It is not always as good as gold. Contact allergy to dental gold as a potential cause of burning mouth syndrome: A case report

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ABSTRACT

Introduction: Inflammation and pain of the oral soft tissues is a common reason for presentation to a dental professional, and with a wide range of differential diagnoses, successful management often provides a significant clinical challenge. One such cause of these symptoms is burning mouth syndrome (BMS), a diagnosis of exclusion based on the presence of a burning sensation of the mouth every day over a period of at least four to six months in the absence of other clinical or laboratory finding to explain the symptoms. Burning mouth syndrome is notoriously difficult to treat and causes significant pain and psychological distress to those who suffer from it. Therefore, thorough investigation and adequate management of potential underlying causes are essential before a definitive diagnosis is made. Case Report: In this report, we discuss the case of an 80-year-old gentleman with a complex medical background referred to our dermatology clinic for epicutaneous patch testing following a diagnosis of Type 1 BMS who had undergone multiple interventions and trialed multiple therapies with very limited improvement in his symptoms. Epicutaneous patch testing revealed contact allergy to gold sodium thiosulfate, suggesting possible contact allergy to dental restorative alloy as an underlying cause for his symptoms. Conclusion: Contact allergy may be a causative or contributing factor in patients who present with burning mouth or who are diagnosed with BMS, as in the case of our report, and epicutaneous patch testing should form an important part of the workup for these patients.

Keywords: Burning mouth syndrome, Contact allergy, Gold

INTRODUCTION

Burning mouth syndrome (BMS) is a complex condition defined by the presence of a burning sensation to the tongue or other oral soft tissue surfaces over a period of at least four to six months in the absence of any other causative clinical or laboratory finding [1]. It results in significant psychological distress to those who suffer from it, and often there is a significant delay between the onset of symptoms and diagnosis. As it must be a diagnosis of exclusion [1], there are a number of other causes for the symptoms of BMS which must be excluded prior to diagnosis, ranging from nutritional deficiencies, systemic medical conditions, medications, and the presence of pathogens in the oral cavity, such as candida [2]. Another important cause that must be ruled out is contact allergy [2], which may be to materials used in dental restorations...
and oral hygiene products. These contact allergies may be identified on epicutaneous patch testing as performed by a dermatologist.

In this case report, we discuss an 80-year-old gentleman referred to our dermatology clinic for epicutaneous patch testing following a diagnosis of Type 1 BMS, where results revealed contact allergy to gold sodium thiosulfate, suggesting possible contact allergy to dental restorative alloy as an underlying cause for his symptoms.

CASE REPORT

An 80-year-old gentleman presented to our clinic with a two-year history of pain to the mid-anterior mandible and burning sensation of the tongue and intermittently the mucosal aspect of his lips, worse in the late afternoon and evening. He also noted subjective xerostomia which was felt to be related to medication. His past medical history was significant, and included hypertension, non-insulin-dependent diabetes mellitus, paroxysmal atrial fibrillation/flutter with dual-chamber pacemaker, benign prostatic hypertrophy, benign paroxysmal positional vertigo, osteoporosis, and a previous fractured sacrum. He had a full, natural dentition with no prosthesis. His regular medications included apixaban, metoprolol, metformin, dutasteride, and tamsulosin, gliclazide, escitalopram, omeprazole, vitamin D, oxazepam as required, and prochlorperazine as required, all of which had been commenced years prior to the onset of oral symptoms. His only known allergy was to penicillin.

Examination by an experienced oral pathologist revealed low-normal saliva flow rate and no evidence of organic disease of the oral mucosa or oropharynx, and no other abnormalities were noted on clinical examination. All regular medications had been commenced years prior to onset of symptoms, and given this assessment by the oral pathologist, xerostomia was considered unlikely to be a significant causative factor for the symptoms. Blood tests including inflammatory markers were normal. Magnetic resonance imaging (MRI) of the head was performed which showed no intracranial or peripheral abnormality found to account for the patient’s symptoms. It was therefore suggested that his symptoms may be consistent with a Type 1 BMS.

His symptoms proved difficult to control despite trial of multiple therapeutic agents. He experienced no relief of his symptoms with the use of opioid analgesics including oxycodone 5 mg as required, and an oxycodone/naloxone combination at a dose of 10/5 mg, and amitriptyline 25 mg taken at night over a period of greater than six months. He was commenced on oral clonazepam, with minimal resolution of symptoms at a dose of 0.5 mg twice daily, which was self-ceased after a period of nine months. He had trialed alpha-lipoic acid, and capsaicin 0.025% mouthwash twice a day over a period of six weeks, again with no significant improvement in symptoms. He was subsequently seen by a pain specialist, and again experienced no improvement of symptoms following trial of an intravenous lignocaine infusion and later a right-sided stellate ganglion block with local anesthetic and corticosteroid. Interestingly, the one agent that did provide some relief was a topical preparation of triamcinolone, neomycin sulfate, gramicidin, and nystatin applied to the tongue, which he has continued to use.

