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Case Report

Concomitant squamous cell carcinoma and actinomycosis—An unique and rare occurrence

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ABSTRACT

Introduction: Actinomycosis of the oral cavity is not common, though actinomyces species often occur as normal commensals in the oral cavity. They gain entry into deep tissue to cause infection and consequent tissue damage whenever there is damage to the oral mucosa by injury or infection or neoplastic process.
Case Report: The case being discussed here is of a male, aged 62 years, who presented with an ulcero-proliferative lesion over left buccal mucosa. A punch biopsy was carried out on the lesion. Histopathology laboratory received three gray-white tissue pieces. Histological examination of the tissue pieces revealed features of moderately to poorly differentiated squamous cell carcinoma. Few colonies of radiating filamentous structures reminiscent of colonies of actinomyces species were noted. The sections were further stained with relevant stains for filamentous bacteria such as Periodic Acid Schiff (PAS) stain which confirmed the colonies of Actinomyces spp.
Conclusion: The case being submitted here is an unique and relatively rare finding of concomitant squamous cell carcinoma and oral actinomycosis with only few cases being reported in publish English medical literature.

1. Introduction

Actinomycosis is a rare chronic disease caused by Actinomyces species, which usually colonize human oral cavity, digestive and urogenital tracts.[1,2] The Office of Rare Diseases at the National Institute of Health has also listed this disease as a "rare disease ".[3] Actinomyces species are known to be a saprophytic constituent of flora of oral cavity which cause infection when they gain entry through breach in the mucosa caused by infection or injury as in case of dental extraction. The Actinomyces species are facultative anaerobic or microaerophilic, Gram-positive filamentous bacteria.[3–7] Actinomycosis affects various organ systems of the human body, and based on this actinomycosis has been classified broadly into three types: 1. Cervico-facial; 2. Thoracic; 3. Abdomino-pelvic.[3,4] Actinomyces are considered among opportunistic micro-organisms. Infections occur endogenously and are known to occur more commonly in males with ratio of infections in males and females being 3:1, Infections are known to affect people in the age-group of 20–50 years. The bacteria enter the body through disruption and breach in the mucous membrane or the skin, most commonly during tooth extractions, abdominal or pelvic surgery or in patients with prolonged history of having intrauterine contraceptive device (IUD), which may also carry the risk of infections while inserting it. In locations with favourable growth conditions, particularly anaerobic conditions, Actinomyces cause abscesses that may lead to formation of sinuses or fistulas. In humans, for actinomycosis to occur Actinomyces

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israeli, Actinomyces propionica and Actinomyces naeslundi are most commonly responsible although 39 or more species of Actinomyces have been described. The characteristic feature of actinomycotic inflammation is the presence of "actinomycosis grains", sulfur granules or sulfur-like yellowish colonies of Actinomyces which form characteristic 0.1–5 mm granules usually found in purulent inflammatory exudate that drains often through sinuses.[6]

Relatively detailed appraisal of medical literature showed published reports of actinomycosis of oral cavity, digestive and urogenital tracts, many of which claim that actinomycotic lesions mimicked malignancies arising in those sites. It is also reported that oral actinomycosis deceptively present as neoplastic processes, which raise serious clinical diagnostic challenges, but existing literature is conspicuously bereft of any veritable report of co-existence of oral actinomycosis and oral squamous cell carcinoma.[2–12] Hence, the present case report is unique in that it could turn to be a very rare and first of its kind that conclusively establishes co-existence of oral actinomycosis and invasive oral squamous cell carcinoma.

2. Case Report

The case being presented here is of 62-year-old man who presented with clinical history having an ulcero-proliferative lesion over the left buccal mucosa which had infiltrated the entire buccal mucosa, the underlying connective tissue stroma and skeletal muscle to penetrate the overlying skin of the left cheek. A clinical suspicion of oral malignancy led to further evaluation of the condition by relevant clinical and imaging studies. Routine laboratory investigations and imaging studies were clinically unremarkable. Finally, a punch biopsy of the lesion was carried out and the specimen was sent for histopathological appraisal.

2.1. Gross examination

Three irregular gray white and slightly friable tissue fragments were received; the largest tissue fragment measured 2.8 cm and smallest measured 2 cm in their maximum dimensions. The tissue sections submitted were routinely processed and stained with Haematoxylin and Eosin stains.

2.2. Microscopic examination

Sections stained with Haematoxylin and Eosin revealed mucosal epithelium lined by hyperkeratotic stratified squamous epithelium with extensive areas of denudation. The subjacent scant stroma showed irregular nests and sheets of round to ovoid cells with ample pale eosinophilic cytoplasm and moderately to markedly pleomorphic vesicular nuclei displaying prominent nucleolus. Focal areas of necrosis and dense infiltration by mixed inflammatory

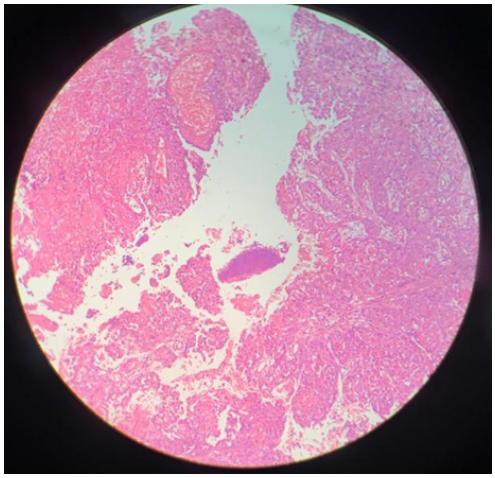

Fig. 1: (10x, Hand E) Photomicrograph of concomitant invasive squamous cell carcinoma with colonies of Actinomycosis species.

