A Pigmented Bullous Maxillofacial Lesion in a Diabetic Patient: A Diagnostic Dilemma

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INTRODUCTION

Diabetes mellitus (DM), the most common endocrine disorder currently affecting 246 million people worldwide and is expected to affect a staggering 380 million by 2025.1 90% have Type II, non-insulin-dependent (NIDDM), whereas 10% have insulin-dependent Type I (IDDM). Type II patients develop more frequent cutaneous infections, whereas Type I patients develop more auto-immune type cutaneous lesions.2,3 Numerous skin lesions are associated with either Type I or Type II DM. DM is characterized by high serum glucose levels and by disturbances of carbohydrate and lipid metabolism and resultant long-term systemic complications. DM has replaced syphilis of pre-antibiotic days as the great clinical imitator with a wide array of sign and symptoms affecting every organ of the body. The incidence of cutaneous or dermatological lesions in diabetic patients varies from 30% to 71% according to different authors.4,5

The cutaneous lesions of DM are numerous, complex and varied. Moreover, sometimes it becomes difficult to classify them. However, the lesion that we found in our patient does not fit in the classification of dermatological lesions of DM. We present a rare case report of a pigmented bullous maxillofacial lesion on right side of neck, which was associated with reactive lymphadenitis due to infection from carious right mandibular the second molar.

CASE REPORT

A 77-year-old female reported to the Department of Oral and Maxillofacial surgery with a complaint of non-healing wound on right side of neck. She had swelling on the right side of the neck past last 8 months (Figures 1 and 2). She developed a slow growing painless swelling over right submandibular area for which she underwent Fine-needle aspiration cytology (FNAC) and was diagnosed as a case of submandibular lymphadenitis. An excisional biopsy was planned under general anesthesia, but could not be operated due to other associated comorbidities. After 1 month, she developed pigmented bullous lesion at the site of FNAC.

The patient was operated for cataract 5 years back and had DM (Type II) and Hypertension since 20 years. She had a history of cardiac arrest and convulsions 8 months back, from which she recovered well and was put on antiplatelet, vasopressors, and anticonvulsant drugs. She was taking following drugs:
Tablet aspirin 150 mg, tablet atorvastatin 40 mg, tablet levetiracetam 250 mg, tablet metoprolol 25 mg, tablet furosemide 40 mg once daily, and tablet gliclazide-XR 500 mg twice daily.

On general physical examination, she was well-oriented. Parameters were within normal range and she had pallor. There was generalized petechiae presentation on right arm, dorsal surface of left hand, nape of neck, right lateral brow, and left side of cheek (Figure 3a-e).

Maxillofacial Examination
Irregular skin lesion approximately 5 cm × 3 cm extending superoinferiorly from right lower border of mandible to anterior border of sternocleidomastoid muscle and anteroposteriorly from level of hyoid bone to 1.5 cm inferior to lobule of ear was present on right side of the neck. The edge of the lesion was sloping, clearly defined with bullous formation at the postero-inferior margin (Figure 4). The lesion was crusted dark brown, and the surrounding area was wrinkling red.

On palpation, the lesion was slightly tender, inflamed and warm to touch with indurated base having the varying depth of 2-4 mm in different areas. Oral cavity examination revealed poor oral hygiene and orthopantomogram (OPG) showed carious right lower mandibular the second molar (Figure 5).

Differential Diagnosis
Diabetic lesion was thought to be the likely diagnosis because of raised blood sugar levels and history of trauma. The other differential diagnosis was lupus vulgaris, scrofuloderma, and necrotizing fasciitis.

Bleeding disorder was also considered because of generalized presentation of petechiae.

Investigations
Laboratory tests facilitated the diagnosis of diabetic lesion.
Hemoglobin A1c was elevated at 8.6%, Blood urea was raised at 76 mg/dl, serum creatinine levels were at 2.7 mg/dl. A Mantoux purified protein derivative was placed which was subsequently negative. Polymerase chain reaction for mycobacterium tuberculosis was not detected. Hence, the lesion was not lupus vulgaris/scrofuloderma.

Necrotizing fasciitis was ruled out because the lesion was not spreading across the fascial planes, and there was no necrosis of the fascia and subcutaneous tissue.

Erythrocyte sedimentation rate was raised at 70 mm. Hemoglobin was 8.1 g% and red blood cells (RBC’s) were normocytic normochromic. Most normocytic normochromic anemia appears to be the result of impaired production of RBC. In renal failure, there is a deficiency of erythropoietin (EPO). The life span of RBC is shortened, but the cause of anemia is failure of red blood cell production.

Serum iron study was done and the levels of serum iron, total iron binding capacity and percentage of iron saturation were within normal limits (Table 1).

To rule out the presence of any bleeding disorder, coagulation tests were done (Table 2). Plasma Von Willebrands factor (vWF) and Factor VIII were raised. Activated partial thromboplastin time (APTT) was prolonged. The presence of prolonged clotting times triggered functional as well as serological testing of blood clotting function to identify common autoantibodies such as antiphospholipid antibodies or lupus antibody/lupus anticoagulant (LAC). These antibodies tend to delay in vitro coagulation in phospholipid dependent laboratory tests such as APTT. Patients LAC profile was done and the values were within normal range (Table 3).

**Management**

The patient was commenced on oral doxycycline 100 mg BD. Under antibiotic coverage extraction of lower right second mandibular tooth was done and occlusive dressing with beta dine was given every alternate day.

