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Unusual oral mucositis

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CLINICAL PRESENTATION

A 53-year-old woman, a sales negotiator by profession, was referred to the Oral and Maxillofacial Diseases Clinic of Kuopio University Hospital for oral mucosal changes. The patient had epilepsy and allergic rhinitis. Her regular medication included carbamazepine and clonazepam, as well as occasional cetirizine, mometasone furoate, salbutamol, and budesonide. Additionally, the patient was taking vitamin B and lactic acid bacteria supplements. The patient had aspirin, environmental, and several plant food allergies. She had no history of smoking or alcohol use. The patient's mother had Sjögren syndrome.

On examination the patient appeared to be in good general health. Her natural dentition was in good condition with several restorations. There was extensive diffuse erythema and a few fibrin-coated ulcerations in the left buccal and upper labial mucosa. The left buccal mucosa felt indurated on palpation. Erythema was detected in the left side of the maxillary gingiva and hard palate, and minor white striae were noted bilaterally in the floor of the mouth. In the left oropharyngeal and left tonsillar pillar areas, ulcerative lesions with erythema were found (Figure 1). There were no periodontal pockets, but slightly increased mobility in the left mandibular third molar and second premolar was detected. Panoramic radiograph revealed noncontributory findings. Palpation of the neck revealed no abnormalities. The patient reported having no skin or genital lesions. There was no history of physical or chemical trauma to the oral mucosa. The oral mucosal lesions were mostly asymptomatic, but the patient had noticed occasional bleeding when brushing her teeth.

Four years before the current referral, the patient had visited a general dental practitioner because of oral mucosal lesions. The lesions were reported to be lichen planus−like and apparently symmetric. Later, red lesions and hemorrhagic ulceration were noted in the left buccal mucosa, lichenoid lesions in the right maxillary gingiva, and erythema in the left maxillary gingiva. The patient was then referred to the Oral and Maxillofacial Diseases Clinic of Lapland Central Hospital. Clinical examination revealed erythema in the left maxillary gingiva, white striae in the mandibular anterior sulcular mucosa, and reticular lichen planus−-like lesions in the right buccal mucosa and more pronounced in the left buccal mucosa. Spontaneous hemorrhage was noted in the left buccal mucosa. A clinical diagnosis of oral lichen planus (OLP) was made. No biopsy was taken. The patient was prescribed a 3-week course of 0.1% topical betamethasone valerate cream. Five weeks later, the mucosa in the right side of the oral cavity and mandibular anterior sulcular area was normal and asymptomatic. The erythema in the left maxillary gingiva had diminished. The clinical appearance of the left buccal mucosa was better but there was still some bleeding.

DIFFERENTIAL DIAGNOSIS

The clinical differential diagnoses of chronic mainly erythematous and ulcerative oral mucosal lesions include infectious diseases, nutritional deficiencies, immune-mediated disorders, preneoplastic lesions and neoplasias, and idiopathic conditions.

Infectious diseases

Oral candidiasis often manifests as erythematous lesions, although they are typically nonulcerative and often symmetric. The years-long duration of the oral lesions and lack of strong predisposing factors to oral candidiasis do not favor candida as a primary cause of the lesions. However, candidiasis superimposed on another pathology is a reasonable consideration in this case.

Nutritional deficiencies

Deficiency in vitamin B2, B6, or B12 or iron may cause oral mucosal erythema that typically affects the tongue and angles of the mouth. Because the patient did not have glossitis or angular cheilitis or any systemic signs or symptoms such as fatigue associated with vitamin B or iron deficiencies, these were not considered likely in the present case.