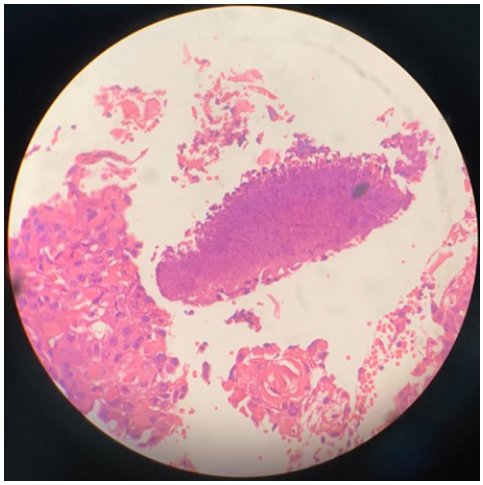

Fig. 2: (40x,H and E) photomicrograph of Invasive squamous cell carcinoma with concomitant colonies of Actinomycosis species.

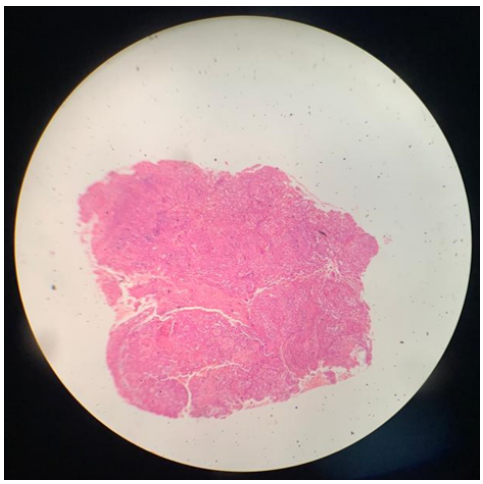

Fig. 3: (4x H and E) Photomicrograph of invasive Squamous cell carcinoma with concomitant colonies of Actinomycosis species.

cells were also noted. At places, Occasional basophilic clumps composed of delicate filamentous structures surrounded by eosinophilic material were also noted, which led to the diagnosis of squamous cell carcinoma with associated features suggestive of orofacial actinomycosis.

3. Discussion

Oral actinomycosis is known to be very rare and very few cases of oral actinomycosis[3,5,8,9] are reported in exhaustively reviewed literature, though actinomyces species occur as normal oral commensals. Some of the reported cases of actinomycosis mimicked malignancy and posed contentious diagnostic dilemmas.[3,6,10–12] Thukral R. et al[3] published a case of actinomycosis of left jaw, which mimicked post-dental extraction osteomyelitis opportunistic bacterial flora since the presenting features were unusual without classic features of abscess, sinus formation and woody fibrosis. Similarly, Kaszuba M, et al[6] described a case of actinomycosis which mimicked abdomino-pelvic malignancy. Yannick Deswysen et al[10] and Baig SN[11] reported actinomycotic lesions occurring in oesophagus which were misleading and mimicking oesophageal cancer. In the same way, Varghese, B.T.[12] published a case report of actinomycosis of parotid gland masquerading as salivary gland neoplasm. On the other hand, Grey T. Lindsay et al[7] reported a case of pre-existing rectal carcinoma becoming unresectable due to complications caused by superadded actinomycotic infection. Lim JA et al[9] published a case report of actinomycosis of the palate that had led to masking of co-existing B-cell lymphoma of the palate and adrenals. All these cases of actinomycosis, indicate that, despite actinomycotic infections being very rare, actinomycosis can affect not only various organ systems but also create deceptive diagnostic predicaments. Paradoxically, these veritably described cases of actinomycosis are conspicuously bereft of any reports of concomitant or co-existent oral actinomycosis and invasive oral cancer. In this regard the case being presented here of co-existent oral actinomycosis and invasive squamous cell carcinoma of the oral cavity is very distinctly unique and one of the extremely rare cases to be reported so far in medical literature.

4. Conclusion

Comprehensive reappraisal of literature of various clinical reports on actinomycosis establishes the fact that oral actinomycosis indeed is very rare, though some of the species of Actinomyces are known to colonize the oral cavity, and furthermore concomitant or co-existence of oral actinomycosis and invasive oral squamous cell carcinoma is extremely uncommon to be considered as incipient. The distinctiveness of the case being discussed here is that it did not conform to the characteristic presenting as the primary lesion manifested as ulceroproliferative lesion with neither abscess nor sinus formation. Meticulous histopathological appraisal of the lesion, supplemented by ancillary studies such as special histological stains to demonstrate microbial organisms and bacterial studies, is considered gold standard in diagnosis of these lesions which have potential risk of either of the component lesions masking or mimicking the other component lesion posing diagnostic incertitude.

5. Source of Funding

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6. Conflict of Interest

None Declared.

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