

CASE REPORT

Curry-assisted diagnosis in the rheumatology clinic

Sarah L. Donaldson^{1,*}, Maura Cobine-Davies¹, Ann W. Morgan²,
Andrew Gough³, and Sarah L. Mackie²

¹Leeds Teaching Hospitals NHS Trust, Leeds, UK, ²Leeds Institute of Rheumatic and Musculoskeletal Medicine, University of Leeds, Leeds, UK, and ³Rheumatology Department, Harrogate and District Foundation NHS Trust, Harrogate, UK

*Correspondence address. 25 Oakdale Glen, Harrogate, North Yorkshire HG1 2JY, UK. Tel: +44-7745700247; E-mail: s.donaldson@doctors.org.uk

Abstract

We report five cases of glucocorticoid-responsive mouth symptoms in polymyalgia rheumatica/giant cell arteritis (GCA); three cases of tongue pain exacerbated by hot/spicy food, a case of scalp pain made worse by eating hot/spicy food and a case of sore tongue as a presenting feature of GCA. These cases emphasize the importance of asking about mouth symptoms and changes in taste when evaluating patients with suspected GCA.

INTRODUCTION

Giant cell arteritis (GCA) is a systemic large-vessel vasculitis (LVV) affecting people older than 50 years. It classically causes headache and ischaemia of cranial structures, resulting in jaw claudication and visual disturbance. GCA may be accompanied by polymyalgia rheumatica (PMR). Likewise, PMR can be accompanied by subclinical GCA/LVV [1]. Early diagnosis of GCA is important to avoid irreversible complications such as visual loss, stroke and scalp or tongue necrosis.

Mouth involvement in GCA is well documented in the literature and usually takes the form of paraesthesiae, glossitis and tongue claudication [2]. Tongue claudication indicates tongue ischaemia and should be detected early in order to avoid progression to necrosis, which happens through stenosis and occlusion of the lingual and contralateral arteries [2]. Tongue necrosis is rare [3–5]. It suggests very active GCA involving multiple vessels [6].

Tongue pain as a feature of GCA without ischaemia or necrosis is less well reported [7, 8]. A case report from 1991 describes a Korean patient with biopsy-proven GCA whose chief complaint was tongue pain and headaches [7]. A 1967 case report of biopsy-proven GCA describes an elderly gentleman with tongue

pain on eating [8]. The author mentions that burning or painful tongue has been reported in three previous cases of GCA [8].

We report five cases of glucocorticoid-responsive mouth symptoms in PMR/GCA; three cases of tongue pain exacerbated by spicy food, a case of scalp pain made worse by eating spicy food and a case of sore tongue as a presenting feature of GCA. Burning mouth syndrome (BMS) is a differential diagnosis of mouth pain exacerbated by spicy foods, but the pain in BMS is not generally glucocorticoid-responsive [9].

CASE REPORT

- (i) A 68-year-old Indian woman presented with a 2-week history of temporal headache, scalp tenderness and jaw claudication on a background of anorexia, weight loss and muscle weakness. She described a 2-year history of upper thoracic pain and a single episode of binocular blurred vision 1 month previously. On examination, her left temporal artery was tender and thickened. The plasma viscosity was 2.2 mPa s (reference range 1.50–1.70 mPa s), CRP 95 mg/l and ESR 120 mm/h. She started 60 mg prednisolone with a good response. A temporal artery biopsy (TAB) confirmed

Received: December 30, 2014. Revised: March 30, 2015. Accepted: April 18, 2015

© The Author 2015. Published by Oxford University Press.

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/4.0/>), which permits non-commercial re-use, distribution, and reproduction in any medium, provided the original work is properly cited. For commercial re-use, please contact journals.permissions@oup.com

GCA. As her glucocorticoid dose was reduced, she experienced headache, gum pain, jaw claudication and tingling tongue pain causing her to stop eating spicy food. Later, she was found to have active LVV. Her thoracic pains improved with higher dose prednisolone, but the jaw claudication and tingling tongue pain induced by spicy food took longer to resolve. She had previously eaten spicy food all her life without problem. She remains troubled by tongue pain on eating even mild spicy food but to a lesser degree than before treatment.

