Case Report

A rare ocular complication following treatment of oral submucous fibrosis with steroids

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ABSTRACT

Oral submucous fibrosis (OSMF) is one of the most commonly found pre-cancerous conditions prevalent in Southeast Asian countries. The treatment method used by most patients is the use of intralesional steroids. With intralesional steroids used commonly, one might come across an unusual ocular complication – central serous chorioretinopathy (CSCR). We report a case of a patient with OSMF who was treated by corticosteroids, subsequent to which he developed CSCR. He was put off steroidal treatment and treated conservatively to which he responded positively.

Key words: Chorioretinopathy, OSMF, steroids

Introduction

Oral submucous fibrosis (OSMF) is a chronic disease with insidious onset, which affects the oral cavity, pharynx and upper digestive tract. It was first reported by Schwartz in 1952 who identified this condition in five Indian women in Kenya and named it as "atrophia idiopathica (tropica) mucosae oris".[1] Since then, the disease has generated considerable interest, and subsequently, Pindborg, Daftary, Cox, and Aziz have significantly contributed toward the identification and treatment of this condition. With an estimated prevalence of 3.2% in Southeast Asian countries (Indian subcontinent 0.5%), it has got a malignant transformation rate of 7.6%.[2] Though various etiologic factors have been proposed ranging from autoimmunity to genetics, the factor linked directly to its causation is betel nut usage.[3] An estimated 20% of the world's population use betel nut in some form. Patients present themselves with significant masticatory dysfunction and discomfort. Advanced cases carry the risk of developing squamous cell carcinoma.

Many management strategies have been proposed for the treatment of OSMF including physical therapy, high dose antioxidants, intralesional injections with steroids and other immunomodulatory drugs along with a myriad of surgical interventions, all of which have shown limited efficacy. Intralesional steroids still remains a choice of treatment in OSMF. An unusual ocular complication was noticed while treating an OSMF patient which was thought to be worth reporting.

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Case Report

A 33-year-old male patient with average built had reported to the Department of Oral & Maxillofacial Surgery (SCB Dental College, Cuttack) with a complaint of burning sensation of mouth and decreased oral opening. He had noticed this gradual reduction in mouth opening about 4–5 years back. He was a regular chewer of paan and gutkha (an areca nut/tobacco mixture sold in a single use sachet, also referred to as pan masala). At the time of his first reporting, he had an interincisal distance of 18 mm. Clinical examination revealed that his buccal and labial mucosa were firm, pale and indurated with palpable bands. There was blanching of the mucosa. The soft palate, retromolar

