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CASE REPORT

A Rare Case Report of a Delayed Sarcoid-like Granulomatous Reaction Following Religious Piercing

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Abstract

Piercings are a prevalent cultural and social phenomenon worldwide. Some of the complications of piercings include bleeding, infections, hypertrophic scars,keloid formation, granulomatous reaction, scar sarcoid etc. We report a case of delayed sarcoid-like granulomatous reaction following a religious piercing during the Thaipusam festival. There have beenfive such cases reported in literature to date.

Key Words

Granulomatous Reaction, Sarcoid-like Reaction, Thaipusam, Piercing

Introduction

Thaipusam is a Hindu festival celebrated by the Tamil community in countries like India, Sri Lanka, Malaysia, Singapore and Mauritius. It is commemorated with offerings done to Lord Murugan by bearing decorated structures known as kaavadi and piercings in the cheeks, tongue, back and chest with metal spears known as alagu. Complications of body piercings include bleeding, infection, allergic reaction, scarring, granulomatous reaction, scar sarcoid, etc.[1] A delayed sarcoid-like granulomatous reaction following a piercing is quite an unusual phenomenon and in such scenarios, it is important to rule out diseases like sarcoidosis, leprosy and tuberculosis.

Case Report

A 31-year-old female was referred to our OPD in view of complaints of multiple painful swellings in the oral cavity and lips for 2 months. The lesions caused painful mouth opening and chewing. She did not givea history of any other mucosalor skin lesions. There was nohistory of fever, loss of weight or loss of appetite. On further inquiry, she revealed that she had performed the religious oral piercing called 'Alagu kuthudhal' 5 years ago. She had no history of comorbidities. On examination of the oral cavity, threeskin-colourednodules of sizes 1x2 cm, 2x2 cmand 2x3 cm were noted over the left and right sides of the buccal mucosa and over the right angle of the mouth, respectively. (Fig 1) The nodules were well-defined, firm in consistency and tender on palpation. The submandibular lymph nodes were bilaterally tender and palpable. Clinical differential diagnoses of scar sarcoid, delayed granulomatous reaction, oral tuberculosis, deep fungal infection and histoid leprosy were made. Blood investigations showed an ESR of 34 mm/hr. Serological tests like HIV, HBsAg, anti-HCVand VDRL testswere non-reactive. A chest X-ray and HRCT of the thorax showed reports within normal limits. Histopathological examination of a punch biopsy specimen taken from a nodular lesion revealed multiple granulomas composed of epithelioid cells, a few Langhans giant cells and lymphocytes with no evidence of necrosis or atypia. (Fig 2a) Reticulin staining revealed a scanty reticulin network. (Fig 2b) No mycobacteria were visualised with Ziehl-Neelsen and Fite-Faraco staining. Periodic acid Schiff and Gomori's methenamine silver staining were also negative for fungal elements. The Mantoux skin test

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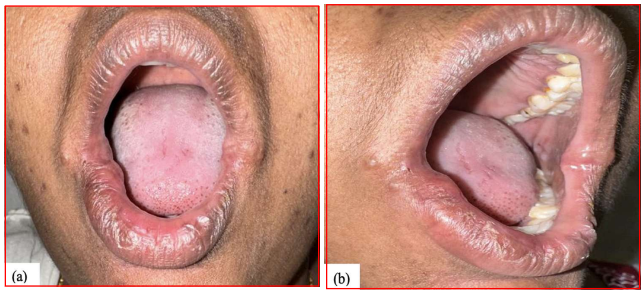

Figure 1: (a) and (b) showing nodular lesions involving the oral cavity and lips.

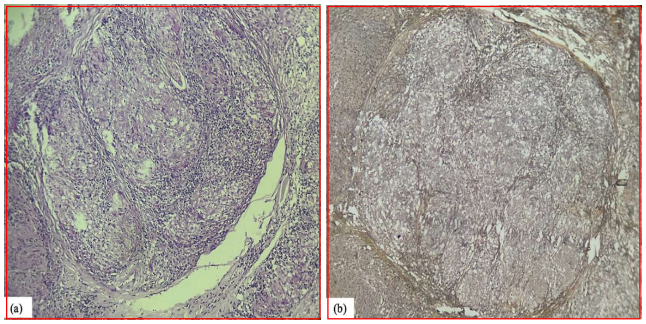

Figure 2: (a) granuloma composed ofa collection of epithelioid cells, Langhans-type giant cells and lymphocytes; (b) Reticulin staining showing scanty reticulin network.

