

Caliber persistent artery in the palate of an Egyptian patient with type 2 diabetes mellitus: a rare case report

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Key words

Caliber-persistent artery • Oral manifestation • Anterior palate

Summary

This paper reports a very rare case of isolated ulcerative lesion of the palate in a patient with type 2 diabetes mellitus. The patient refused to run any radiological tests, other than the periapical radiograph. This rendered the diagnostic dilemma more challeng-

ing. Through histochemical and immunohistochemical stains, the case could be non-invasively diagnosed as caliber-persistent artery. All differential diagnoses were excluded based on concrete findings.

Introduction

Diabetes mellitus encompasses a group of genetically and clinically heterogeneous metabolic disorders characterized by hyperglycemia depletion of antioxidants, and alteration in lipid metabolism. Diabetes mellitus (Type 2) characterizes an insulin resistance which progresses into an insulin deficiency due to a secondary failure in the pancreatic beta-cells. Given the pathophysiology of diabetes-related complications, diabetic patients develop a relatively higher vulnerability of the mucosa upon exposure to various ulcerogens, ischemia or reperfusion, stress, and nonsteroidal anti-inflammatory drugs¹⁻⁴. A wide diversity of inflammatory - either fungal, viral or bacterial- and neoplastic palatal lesions may overlap complicating the diagnosis of unusual representations. Accordingly, meticulous care should be rendered to avoid unnecessary procedures and improper treatments, especially in chronic diabetic cases.

Case history

A 66-year-old female, non-smoker and non-alcoholic, has suffered a painful palatal lesion. The ulcerative palatal lesion had an insidious onset, but turned painful for

two weeks without any response to placebo (Antibiotics, NSAID, anti-fungal and topical gels) (Fig. 1a). An incisional biopsy was cut for microscopic examination. No odontogenic irritants could be blamed. History of any concomitant tumor was negative. No running diseases in the family except for non-insulin dependent diabetes mellitus. The differential diagnosis included caliber-persistent artery with or without vasculitis, mucormycosis, lymphoma, granulomatosis polyangiitis (aka Wegener's granulomatosis), and spindle cell carcinoma.

Serum ANA and C-reactive protein tests were negative. Detection of antineutrophil cytoplasmic antibodies (ANCA) from indirect immunofluorescence on blood was also negative ruling out granulomatosis polyangiitis. The periapical radiograph showed no more than normal maxillary trabeculae, excluding the liability of lymphocytic malignancies (Fig. 2).

Histologically, the structure of this peripheral palatal lesion revealed an arterial blood vessel wall, with dense collagen with two layers of spindle cells running in different directions, surrounded by inflammatory infiltrates (Fig. 3). Because of the surface ulceration and acute inflammation, the ulceration was expected to be either a sign for vasculitis or inflammatory evidence. Neither granuloma formation nor malignant features could be detected (Fig. 4).

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Fig. 1. Clinical picture displaying exophetic ulcerative lesion, located at the anterior palate.