He was subsequently referred to our dermatology clinic for consideration of epicutaneous patch testing to rule out contact allergy as a contributing factor for his presentation. Swabs of the oral mucosa were taken to exclude candida and were negative. Patch testing was performed and included the Australian Baseline Series, the cheilitis series, and his own products including the steroid ointment, lignocaine 2% solution, and a topical capsaicin preparation.

Interestingly, the patch testing revealed contact allergies to hydroperoxides of linalool and gold sodium thiosulfate, and the patient was advised to discontinue use of lignocaine 2% solution and the topical capsaicin preparation.

Given the presence of extensive dental restorations using gold alloy, the results of examination by an experienced oral pathologist revealing low-normal saliva flow rate and no evidence of organic disease of the oral mucosa or oropharynx, and lack of other significant findings on imaging and blood work, this positive result to gold sodium thiosulfate may be a contributing factor to his ongoing and difficult to control symptoms. Of note, the patient’s routine blood work remained within normal limits. As epicutaneous patch testing detects type IV hypersensitivity reactions, abnormalities or other significant findings on routine blood work would not be expected. Specific testing such as radioallergosorbent (RAST) tests were not performed. At the patient’s request, the gold alloy dental restorations have not yet been removed despite the ongoing symptoms, given the major dental intervention required.

DISCUSSION

Burning mouth syndrome

Burning mouth syndrome is defined by the presence of a burning sensation in the oral mucosa, most commonly the tongue, over a period of greater than four to six months, with a lack of other significant, relevant clinical or laboratory findings [1]. Other commonly associated findings are xerostomia and dysgeusia [1, 3]. It is seen most commonly in postmenopausal woman, with studies reporting “oral burning” as a presenting symptom in 10–40% of women presenting for management of symptoms associated with menopause [4]. Type 1 BMS, the subtype with which our patient was diagnosed, is defined as the absence of pain on waking, followed by gradual worsening of burning pain throughout the day, reaching a peak in the
There are a number of previous randomized clinical trials that suggest that topical capsaicin, alpha-lipoid acid, and clonazepam may be indicated for relief of symptoms, as well as psychotherapy and behavioral therapies [3, 5]; however given the complexity of the condition, BMS remains notoriously difficult to resolve, and there appears to be no clear consensus on the cause, risks, or best management for the condition [4].

 Burning mouth syndrome is a diagnosis of exclusion, and causes of symptoms that mimic BMS that must be considered include nutritional deficiencies, not limited to vitamin B or folic acid deficiencies, exposure to dietary allergens, systemic conditions such as diabetes mellitus, medications, presence of infectious agents such as candida, Enterobacter and Klebsiella, and contact allergens found in dental metals, such as nickel, mercury, palladium, and gold [2].

A study by Steele et al. in 2012 [6] conducted a review of 175 patients who presented with a burning mouth and conducted epicutaneous patch testing in 75 of these. Of these patients, 37.3% had positive patch test results, with the most common allergens including nickel, balsam of Peru, and gold sodium thiosulfate. Interestingly, the results of allergen avoidance or removal of the allergens and offending dental restorations were mixed, with some showing significant improvement, and others reporting no change.

The consensus from a range of articles [6–8] where epicutaneous patch testing has been performed in patients who present with burning mouth or a diagnosis of BMS is that contact allergy may be a causative or contributing factor for these patients, and that epicutaneous patch testing should form an important part of the workup.

The role of dermatologist referral

Epicutaneous patch testing is a common procedure performed by dermatologists and remains the mainstay for diagnosis of contact allergies. It involves the application of small patches coated in known allergens in a prescribed order (known as a series) to the patient’s skin, most often on their back. These are left in place for at least 48 hours before they are removed. The result for each individual patch is then interpreted or “read” according to the skin reaction which may vary from no notable change, to mild erythema, to confluent blistering of the skin. This “reading” procedure is repeated again approximately 72 hours later.

The Australian Baseline Series, as performed for this patient, is a set of the 60 most common contact allergens in Australia produced by Chemotechnique Diagnostics (Vellinge, Sweden). It includes nickel, a number of topical corticosteroid preparations, fragrance mixes, and commonly used dyes.