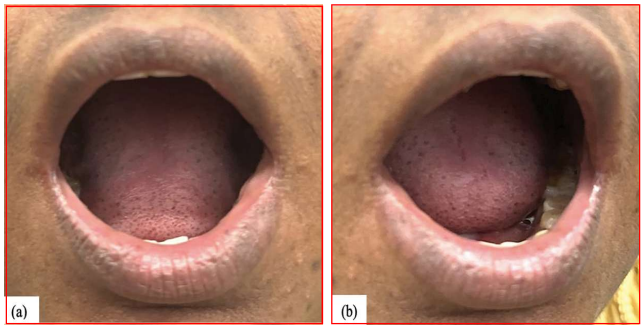

Figure 3: (a) and (b) showing resolution of nodular lesions post-treatment.

showed a reading of 17 mm, while a CB-NAAT analysis of the tissue specimen was negative for mycobacteria. Serum angiotensin-converting enzyme (ACE) and serum calcium levels were normal. Based on the findings, a diagnosis ofa delayed sarcoid-like granulomatous reaction was made. The patient was treated with four sittings of 10 mg/ml intralesional triamcinolone acetonide injection one monthapart. Asthe patient complained of continued pain during mastication and hadan unsatisfactory response to one sitting of intralesional steroid injection alone, she was additionally treated with weekly intramuscular injections of 12.5 mg methotrexate and 200 mg oral hydroxychloroquine daily for 3 months. There was a progressive reduction in the size of the lesions in 6 weeks and a complete resolution of lesions in 4 months of treatment initiation. (Fig 3) The patient was followed up for a period of 1 year after the treatment period and she did not report recurrence or the appearance of any new lesions.

Discussion

Granulomas are a common inflammatory entity observed in dermatopathology. A sarcoidal granuloma could be differentiated from a tuberculoid granuloma by the presence of a less prominent inflammatory infiltrate and by the absence of caseous necrosis.[2] A sarcoid-like granulomatous reaction is considered inpatients who do not satisfy the criteria for systemic sarcoidosis.[3] It is a type IV hypersensitivity reaction that is commonly triggered by malignancies and exogenous agents like body piercings and tattoos. Antigen-presenting cells (APCs) present these antigens via MHC class II molecules to the CD4+ Th cells, which in turnsecretecytokines like IL-2, IFN-λ and TNF-α that cause recruitment of macrophages and ultimately lead to granuloma formation. The granulomatous reaction following the oral piercing done during Thaipusam is a delayed-type hypersensitivity response,probably attributed to the metal shards of thespear or to the holy ash known as the'vibuthi' that is pressed over the raw areas as soon as the spear is removed in an attempt to arrest the bleeding. The ash is commonly made of a combination of burnt cow dung and neem tree wood blended with sandalwood and rose essences.[4] There are a limited number of reports of granulomatous reactions following such religious oral piercings.[4-6] After excluding the other possible aetiologies, these cases have been managed with either intralesional or systemic corticosteroids. Our patient was treated with a lower dose of intralesional triamcinolone acetonide injections and a short course of systemic immunosuppressants like methotrexate and antimalarials like hydroxychloroquine,with which she hada good response. This emphasises the role of steroid-sparing agents in patients who do not respond adequately with steroids alone and also the safety profile of steroid-sparing agents when given for a shorter duration.

The other therapeutic approaches that have been tried for granulomatous reactions include tetracyclines and

allopurinol. Excision can be considered when the patient does not respond to medical management. In addition, as a preventive measure, patients should also be advised to avoid any trauma to the skin in the form of tattooing or piercings in the future.

Conclusion

Sarcoid-like granulomatous reaction following a piercing isa rare entity that presents as a lump a few months or years after trauma. It could affect the patient functionally and cosmetically. While it is challengingto rule out other close clinical differential diagnoses like tuberculosis and scar sarcoidosis, local and systemic immunosuppressants with adjunctives arean adequate therapy to improve the condition.

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