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INTRODUCTION
Sarcoidosis is a chronic multisystemic disorder of unknown origin, characterized by the formation of nonnecrotizing granulomas, composed of mononuclear/macrophages and surrounded by T-lymphocytes. Jonathan Hutchinson reported the first case of sarcoidosis in 1869, but the term sarcoidosis was introduced by Boeck in 1899.[1] Sarcoidosis occurs worldwide, with an average incidence of 16.5/100,000 in men and 19.0/100,000 in women. In the USA, sarcoidosis is more common in people of African-Americans descent than Caucasians, with an annual incidence reported as 35/100,000 and 10/100,000, respectively. The disease most commonly affects young adults, with a slight predilection for females. The incidence is highest for individuals younger than 40 years and peaks in the age group from 20 to 29 years; a second peak is observed for women over 50 years.

All organ systems may be involved, but the most frequently affected organ is the lung. Involvement of the skin, eye, liver, and (particularly hilar) lymph nodes is also common. The most frequently affected organs in the head-and-neck region are salivary glands, with the parotid gland affected in about 6% of the patients, and the cervical lymph nodes. Oral involvement of sarcoidosis is very rare and usually occurs on the buccal mucosa, lips, gingiva, tongue, and palate. Approximately one-fourth of all reported intraoral cases are represented by intraosseous lesions affecting either jaw.[5]

The diagnosis of sarcoidosis is based on clinical picture and radiological features and on the microscopic finding of nonnecrotizing granulomatous inflammation. Other diseases mimicking sarcoidosis, mainly various infections and systemic granulomatoses, must be excluded. There is no single diagnostic test for sarcoidosis, and the presence of granulomas alone does not establish the diagnosis. Histological confirmation is not needed for Löfgren’s syndrome (erythema nodosum, bilateral hilar lymphadenopathy on chest radiograph, and arthralgia), Heerfordt’s syndrome (uveitis, swelling of the parotid gland, chronic fever, and in some cases, facial palsy), and asymptomatic bilateral hilar lymphadenopathy.

We present a case of oral sarcoidosis with an unusual presentation on the oral mucosa.

CASE REPORT
A 59-year-old female was referred to the Department of Dentistry in July 2013. The main complaints were 3-month lasting painless aphthous lesions of the oral mucosa. The lesions were predominantly located on the lower lip in the beginning but subsequently developed also on the buccal mucosa. There were multiple erosive lesions (maximum size 18 mm × 7 mm) present on the buccal mucosa. There were multiple erosive lesions (maximum size 18 mm × 7 mm) present on the buccal mucosa bilaterally during the intraoral examination. The lesions were slightly

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prominent, with a submucosal palpable mass [Figures 1 and 2]. There was no erythema in the adjacent mucosa. No systemic symptoms were present, and cervical lymphadenopathy was absent. The patient reported a personal history of pulmonary sarcoidosis lasting 8 years. The involvement of the brain at the time of original diagnosis was suspected (not proven). She has been on long-term corticosteroid treatment and without any apparent clinical or laboratory symptoms of active sarcoidosis since the initial diagnosis. Medical history of the patient included prednisolone 10 mg daily, omeprazole, and levothyroxine (for hypothyroidism) and telmisartan (for hypertension) at the time of examination.

Clinical appearance of the lesions did not allow the proper diagnosis. Autoimmune disorder or erosive form of oral lichen planus was considered in the differential diagnostics. Therefore, the biopsy of one lesion was performed during the first visit.

Microscopically, besides superficial erosions with mixed inflammation, there were several nonnecrotizing epithelioid granulomas deeper in the submucosa, composed of CD68-positive macrophages and surrounded by lymphoplasmacytic infiltrate [Figure 3]. The presence of acid-fast rods and mycotic microorganisms was excluded by Ziehl–Neelsen and Grocott stainings.

The diagnosis of oral sarcoidosis was confirmed by histopathological examination. No change of systemic steroid medication was recommended by the responsible specialist, but topical treatment was administered. The patient received 0.1% dexamethasone gel three times daily and one intraleisonal application of 7 mg of betamethasone. All oral lesions subsequently disappeared during 3 weeks.