- (ii) A 64-year-old Caucasian gentleman presented with a history of polymyalgic symptoms. His GP had noted raised inflammatory markers (CRP 77 mg/l, ESR 14 mm/h and platelets $567 \times 10^9/l$) and had started him on prednisolone 40 mg with a good response. He had a sore tongue occurring immediately on eating spicy foods, which was glucocorticoid-responsive. He had previously eaten spicy foods without pain prior to diagnosis. Otherwise, there were no features of GCA. The prednisolone was weaned to 7 mg at which point his PMR flared. Methotrexate was added as a steroid-sparing agent and control of both PMR and spice-induced tongue pain was rapidly achieved.
- (iii) A 61-year-old Caucasian woman presented initially to the surgeons with abdominal pain and vomiting as well as fever on a background of weight loss, myalgia, sweats and fatigue. Her CRP was raised (288 mg/l) despite antibiotics. All gastrointestinal investigations were normal including abdominal CT and laparoscopy. The infectious diseases team excluded infection as a cause of her pyrexia. A CT chest, requested to exclude malignancy, revealed thickening of the wall of the arch of the aorta. Following this, a rheumatology opinion was sought and PET/CT confirmed active LVV. She did report headache, jaw claudication and a 'red-raw' tongue sensitive to spicy foods occurring up to an hour after ingestion that was noted in association with the diagnosis of LVV having never occurred on eating spicy food before. On examination, she was tender over the left temporal artery. The tongue

symptoms resolved with glucocorticoid therapy and recurred when her disease flared on glucocorticoid reduction.

- (iv) A 63-year-old Caucasian man presented with systemic inflammatory symptoms. A diagnosis of LVV was eventually made following multiple admissions at different hospitals. His symptoms included abdominal pain, headache, weight loss, fatigue, fevers and chest pain. Interestingly, he experienced scalp tingling immediately after eating spicy foods which preceded diagnosis of LVV by 2 years. He remembers eating curries regularly since 1963 with no problem. At no point was there ever any temporal artery tenderness. A PET/CT showed florid LVV (Fig. 1a–c), which improved with glucocorticoids and methotrexate. He remains troubled intermittently by limb claudication symptoms and scalp tingling associated with eating spicy foods that is glucocorticoid-responsive. He has a weekly curry as his 'test' for disease control. On occasions since diagnosis when he has experienced scalp tingling on eating curry, his inflammatory markers have been raised.
- (v) A 76-year-old Caucasian woman had a 5-year history of BMS (diagnosed by a maxillofacial specialist). The soreness temporarily improved whenever she received short courses of glucocorticoid for a seronegative inflammatory arthritis. This was felt to be unusual since BMS does not usually respond to glucocorticoids. One year after her initial presentation to the maxillofacial department, she reported lumps on her temporal arteries and discomfort on hair brushing. She admitted to only mild headaches and denied jaw claudication and visual symptoms. A few weeks later, however, she reported jaw claudication and increased scalp tenderness. Her temporal arteries were thickened, nodular and tender, and a CRP was 57 mg/l. A TAB revealed florid GCA. Her GP had diagnosed PMR 1 year previously. Her GCA and PMR symptoms resolved with prednisolone 40 mg, but she has struggled over the years when weaning glucocorticoid and has continued to suffer with a glucocorticoid-responsive sore tongue.