Allergens found in dental materials that are most commonly identified on epicutaneous patch testing include amalgam, ammonium tetrachloroplatinate, ammoniated mercury, nickel sulfate, tin, gold sodium thiosulfate, potassium dichromate, cobalt, palladium, copper, and methylhydroquinone [9, 10]. As a result, a number of series of patches specific to dentistry exist, and may have a role in not only investigating potential contact allergy due to the presence of certain substances in a patient’s dental restorations, but also to investigate for possible contact allergy in dentists and dental workers who may considered to be at risk due to occupational exposure.

Contact allergy to dental gold

Though named Contact Allergen of the Year in 2001 [11], the role of gold as a contact allergen has remained controversial. Sources of allergic contact dermatitis to gold are wide ranging, and include jewelry, various occupational exposures, medical devices and implants such as gold-coated endovascular stents and dental alloys, and other therapeutics containing gold [11]. Positive patch test results to gold sodium thiosulfate are seen commonly, with reported rates as high as 30.7% in a study assessing patch test results of 852 patients from 2000 to 2009 [12]. Interestingly, a review by Chen and Lampel [11] noted that only 0–54% of positive patch test results for gold were actually considered clinically relevant, with a rate of 10–15% in two of the largest studies they reviewed.

Further adding to the controversy, the process by which elemental gold undergoes ionization to a suitable extent to become a hapten capable of eliciting an allergic response, especially in the oral cavity, remains poorly defined, but is hypothesized to rely on the presence of amino acids in bodily fluids and may be assisted by the presence of other metals in the specific alloy [11, 13]. Despite this, a number of case reports of contact allergy to dental gold have been published, and the review by Chen and Lampel [11] reports the presence of studies to support a correlation between the concentration of gold detected in a patient’s blood and the extent of gold dental restorations, and between the concentrations of gold in a patient’s saliva and the extent of gold dental restorations.

Oral manifestations of allergic contact reaction to gold are reported to include perioral dermatitis, cheilitis, burning mouth, stomatitis, ulceration, and lichenoid reactions [11]. Ditrichova et al. [14] performed epicutaneous patch testing on 25 patients with lichenoid changes in the oral cavity. Positive results were noted in 15 patients, with a total of 31 positive reactions. These positives were most frequently to dental metals including mercury, amalgam, palladium, and cobalt, and 6.4% of the positive results were for gold.

Patient selection prior to patch testing for potential contact allergy to dental materials is important, and patch testing is said to be indicated only where objective signs and symptoms such as pain, stomatitis, cheilitis, or lichenoid lesions are present [9]. Routine testing in patients with an absence of symptoms is unlikely to be appropriate, and the relevance of positive results cannot
be suitably established [9]. Schaffran et al. [15] conducted a study into the prevalence of gold sensitivity in patients with gold dental restorations but no symptoms. They showed that 33.8% of the 71 patients patch tested had a positive result for gold sodium thiosulfate, even in the absence of symptoms, further highlighting the importance of careful correlation between symptoms and patch test results.

The appropriate course of action once dental gold is identified as a contact allergen is also poorly defined. An article by Raap et al. in 2012 [9] suggests that dental materials should not be removed immediately following the discovery of a positive patch test result, and that careful assessment of the association between the discovered allergen and its presence in the patient’s dental restorations must occur before it is removed or exchanged.

As previously described, studies show mixed results for improvement in symptoms once the dental gold is removed [13], and this must be considered before embarking on the invasive and costly mission of removing the allergen, especially in patients with multiple medical comorbidities, such as ours. One method that could be considered is for the patient’s dentist to prepare a vacuum formed acrylic stent, similar to a mouth guard, to reduce direct contact with the allergen and assess if any symptomatic improvement results.

CONCLUSION

Inflammation and pain of the oral soft tissues is a common reason for presentation to a dental professional and often provides significant challenges for management, with a wide range of possible underlying causes. Burning mouth syndrome remains a diagnosis of exclusion, and it is essential that all patients who are considered to have this diagnosis undergo a thorough workup. A number of studies suggest that contact allergy may be a causative or contributing factor in patients who present with burning mouth or who are diagnosed with BMS, and that epicutaneous patch testing should form an important part of the workup for these patients. Early referral to a dermatologist may assist to identify these potential causes early. The appropriate course of action following identification of a possible contact allergy to restorative dental work remains poorly defined, and the clinical relevance of a positive patch test results must be carefully established. Despite this, identification of a potential cause for a patient’s symptoms may provide them with both hope and relief, making it imperative that all possible causes are investigated before a definitive diagnosis is given.

REFERENCES


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