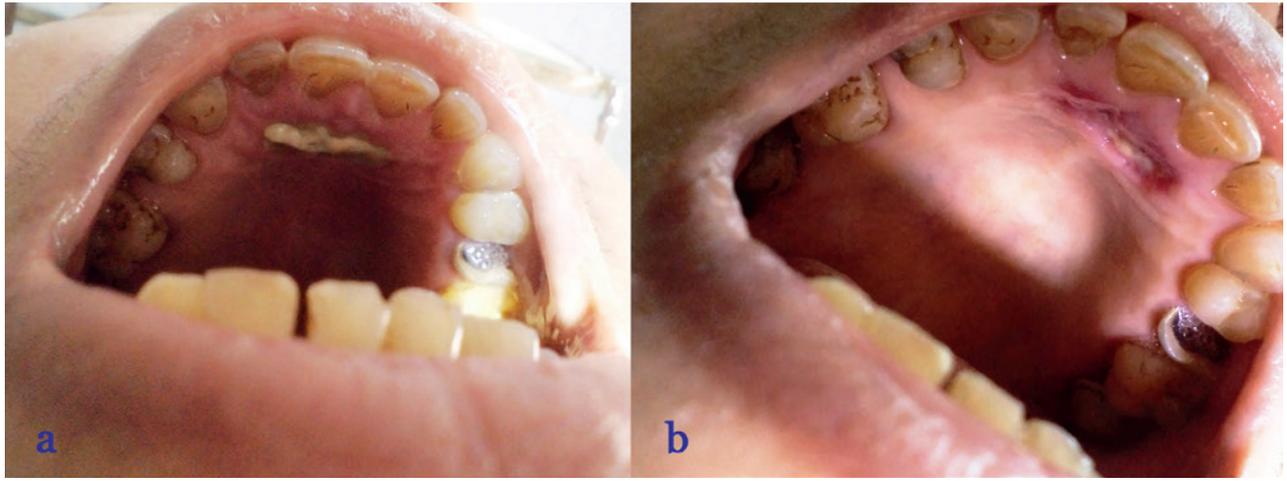


Fig. 2. X-ray showing normal maxillary trabeculae. Neither dental caries nor any radicular lesion is evident.



Fig. 3. Photomicrograph revealing dense collagen of two layers of spindle cells running in different directions, surrounded by inflammation (H&E stained, Original magnification:10x).

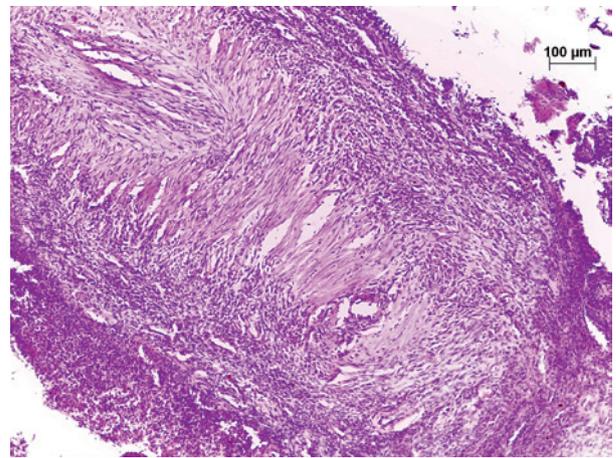


Fig. 4. Photomicrograph of higher magnification characterizing the inflammatory infiltrates around the arterial wall. (H&E stained, Original magnification: 40x).

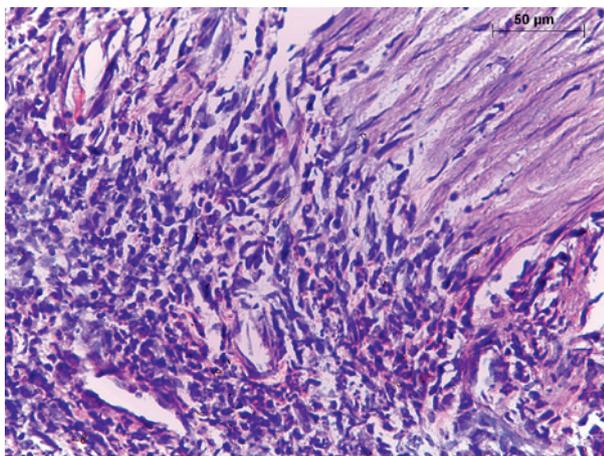
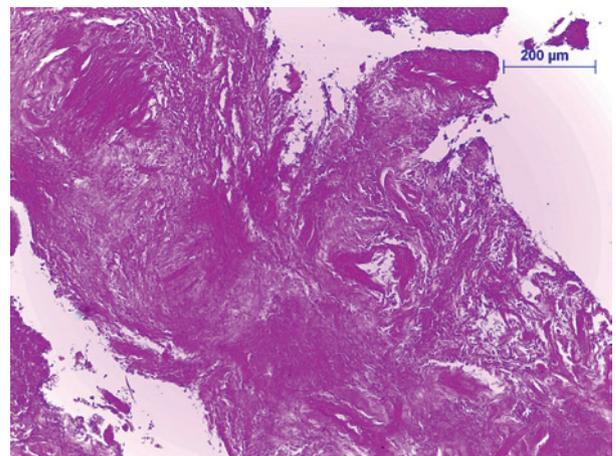


Fig. 5. Photomicrograph displaying no more than inflammatory infiltrates around an arterial wall. No huphae could be traced (PAS stained, Original magnification: 4x).