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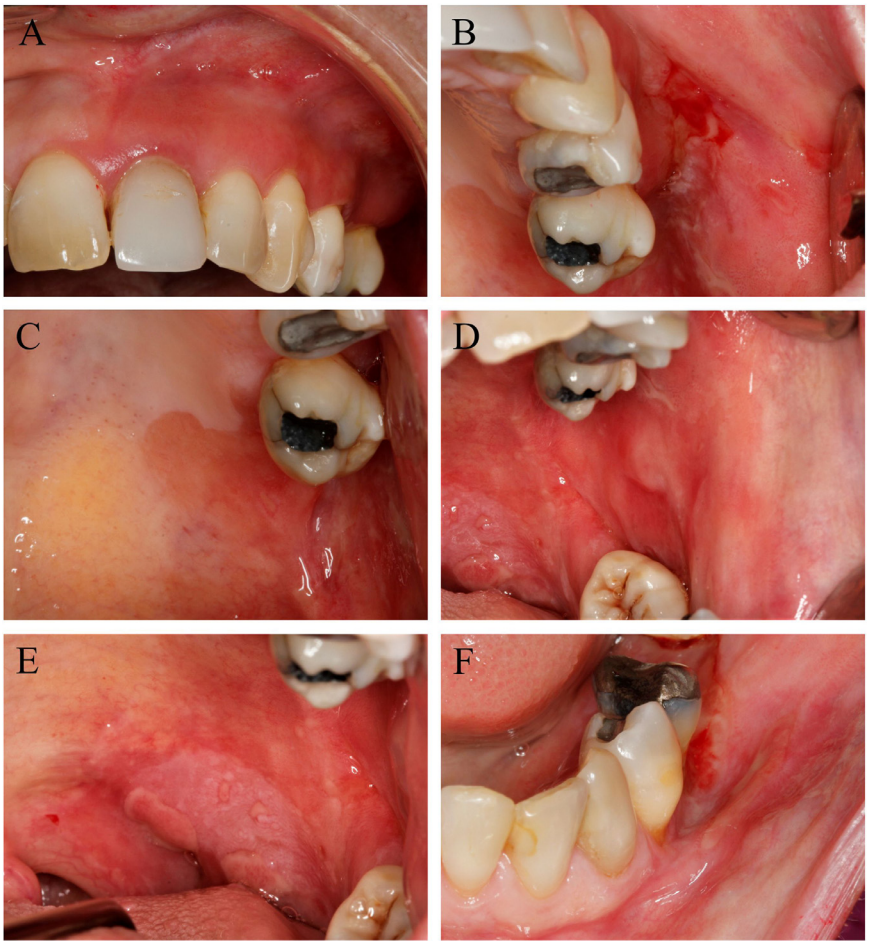

Fig. 1. Erythema of the left side of **(A)** maxillary gingiva, **(B)** maxillary buccal sulcus, **(C)** palatal mucosa, **(D)** buccal mucosa, **(E)** anterior pillar and oropharynx, and **(F)** mandibular buccal sulcus. Ulceration is also seen in the **(B)** buccal sulcus and **(E)** anterior pillars.

Immune-mediated disorders

Many different agents, including resins, metals, flavorings, preservatives, and drugs, can cause acute or chronic oral allergic reactions, which may manifest as oral mucosal erythema, edema, ulceration, and lichenoid lesions as well as symptoms such as burning and itching.[1] Sometimes the offending agent cannot be identified. Oral lesions of contact allergy to mercury or copper sulfate in amalgam fillings are typically local and in direct contact with the filling(s). In our case, however, some of the lesions were present in areas with no contact with dental fillings. Because allergic contact stomatitis due to food or oral hygiene products normally causes widespread lesions, it was not considered likely in our case.

Plasma cell gingivitis or gingivostomatitis (PCG) is an acute allergic reaction to certain agents present in, for example, chewing gums, peppers used for cooking, herbal toothpastes, and mint candies.[2] It was quite common in the late 1960s and early 1970s but has since almost vanished. PCG shows marked erythema of gingiva and can sometimes also involve the palatal mucosa, lips, or tongue. The condition is usually painful and ulceration is uncommon. PCG was considered as a diagnosis but was thought to be unlikely because the patient was not using any of the reported offending agents and the mucositis was chronic and almost exclusively unilateral. In addition to acute hypersensitivity reactions, systemic drug administration may cause chronic lichenoid or lupus- or pemphigoid-like reactions.[3] Although chronic allergic reaction to a systemic drug was included as a differential diagnosis, no temporal relationship between the drugs used and the appearance of oral lesions could be identified.