**DISCUSSION**

Sarcoidosis has been known for more than 120 years but remains a challenging diagnosis. The absence of reliable markers, multiple organ involvement, and various clinical and radiological findings make the diagnosis challenging issue. About 30%–50% of the patients present with extrapulmonary manifestations of the disease. If typical pulmonary lesions are absent, the diagnosis is based on histological verification and exclusion of other disorders. Modern imaging techniques, such as positron emission tomography/computed tomography and novel biomarkers, such as neopterin, interleukin-2 receptor, and serum angiotensin converting enzyme, may help in the diagnostics. The skin manifestations of sarcoidosis are common (about 15%) and may have various clinical appearance. Subcutaneous lesions, maculopapular rash, erythema nodosum, keloids, and changes in pigmentation can all be seen in skin sarcoidosis. The head-and-neck region can be affected in up to 15% of the cases. Salivary gland involvement resulting in xerostomia is the most frequent. The minor salivary glands can be affected as well; the nose and larynx can be also affected (in around 0.5%–6%). The manifestation of sarcoidosis in the oral cavity is much less common and may be very rarely the first presentation of the disease.

The first description of oral sarcoidosis was reported by Schroff in 1942. He reported a suspected case of sarcoid granulomas of the oral mucosa. Poe published the first histologically confirmed case of sarcoidosis affecting the mandible 1 year later. Since then, only around eighty cases of oral sarcoidosis have been described in the literature. The most common sites of oral sarcoidosis include jaw bones, followed by buccal mucosa, gingiva, floor of the mouth, tongue, and palate. Only few cases with isolated buccal mucosal involvement have been reported [Table 1]. The most common buccal
Table 1: Adapted from Bouaziz et al.[5]

| Author                  | Year | Age | Race | Sex | Chief complaint | Treatment               |
|-------------------------|------|-----|------|-----|-----------------|-------------------------|
| Schroff J.              | 1942 | 48  | C    | F   | Swelling        | Not reported            |
| Kolas S.                | 1960 | 15  | C    | M   | Multiple lumps  | Steroids                |
| Roche W.C.             | 1966 | 33  | B    | M   | Swelling        | Oxygen                  |
| O‘Brien JJ              | 1969 | 30  | M    | F   | Pain            | Not reported             |
| Hoggins GS, Allan D    | 1969 | 45  | C    | M   | Swelling        | Surgery, radiation      |
| Gold RS, Sager E       | 1976 | -   | -    | -   | Pain            | Not reported             |
| Greer RO, Sanger RG    | 1977 | 37  | C    | F   | Swelling        | No treatment            |
| Orlian AI, Birnbaum M  | 1980 | 43  | C    | F   | Swelling        | Surgery                 |
| DeLuke DM, Scuibba JJ  | 1985 | 35  | B    | F   | Swelling        | Steroids                |
| Klesper B et al.       | 1994 | 16  | C    | F   | Swelling        | Not reported             |
| Blinder D et al.       | 1997 | 43  | C    | F   | Swelling        | No treatment            |
| Piattelli A et al.     | 1998 | 44  | C    | F   | Nodules         | No treatment            |
| Jackowski K et al.     | 2005 | 39  | C    | M   | Mass            | Surgery                 |
| Kasamatsu A et al.     | 2007 | 71  | A    | M   | Nodule          | Spontaneous remission    |
| Bouaziz A et al.       | 2012 | 33  | C    | M   | Cystic lesion   | Spontaneous remission    |

mucosal symptom is a swelling, but both ulcers and nodules can occasionally be present. As the only way how to diagnose sarcoidosis is microscopic examination, the tissue sampling is warranted in all the cases of unclear swelling or ulcers in the mouth. Although the presence of nonnecrotizing granulomas supports the diagnosis, other diseases must be excluded before the final diagnosis is established. These include tuberculosis and other mycobacterial infections, deeper fungal infections, orofacial granulomatosis, foreign body reaction, and other granulomatous disorders such as Crohn’s disease or granulomatosis with polyangiitis (formerly Wegener’s). Plain chest X-ray and consultation with a pneumologist should be performed in every patient with granulomatous inflammation in the oral cavity.

There is no treatment consensus of sarcoidosis lesions in the oral cavity. Reported modalities comprise observation only as well as surgery, eventually combined with radiotherapy.[6-8] Surprisingly, the combined treatment comprising systemic, local, and intralesional administration of steroids has been only rarely reported in the literature.[9] Diagnosis and treatment of localized extrapulmonary sarcoidosis remain a clinical challenge. The major pitfall is the recognition of the disease and proper evaluation of all possibly affected organ systems. Oral cavity involvement is very rare. It can be seen as the first sign of the disease on very rare occasions. Oral manifestations could develop anytime during the disease. No standardized treatment has been established yet, but combining surgery and topical and/or intralesional application of steroids seem to be effective in the most cases.

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Conflicts of interest
There are no conflicts of interest.

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