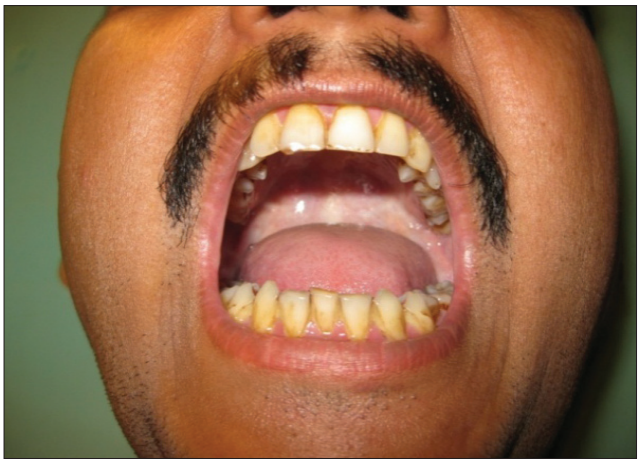

Figure 1: Adequate mouth opening post treatment

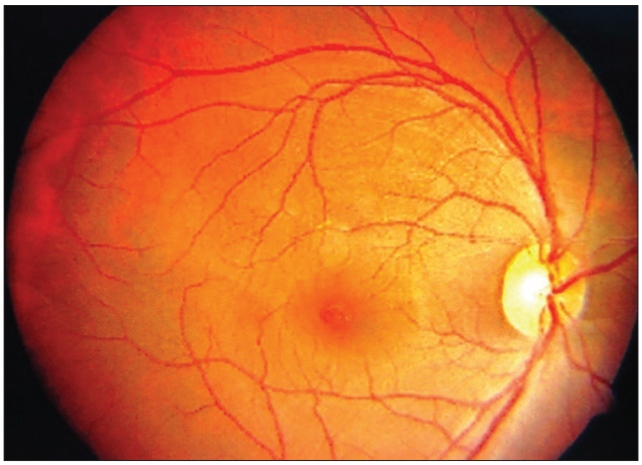

Figure 2: Color fundus photograph of the right eye showing normal macula

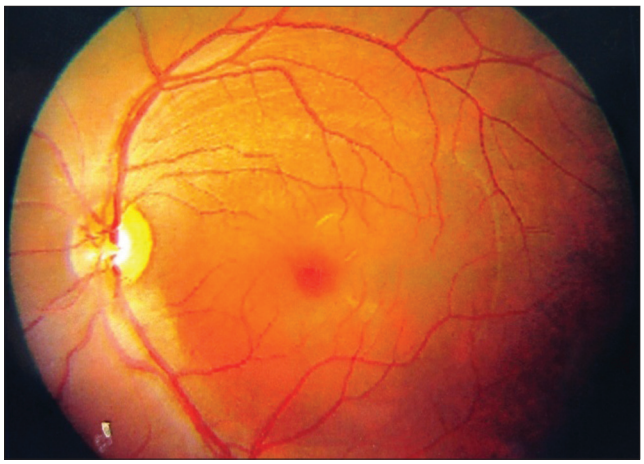

Figure 3: Color fundus photograph of the left eye showing one smooth bulging in the subretinal layer in macular region

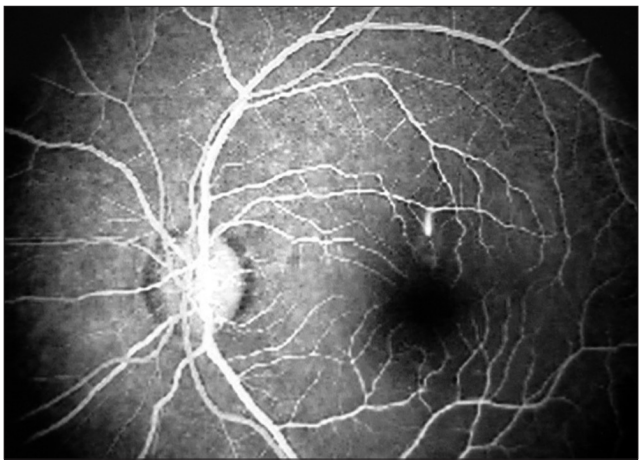

Figure 4: FFA – In AV phase, showing a small dot of dye leaking from superior parafoveal region

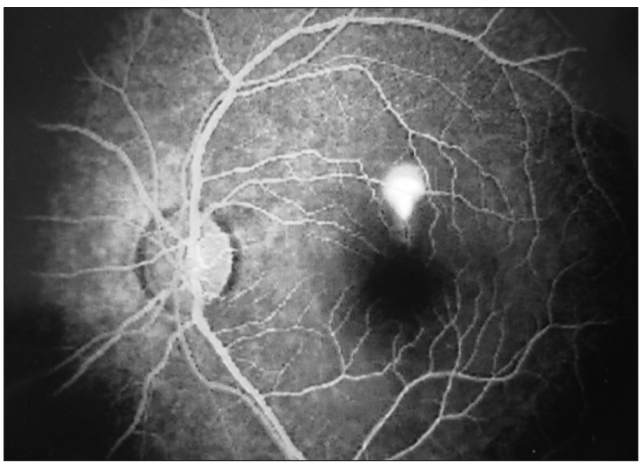

Figure 5: FFA – Late phase showing extensive dye leakage in the same zone giving a characteristic "Smokestack" pattern

area and faucial pillars were found to be involved. A diagnosis of OSMF was made and he was put on an antioxidant therapy initially. He was asked to totally abstain from chewing gutkha or any other form of tobacco.