Lichen planus is a chronic mucocutaneous immune-mediated disease of unknown etiology. It has a prevalence of 1% to 2% and typically affects middle-aged persons, with a female predominance. OLP may be present in various clinical forms (reticular, papular, atrophic/erythematous, plaque-like, erosive/ulcerative, and bullous). The most common form is reticular OLP, which manifests as white striae in the oral mucosa and

is relatively often accompanied by the other forms, especially the atrophic and ulcerative. OLP lesions are typically symmetric and can affect any oral mucosal site but are most often seen in the buccal mucosa, gingiva, and tongue.[4] Oral lichenoid lesions (OLLs) are clinically and histopathologically similar to OLP and occasionally indistinguishable from OLP. In contrast to OLP, OLLs are considered to have an etiologic factor, such as contact allergy or drug reaction, although this factor cannot always be found. OLL may present as unilateral lesions.[5] In our patient, the previous and present oral lesions had features of OLP and OLL. The asymmetric presentation would clinically rule out OLP in this case. In addition, the large areas of erythema without coexisting white striae and sharp demarcation of the erythema in areas did not suggest OLP or OLL.

Mucous membrane pemphigoid (MMP) and pemphigus vulgaris (PV) belong to a group of autoimmune blistering diseases that typically affect the oral mucosa.[6] MMP and PV commonly affect persons over age 50, and MMP has a female predilection. MMP and PV manifest as erythema, vesicles, or bullae, erosions and ulcerations in the mucosal surfaces (oral, nasal, pharyngeal, laryngeal, esophageal, genital, anal and/or conjunctival) and skin, although skin involvement is rare in MMP.[6] The oral cavity is usually the first site of lesions, and they are typically located in the buccal mucosa, gingiva, palatal mucosa, and oropharynx. Ocular manifestations are more common in MMP than in PV and include conjunctivitis and subepithelial fibrosis leading to symblepharons, entropion, and trichiasis and may eventually cause blindness. Our patient reported never having any blisters in the oral mucosa or skin or ocular symptoms. In addition, blisters were not seen on clinical examination. Despite not receiving systemic corticosteroid or other immunosuppressive therapy, the patient's condition did not progress, and that would make at least PV unlikely in this patient.

Preneoplastic lesions and neoplasias

Erythroplakia is a red mucosal patch that cannot be clinically or pathologically diagnosed as any other condition.[7] Almost all erythroplakias present dysplasia or carcinoma in situ on histopathologic examination. Erythroplakia is typically solitary and well demarcated and has a bright red velvety surface with a size less than 1.5 cm in diameter.[7] Erythroplakic lesions have been reported in association with oral lichenoid disease.[8] In the present case, the erythematous mucosa was locally sharply demarcated resembling erythroplakia. The large size of the lesions made erythroplakia unlikely as a primary diagnosis. However, erythroplakia arising in association with oral lichenoid disease was considered as a possibility in this case.

Erythema, ulceration, and induration of the oral mucosa with no apparent cause could be a sign of squamous cell carcinoma. However, the extent of the lesions and long duration without progression to clinically evident malignant disease did not favor squamous cell carcinoma in the present case.

Idiopathic conditions

Plasma cell mucositis (PCM) is a rare, benign idiopathic disorder involving the upper aerodigestive tract.[9] The condition is typically chronic and may be associated with synchronous or metachronous autoimmune or immune-mediated diseases.[10] The patients are most often over age 50, and a slight male predominance has been reported.[11] The most common sites of involvement for PCM are the gingiva, lips, palatal and buccal mucosa, and larynx.[10,11] PCM typically manifests as intense erythema of the mucous membranes. The surface of the erythematous lesions may be velvety, papillomatous, nodular, or cobblestone-like. Symptoms of PCM include pain, dysphagia, hoarseness, and sore throat.[10] PCM was included in the clinical differential diagnosis because our patient had many of the aforementioned signs.