To exclude any fungal involvement, PAS stain was used to identify in hyphae, if present. No fungal traces could be seen (Fig. 5). Immunohistologically, performing a smooth-muscle actin stain could confirm the arterial component in the lesion. The lesion was strongly positive for smooth-muscle actin. Other tests were run to exclude any malignancy, including S-100, CK7, CK18, and p53, which were all negative. In 10 days, the palatal site healed up dramatically confirming the inflammatory nature of this lesion.

Discussion

As regards chronic diabetic complications, bone marrow-derived hematopoietic cells were recently considered proinflammatory and infiltrative to tissues inducing microvascular complications in addition to the dysregulated biochemical pathways downstream of hyperglycemia⁵. Miko et al first described, in 1980, a developmental anomaly referred to by them as persistent caliber artery. The artery becomes superficial toward the midline, and the persistent size makes it palpable, usually a few millimeters inferior to the vermilion border. Being idiopathic in nature and rare in incidence, predisposing factors included intense sun exposure, senile atrophy of tissue associated with vascular ectasia, secondary to ageing, and injury from long term pipe smoking⁶⁻⁷. Although caliber-persistent artery (CPA) is not totally delineated in the medical literature, given the rarity of the reported cases, this case showed typical findings of CPA. There appears a dysregulated hematopoietic element induced a low-grade irritation causing the arterial wall to thicken, which made the site liable to ulceration in the harsh oral environment. Either idiopathically induced or pathophysiologically caused by the underlying diabetic condition, the poorly vascularized anterior palate might contribute to the severity of the condition. Although CPA is a disease commonly seen in geriatric individuals, Awni and Conn⁸ have interestingly reported this rare entity in two young cases out of five cases. Santagata et al.⁹ have remarkably reported CPA in the upper lip of a 25-year-old female. In our case, other possibilities of malignancies, infection, or autoimmunity were definitely ruled out.

For the deep fungal infections which are not uncommon in diabetic patients, mucormycosis was frequently seen devastating the palate and encountering the nasal cavity. Mucormycosis is ubiquitous in nature and humans usually have a strong natural resistance to the infection. Mucormycosis becomes pathogenic when the patient's general resistance has been altered by metabolic disorders, immunosuppressive therapy, malignancy or other chronic debilitating disorders. An underlying disease, frequently diabetes mellitus, is always evident⁹⁻¹¹. In this case, the recruitment of PAS stain, with its negative detection of hyphae, signed any deep fungal infection out.

Granulomatosis polyangiitis is an uncommon immune-based inflammatory necrotizing vasculitis with no certain etiology proven. Upper respiratory signs and symptoms include ear epistaxis, sinonasal dryness, crusts, obstruction, sinonasal destruction, subglottic stenosis, hearing loss, tinnitus and vertigo. Lower respiratory manifestations comprise parenchymal nodules, endobronchial lesions, pulmonary infiltrates, and pulmonary hemorrhage or embolism. Serologically, 90% of patients of granulomatosis polyangiitis are ANCA positive¹². Nothing of the above mentioned signs and symptoms was evident in our case. Malignancies were histologically and immunohistochemically excluded.

Conclusions

A better understanding of the versatile pathophysiology of diabetes-related complications is mandatory. The diagnostic procedures, especially in diabetic patients, should be careful and non-invasive. This should combine consultative efforts between medics and paramedics toward providing the best diabetic care for the patients.

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