He was treated with weekly intralesional injection of steroids [triamcinolone 1 ml (10 mg/ml) + hyaluronidase 1500 IU + placentrex 2 ml] given in fragmented doses. After a period of 2 months, the patient had noticed improvement in the burning sensation of mouth with a feeling of well-being. His interincisal opening had increased significantly [Figure 1]. But after the fifth dose of steroidal injections, he complained of heaviness and increased watering of his left eye. He noticed a blind spot in his field of vision. He was immediately referred to an ophthalmologist, who after examination diagnosed it to be central serous chorioretinopathy (CSCR) [Figure 2] Normal Eye. [Figure 3, 4 and 5], affected eye. On his consultation, the steroidal treatment was stopped as it was considered an etiologic factor for the development of CSCR. He was put under thorough observation under the care of the ophthalmologist and was asked to go for active jaw exercises, antioxidants and stoppage of habit. Follow-up examination after 6 months has revealed that his vision in the affected eye has improved though he occasionally notices blurring of the vision. Surgery was not considered as his response

to conservative treatments has shown good results. Now, his interincisal opening is 32 mm and he is still expected to gain more with active jaw exercises.

Discussion

Various modalities of treatment ranging from conservative regime to extensive surgical interventions have been tried for achieving a clinical cure but none has proved to be absolutely fruitful. Use of steroid alone ranging from local application to intralesional injections or with hyaluronidase and placentrex has been tried by many authors.

Steroid use is believed to decrease inflammation and collagen formation, thereby reducing the symptoms and resulting in increased mouth opening. Steroids have been found to produce side effects like severe adrenal insufficiency, edema, osteonecrosis, osteoporosis, myopathies, peptic ulcers, hypocalemia, euphoria, psychosis and myasthenia.

Central serous retinopathy (CSR), also known as central serous chorioretinopathy (CSCR), is a visual impairment, often temporary, occurring usually in one eye, mostly affecting males in the age group 20–50 years but may also affect women. The disorder is characterized by leakage of fluid in the central macula, which results in blurred or distorted vision (metamorphopsia). A blind or gray spot in the central vision is common, along with flashes of light (photopsia). The diagnosis usually starts with a dilated examination of the retina, followed by confirmation with Fluorescein Angiography. The angiography test usually shows one or more fluorescent spots with fluid leakage. In 10–15% of the cases, these will appear as a "classic" smoke stack shape. An Amsler grid could be useful in documenting the precise area of the visual field involved. CSCR is a fluid detachment of macula layers from their supporting tissue. This allows choroidal fluid to leak into the subretinal space. The buildup of fluid seems to occur because of small breaks in the retinal pigment epithelium. CSCR has also been associated with cortisol and corticosteroids. Persons with higher levels of cortisol than normal also have a higher propensity to suffer from CSCR. There is extensive evidence to show that corticosteroids ("cortisone"), commonly used to treat inflammations, allergies, skin conditions and even certain eye conditions, can trigger CSCR, aggravate it and cause relapses. Bouyan *et al*.[4] have reported about two patients with CSCR who had been treated with corticosteroids for their diseases. Both the patients had noticed visual loss which improved clinically following discontinuation of steroids. In another case report by Chaine *et al*,[5] a group of 14 patients had reported with serous detachment of macula due to CSCR after long-term steroidal therapy.

The prognosis for CSCR is generally excellent. Over 90% of patients regain 20/30 vision or better within 6 months. Some visual abnormalities can remain even if visual acuity is measured at 20/20. Lasting problems can include decreased night vision, color discrimination problems, and some distortion. Long-term complications can include subretinal neovascularization and pigment epitheliopathy.

There is no known effective treatment for the disease. Laser photocoagulation and transpupillary thermotherapy are some of the advocated methods.

Conclusion

Though steroid has been a choice of treatment for OSMF, it has to be used keeping its various side effects in mind. Unusual symptoms peculiar to individual patients should be noticed early and therapy discontinued if required. Other conventional methods may be used as the situation demands but the aim should be to identify and arrest the disease process at the earliest. Surgical modality is suitable in many cases if adequate post-surgical physical therapy is given to prevent recurrences.

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