DIAGNOSIS AND MANAGEMENT

A swab was taken from the oral cavity for yeast culture and showed small amounts of *Candida albicans*, considered to represent a carrier state.

Due to the unusual clinical presentation, biopsies were considered necessary. Before the biopsies, the lesional areas (left buccal mucosa, labial mucosa, and gingiva) were stained with toluidine blue. Positive staining was mostly observed in the ulcerative areas. Biopsies were performed from the left mandibular buccal sulcus, left buccal mucosa, left maxillary buccal sulcus, and left side of the hard palate.

Routine histopathologic examination revealed a parakeratotic, occasionally acanthotic, stratified squamous epithelium covering some of the specimens, whereas others were devoid of epithelium. Mild epithelial spongiosis and exocytosis was present. A chronic plasma cell and lymphocytic inflammation was seen subepithelially, with intense plasma cell infiltrate present in all the specimens (Figure 2; a high-resolution version of this slide is available as eSlide VM06399). An abscess-like cavity was seen in 1 of the specimens. No dysplastic or malignant features were noted.

Immunohistologic staining of the specimens was performed to rule out plasmacytoma and IgG4-related disease. Kappa and lambda staining revealed polyclonal plasma cell infiltrate (Figure 2). CD56 and cyclin staining were negative. The ratio of IgG4-positive plasma cells was low.

