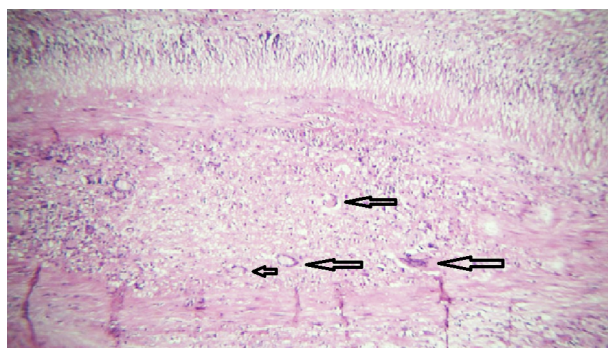
Urine analysis, rheumatoid factors, circulating immune complexes, antinuclear antibodies and the chest X-ray were normal. The brain computed tomography (CT)

**Table 1.** Clinical Laboratory Findings

The Serum Test	Patients Value	Unit	Measurement Device
White blood cells	14500	(cells/mL)	Sysmex® XN-Series
Red blood cells	4.34	( x 106 /mL)	Sysmex® XN-Series
Hemoglobin	12.4	(g/dL)	Sysmex® XN-Series
Hematocrit	37.8	(%)	Sysmex® XN-Series
Platelets	418000	mm <sup>3</sup>	Sysmex® XN-Series
<b>Differential white blood cell</b>			
Neutrophil	85	(%)	Sysmex KX-21N
Lymphocytes	10	(%)	Sysmex KX-21N
Monocyte	3	(%)	Sysmex KX-21N
Eosinophil	1	(%)	Sysmex KX-21N
Basophils	1	(%)	Sysmex KX-21N
CRP	142.8	(mg/L)	AQT90 FLEX analyzer
E.S.R. 1 hrs.	94	(mm/hrs)	Westergren-based ESR 'analyzer' H02-A5
ANCA	0.13	(U/ML)	ELISA
Complement component C3	147	(mg/dL)	AU2700/5400 Beckman Coulter Analyzers
Complement component C4	22	(mg/dL)	AU2700/5400 Beckman Coulter Analyzers

Abbreviations: CRP, C-reactive protein; ESR, estimated sedimentation rate; ANCA, Anti-neutrophil cytoplasmic antibody.

scan, magnetic resonance imaging (MRI) and trans cranial color sonography all showed normal results. In the biopsy of the temporal artery in two blocks, the specimen consisted of  $4.3 \times 0.4$  cm artery, which showed an arterial wall with nodular intimal thickening, accompanied by thrombosis. Using the Hematoxylin and Eosin (E&O) staining, granulomatous inflammation was observed in the media consisting of macrophages, multinucleated giant cells, lymphocytes and eosinophils (Figure 1).



**Figure 1.** 40X, Hematoxylin and Eosin (E&O) staining, Granulomatous Inflammation in the Media. Giant Cells are Marked by Arrows

The patient was managed as a known case of Giant-cell

arteritis (GCA) and was treated with 1 mg/kg daily oral prednisone. After three months' follow-up, and tapering corticosteroids, the patient had no complications.

### 3. Discussion

In our rare case, severe two-sided headache, simultaneous bilateral temporal arteries and inflated and palpable temporal arteries were included in the patient's chief complaints without jaw and tongue claudication, blindness or optic nerve neuropathy.

Temporal arteritis is often diagnosed clinically based on the criteria of the American College of Rheumatology, however it might be frequently misdiagnosed, because it has a wide range of signs and symptoms; thus, high clinical suspicion is required for its diagnosis (2, 6). Increased erythrocyte sedimentation rate (ESR) and C - reactive protein (CRP) give a specificity of 97% and jaw claudication and neck pain are strongly suggestive symptoms, but the gold standard diagnostic tool is the biopsy investigation (9), which should be performed on both sides. In cases of proper treatment with corticosteroid, its prognosis is similar to healthy controls and the major threats of temporal arteritis can be prevented (10).

The people with the age over 50 is one of the five diagnostic criterias and in the largest case series by Bengts-

son et al (1981), all patients with temporal arteritis were above fifty years, and 95% of patient were over 60 years (7). However, the present case report, along with the previous reports, emphasize on consideration of TA in younger ages. Putting aside the juvenile temporal arthritis (7-35 years) (11, 12), few studies have reported cases aged 40 to 50, like Agbanlog and Cruz-Bermudez, who have reported a 41-year old patient with left-sided headaches, fever, blurred vision, diplopia and left jaw claudication, who was free of complications after three months of treatment with prednisone (8), similar to the case in the present report. They have strongly suggested consideration of temporal arteritis (TA) in all ages. Regarding the few cases presented under 50 years, the prevalence of symptoms at this age has to be investigated in further studies, which might help diagnose temporal arteritis in younger patients.

In addition, few cases of bilateral cases have been previously reported, but they were all in elderly patients. Coors and Simon have reported one case of bilateral temporal arteritis, who was 68 years old, presenting with skin erythema and temporal artery swelling on both sides (7). Carmenini et al. also reported a 76 year-old patient, presenting with bilateral claudication satiation, which is a quick response to corticosteroids (13). In reported a case presented with sudden blindness of her left eye. All the above-mentioned studies have reported regression of symptoms short after treatment with corticosteroids, which was similarly found in the present study. Due to the few number of cases of bilateral temporal arteritis, there is a need for further investigation on bilateral cases to determine possible differences in terms of clinical differences and effective treatments.

Thus, the present study is reporting the first case of bilateral temporal arteritis in a patient under 50-years-old. As far as the authors are concerned it has been shown that high clinical suspicions and awareness of physicians are

required for a more accurate diagnosis of temporal arteritis in younger patients to prevent the severe complications with simple treatment with corticosteroids.

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