Human insulin was used for blood glucose control (6 units in the morning and 4 units in the evening). Moreover, after consultation with the neuro physician levetiracetam (anticonvulsant drug) was slowly tapered and discontinued altogether.

The patient had an uneventful postoperative course, and the ulcer healed and the petechiae resolved spontaneously (Figures 6a-d).

**DISCUSSION**

Diabetic lesions are classified into four categories: (I) Skin diseases with strong association and others with less

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**Table 1: Serum iron study**

<table>
<thead>
<tr>
<th>Test</th>
<th>Patient</th>
<th>Normal values</th>
</tr>
</thead>
<tbody>
<tr>
<td>Serum iron (µg/dl)</td>
<td>56</td>
<td>50-177</td>
</tr>
<tr>
<td>Total iron binding capacity (TIBC) (µg/dl)</td>
<td>386</td>
<td>250-430</td>
</tr>
<tr>
<td>% age of iron saturation</td>
<td>14.5</td>
<td>15-50</td>
</tr>
</tbody>
</table>

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**Table 2: Coagulation tests**

<table>
<thead>
<tr>
<th>Test for coagulation</th>
<th>Patient</th>
<th>Normal values</th>
</tr>
</thead>
<tbody>
<tr>
<td>vWF (%)</td>
<td>227</td>
<td>89</td>
</tr>
<tr>
<td>Factor VIII (%)</td>
<td>263</td>
<td>84</td>
</tr>
<tr>
<td>Factor IX (%)</td>
<td>58</td>
<td>85</td>
</tr>
<tr>
<td>Prothrombin time</td>
<td>14.4</td>
<td>11-16 s</td>
</tr>
<tr>
<td>APTT</td>
<td>45.1</td>
<td>30-40 s</td>
</tr>
<tr>
<td>INR</td>
<td>1.19</td>
<td></td>
</tr>
</tbody>
</table>

INR: International normalized ratio, APTT: Activated partial thromboplastin time, vWF: Von Willebrands factor

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**Table 3: LAC profile**

<table>
<thead>
<tr>
<th>LAC profile</th>
<th>Patient (s)</th>
<th>Normal values (s)</th>
</tr>
</thead>
<tbody>
<tr>
<td>PTT (LAC)</td>
<td>46.4</td>
<td>38.8-46.6</td>
</tr>
<tr>
<td>DRVV screen</td>
<td>40.3</td>
<td>39.3-43.1</td>
</tr>
</tbody>
</table>

PTT: Partial thromboplastin time, LAC: Lupus anticoagulant, DRVV: Dilute Russell’s viper venom
distinct association with DM, (2) Cutaneous infections, (3) Dermatologic disorders related to diabetic complications, and (4) Skin conditions related to diabetic treatment. The lesion seen in our patient does not fit into the classification of dermatological lesions of DM.

Diabetic lesion in the maxillofacial region is a rare presentation. In our patient, the infected right mandibular second molar tooth lead to submandibular lymphadenitis and raised blood sugar levels and trauma following FNAC were the triggering factors for the formation of the lesion.

With the extraction of the offended tooth and dressings, the lesion healed completely (Figure 6a). This case report warrants recognition as clinicians may encounter such cases in clinical practice. As the source of infection was a carious tooth, the importance of oral hygiene cannot be neglected in a diabetic patient. Although raised blood sugar levels and trauma were the contributing factors.

Factors leading to initiation and those determining rate of progression of diabetic renal disease are gaining increasing recognition. Both development and early progression of diabetic nephropathy (DN) are the most likely to occur following years of poor diabetic control. Type II diabetic patients with DN, but without severe renal function impairment are anemic (normochromic and normocytic) and is associated with a relative EPO deficiency. Strict control over a decade both delays onset of the disease and slows progression chiefly of its early phase. Normocytic Anemia often occurs relatively early in the course of the disease before renal failure is established as seen in our patient with normal serum ferritin levels thus excluding iron deficiency.

The mechanisms that may contribute to this anemia include shortened red cell survival, decreased EPO production, blood loss because of defective platelet function, and impaired erythropoiesis secondary to inhibitors or toxic metabolites. As the renal function deteriorates, the anemia becomes more marked. The anemia characteristically responds to exogenous EPO administration in the form of recombinant human EPO in the presence of adequate iron replacement.

Plasma vWF, factor VIII (a marker of endothelial damage/dysfunction) levels are elevated in patients with Type II DM, particularly in the presence of microalbuminuria and a history of coronary artery disease. Hyperlipidemia is common in nephropathy, as are other risk factors for vascular disease, including changes in the concentrations of fibrinogen and other clotting factors. The raised clotting factors in the patient was a long-term complication of diabetes. Generalized presentation of petechiae was due to fragility of the blood capillaries, due to long standing DM.

Though LAC was suspected in the initial stages. Lupus antibody is an immunoglobulin that binds to phospholipids and proteins. In living systems causes an increase in APTT and prevents blood clotting.

**CONCLUSION**

Therefore in the present clinical report, the final diagnosis was diabetic lesion, which was pigmented bullous in nature and was difficult to diagnose, as the lesion resembled tuberculous ulcer. Clinically, as well as radio graphically, the cause of infection was diagnosed as abscess secondary to carious right lower second molar tooth.

Long-standing uncontrolled diabetes and trauma following FNAC lead to the formation of the lesion. Diabetics should maintain good oral hygiene, keep a record of their blood glucose levels and maintain strict glucose control and avoid injury.

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