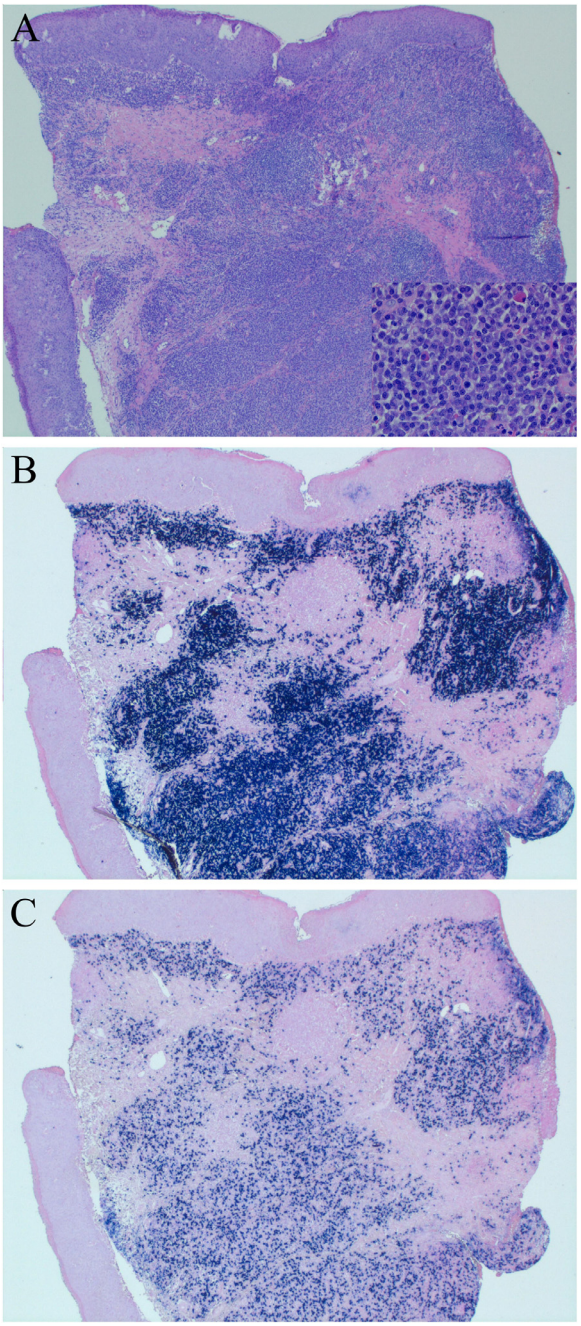

Fig. 2. **(A)** Hematoxylin and eosin−stained section of the left buccal mucosa shows parakeratinised epithelium with mild spongiosis and exocytosis and dense plasma cell infiltrate in the subepithelial connective tissue (25× magnification, inset 200× magnification). **(B)** Lambda and **(C)** kappa immunohistochemical staining with BCIP/NBT as chromogen shows polyclonality of the plasma cell infiltrate. A high-resolution version of this slide for use with the Virtual Microscope is available as eSlide: VM06399.

Management of the oral lesions with a 2-week course of 0.1% triamcinolone acetonide solution 3 times a day was initiated, and this alleviated the symptoms.

The patient was referred to an otorhinolaryngologist for examination and biopsy of the left tonsillar pillar lesions. Oral and oropharyngeal mucosal lesions as described were detected, in addition to a mobile, nontender lymph node palpated in the right side of the neck in the area of carotid bifurcation. A biopsy was taken from the left palatoglossal arch. Histopathologic analysis of the specimen showed an acanthotic stratified squamous epithelium with a parakeratotic surface. The tips of the rete ridges were bulbous and intercellular spaces were widened. Subepithelially, an intense chronic inflammatory cell infiltrate was present. An immunofluorescence examination of the oral mucosa was recommended.

The patient was referred for ultrasound examination of the neck, which showed signs of past thyroiditis but was otherwise within normal limits.

Several blood tests, including complete blood count and assays for plasma glucose, serum vitamins B6 and B12, folate, iron, transferrin receptor, erythrocyte sedimentation rate, IgG1-4, IgA, IgE, tissue transglutaminase-immunoglobulin A, antineutrophil cytoplasmic antibodies, antinuclear antibodies, extractable nuclear antigen, human immunodeficiency virus antigen/antibody, *Treponema pallidum* antibodies, angiotensin converting enzyme (ACE), and lysozyme (LZ) and urine analysis were done to rule out systemic diseases. The tests showed somewhat elevated levels of ACE, LZ, and vitamin B6. To rule out sarcoidosis, a pulmonologist was consulted and a chest x-ray was taken. It was within normal limits, and no additional pulmonary examinations were deemed necessary because the patient had no respiratory symptoms.

Six months later, the serum LZ level was elevated to a similar degree as previously, but ACE showed a further increase. An internist and pulmonologist were consulted on the significance of the abnormal results. The elevated LZ and ACE levels were suspected to be secondary to oral mucosal inflammatory condition and therefore were nonspecific findings. However, a control chest x-ray was recommended. This was taken 5 months after the first imaging and was again within normal limits.

Later, a fresh tissue sample of the oral mucosa was taken for immunofluorescence examination to rule out autoimmune oral mucosal diseases such as mucous membrane pemphigoid. This showed positive IgM at vessel walls, which suggested vasculitis. However, the previous histopathologic examinations did not show any features of vasculitis, nor did the patient have any clinical signs of vasculitis.

A working diagnosis of plasma cell mucositis was made based on the clinical, histopathologic, and radiologic findings and laboratory analyses.

About 1 year after the first consultation at Kuopio University Hospital, a white and erythematous

lichenoid lesion was noted in the right mandibular sulcular area, in addition to the lesions in the left side of the oral mucosa. Later, the patient developed genital symptoms and visited a gynecologist. The vulva and vaginal mucosa were erythematous and the vaginal mucosa was hemorrhagic. The minor and major labia were fused. Clinically, the lesions were considered consistent with lichen planus. However, a biopsy from the vulva yielded a histopathologic diagnosis of chronic nonspecific inflammation. The patient has regular follow-ups with her gynecologist.