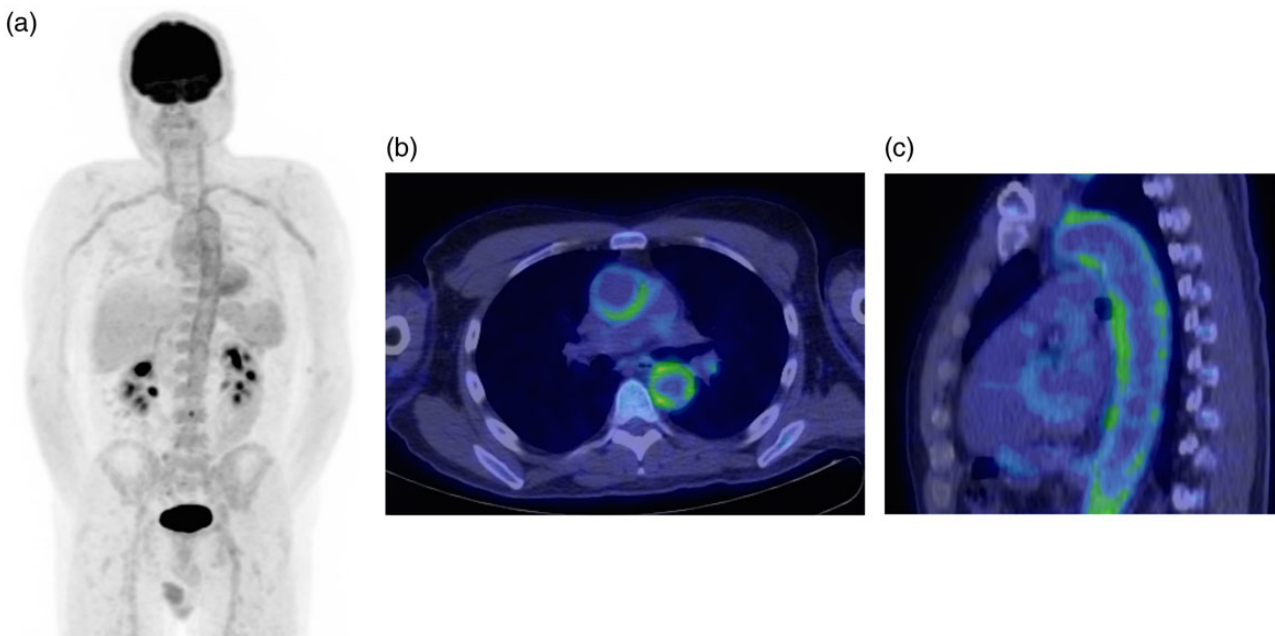


Figure 1: Axial and sagittal fused PET-CT images showing increased activity within the wall of the aorta of Patient 4.

DISCUSSION

The association between GCA and glucocorticoid-responsive burning sensations in the mouth has not, to our knowledge, been previously reported and appears distinct from tongue claudication. This may not be a disturbance of taste (detected via the taste-bud receptors on the tongue), but rather hypersensitivity to capsaicin in chilli peppers. Local ischaemia and acidosis, in combination with systemic inflammation, are the most likely stimuli. Interestingly, four patients reported here had either a long history of symptoms or LVV, with high inflammatory markers. Eating spicy food was not a new habit for these patients and they were not taking medications thought to be relevant to this symptom. We were unable to quantify the exact amount of chilli consumed and there was no suggestion of an allergic response in any case so biomarkers were not measured. In those patients with persistent mouth symptoms despite remission/glucocorticoid treatment, it is important to test for nutritional deficiencies including vitamin B and iron status.

Glucocorticoid-responsive mouth pain induced by chilli in food may be a useful diagnostic clue to GCA, as well as a potential sign of relapse. General practitioners, dentists and oral and ENT surgeons should be aware of glucocorticoid-responsive mouth symptoms as an atypical presentation of GCA.

FUNDING

S.L.M. is funded by a National Institute for Health Research Clinician Scientist Award. The views expressed in this article are those of the authors and not necessarily those of the NHS, the

National Institute for Health Research or the Department of Health.

REFERENCES

1. Moosig F, Czech N, Mehl C, Henze E, Zeuner RA, Kneba M, et al. Correlation between 18-fluorodeoxyglucose accumulation in large vessels and serological markers of inflammation in polymyalgia rheumatica: a quantitative PET study. *Ann Rheum Dis* 2004;**63**:870–3.
2. Maahs GS, Fabricio DD. Tongue necrosis in a patient with cranial arteritis. *Rev Bras Otorrinolaringol* 2007;**73**:717.
3. Cianter M, Adlam DM. Glossodynia and necrosis of the tongue caused by giant cell arteritis. *Br J Oral Maxillofac Surg* 2008;**46**:231–3.
4. Kumar R, Gupta H, Jadhav A, Khadilkar S. Bitemporal scalp, lip and tongue necrosis in giant cell arteritis: a rare presentation. *Indian J Dermatol* 2013;**58**:328.
5. Brodman M, Dorr A, Hafner F, Gary T, Pilger E. Tongue necrosis as first symptom of giant cell arteritis (GCA). *Clin Rheumatol* 2009;**28**:S47–9.
6. Salvarani C, Cantini F, Boiardi L, Hunder GG. Polymyalgia rheumatica and giant cell arteritis. *N Engl J Med* 2002;**347**:261–71.
7. Nuovo J. Temporal arteritis: an atypical presentation. *J Am Board Fam Pract* 1991;**4**:115–8.
8. Henderson AH. Tongue pain with giant cell arteritis. *Br Med J* 1967;**4**:337.
9. Cerchiari DP, de Moricz RD, Sanjar FA, Rapoport PB, Moretti G, Guerra MM. Burning mouth syndrome: etiology. *Braz J Otorhinolaryngol* 2006;**72**:419–23.