The patient's oral condition has been monitored for 34 months. The oral mucosal lesions have shown clinical fluctuation but remained relatively unchanged. The patient has occasionally used a course of topical 0.1% triamcinolone acetonide suspension or 0.1% betamethasone valerate cream with a reduction of oral symptoms. Changing the toothpaste to a gentle formula has reduced the bleeding on brushing. The patient reported using olive oil to moisturize the oral mucosa. The patient has regular dental check-ups and periodontal scaling twice a year.

To date, the patient is in good general health and has no symptoms suggestive of any general conditions that could explain the oral mucosal lesions. However, she was recently diagnosed with a mild chronic cholecystitis and cholesterolosis.

DISCUSSION

A diagnosis of plasma cell mucositis was initially made based on the clinical, histopathologic, imaging, and clinical laboratory findings. However, the lichenoid lesions seen clinically initially and again later in this patient are not typical in PCM. This could suggest that the patient has a combination of plasma cell mucositis and oral lichenoid disease. In fact, a case of plasma cell mucositis on a background of lichen planus has been previously reported.[9] In the present patient, however, none of the clinically lichenoid lesions were biopsied, and none of the biopsies taken showed lichenoid inflammation histopathologically.

Abscess-like cavity formation seen histopathologically in 1 of the specimens is not a typical finding in PCM. In our case, the abscess-like finding could be explained by secondary infection of the ulcerated oral mucosa.

During follow-up, the patient developed genital inflammatory mucosal lesions that were clinically lichenoid, but a biopsy did not confirm this impression, showing nonspecific chronic inflammation. PCM is clinically and histopathologically similar to plasma cell vulvitis/balanitis (Zoon's vulvitis/balanitis or vulvitis/balanitis circumscripta plasmacellularis).[12–14] In the present case, the oral and genital lesions shared some similarities clinically, but the histopathologic findings were different. It is therefore impossible to say whether they represent manifestations of the same disease spectrum.

PCM is an exceedingly rare chronic inflammatory mucosal disorder confined to the upper aerodigestive tract.[9] It has also been called circumorificial plasmacytosis, idiopathic plasmacytosis, mucous membrane plasmacytosis of the upper aerodigestive tract, oral papillary plasmacytosis, and plasma cell orificial mucositis.[10,15] To date, over 50 cases of PCM have been described in the literature.[11,15-19]

The etiology of PCM is uncertain, although many patients have simultaneous autoimmune disorder such as seronegative rheumatoid arthritis, Sjögren syndrome, autoimmune hepatitis, polymyositis, celiac disease, or type I diabetes mellitus.[10,13] An elevated erythrocyte sedimentation rate is sometimes seen in conjunction with PCM.[10]

The average age of patients with PCM is 56 years, and the disease seems to affect males slightly more often than females.[10,11] PCM typically affects the supraglottic larynx, lips, gingiva, and palatal mucosa, but it may be seen in the glottic larynx, pharynx, nasal mucosa, buccal mucosa, and tongue.[11,13] PCM manifests clinically as bright erythema of the mucosa with a cobblestone-like, granular, papillomatous, or nodular surface change. Swelling of the attached gingiva or lips may occur.[10,11] Ulceration is shown to be present in over one-fifth of PCM cases.[11] The mucosa may appear thickened and firm on palpation as if scarred[10]; this feature was present in the buccal mucosa of our patient. PCM lesions are typically multifocal and bilateral. The lesions in the present case were mostly unilateral. Symptoms of PCM include pain, dysphagia, soreness, cough, and voice changes such as stridor, hoarseness, and dysphonia.[9,10]

Clinical differential diagnoses of PCM include sarcoidosis, granulomatosis with polyangiitis (GPA, Wegener's granulomatosis), rhinoscleroma, mucous membrane pemphigoid, pemphigus, lichen planus, erythroplakia, squamous cell carcinoma, fungal infections, plasma cell gingivitis, and lymphoproliferative disorders.[9,10] Additionally, differential diagnoses of PCM confined to the lips comprise allergic contact mucositis, cheilitis granulomatosa, Melkersson-Rosenthal syndrome, plasmoacanthoma, and angioedema.[9,10]

Typical histopathologic features of PCM are epithelial hyperplasia and spongiosis with long and narrow rete ridges and a dense polyclonal plasma cell infiltrate in the subepithelial connective tissue.[9,15] Some cases show lymphocytes as well.[15,20] Ulceration and nonspecific secondary inflammation may occur.[9]

Histopathologic differential diagnoses of PCM include other upper aerodigestive tract conditions with dense plasmacytic infiltrate. Extramedullary plasmacytoma is typically a local mass and has a monoclonal

plasma cell population that can be demonstrated with immunohistochemistry. Of note, an unusual case of PCM with evidence of a monoclonal plasma cell infiltrate has been reported.[10] PCG cannot be differentiated from PCM histopathologically, but clinical features must be taken into account. PCG is a hypersensitivity reaction with an acute onset and is typically resolved when the use of the allergenic agent is eliminated.[10] Other, albeit more unlikely, histologic differential diagnoses include plasma cell granuloma, lymphoplasmacytic lymphoma, and plasmoacanthoma.[10] In addition, syphilis and plaque-related gingival inflammation show considerable plasma cell infiltrate. Hence, the diagnosis of PCM requires clinical-pathologic correlation.

In the immunofluorescent examination, IgM antibodies were found in vessel walls, which suggests leukocytoclastic vasculitis, possibly pointing to Behçet disease or GPA. However, no features suggestive of vasculitis were present in the routine histopathologic examination. In addition, because there was no ocular or dermal involvement and the genital lesions did not resemble aphthae clinically or histopathologically, our patient does not meet the diagnostic criteria for Behçet disease.[21] Although vasculitis could be difficult to observe in oral mucosal biopsies in GPA, the inflammatory cell infiltrate adjacent to vessels would show mixtures of histiocytes, lymphocytes, neutrophils, eosinophils, and multinucleated giant cells.[22] In the present case, the inflammatory infiltrate consisted mainly of plasma cells.

The elevated ACE levels seen in our patient can be indicative of sarcoidosis. In chronic sarcoidosis, pulmonary symptoms are typical but about one-fifth of patients may be asymptomatic. Chest x-ray shows signs suggestive of sarcoidosis in almost all cases at some point of the disease process.[23] In our case, chest x-ray findings were normal. In addition, several oral biopsies taken did not reveal signs of granulomatous inflammation typically seen in sarcoidosis.

The treatment of PCM usually consists of topical, intralesional, or systemic corticosteroids. This treatment may provide only temporary relief.[9] In addition, PCM has been treated with variable success using resection, CO_2 laser, argon plasma coagulation, low-dose radiation, vitamin A derivatives, topical tacrolimus, adalimumab, infliximab, golimumab, cyclosporin, ketoconazole, chlorhexidine mouthwash, methotrexate, chlorambucil, dapsone, mycophenolate mofetil, colchicine, azathioprine, cyclophosphamide, vincristine, and griseofulvin.[9,10] Recently, injectable platelet-rich fibrin has been tried as a treatment for PCM with success.[20] When the disease compromises the distal airways, tracheostomy may be needed in severe cases.[15]

The prognosis of PCM is considered favorable, and on occasion PCM may regress spontaneously.[24] To date, progression of PCM to plasma cell neoplasm or lymphoma has not been reported.[9] One case report described squamous cell carcinoma arising in the lip of a patient with a 20-year history of PCM, but this association could be incidental.[25]

CONCLUSIONS

PCM is a rare and diagnostically challenging cause of oral mucositis. The diagnosis of PCM is based on correlation of clinical and histopathologic findings and may require multiple biopsies as well as imaging and clinical laboratory investigations. A multidisciplinary approach is typically required in the diagnosis and treatment of PCM.

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