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101 Interesting Cases in Clinical Medicine

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CASE 4

MIDDLE-AGED GENTLEMAN WITH ENLARGEMENT OF TONGUE

CASE SUMMARY

A 54-year-old male presented with gradual swelling and enlargement of tongue for the last 1 year. He complained of repeated accidental tongue bites and multiple small oral ulcerations all over the tongue, which were painful. The pain was so intense that he developed swallowing and speech difficulty. His dysphagia started with solid food, then it progressed to interfere with intake of liquids too and subsequently he lost approximately 15 kg weight because of this condition. He had long-standing bilateral knee joint pain and walking difficulty. Sometimes, both knees became so painful that he needed to use walking aid. He also developed generalized body ache during this course of illness. On his past medical history, he had vitiligo for the last 24 years. He was a diagnosed case of ischemic heart disease and had a history of stenting with percutaneous coronary intervention to left anterior descending and right coronary arteries about 3 years back before his current symptoms arose but during this 1 year course of illness, he noticed gradual deterioration in his general health with generalized fatigue, bilateral leg swelling and significant exertional dyspnea. On query, he mentioned having occasional passage of loose stool without any blood. Along with all these complaints, he was unable to perform his daily activities. So, he got admitted to our hospital.

On examination, patient was ill-looking and mildly anemic. Bilateral pitting leg edema was present. Vitiligo was present all over his body and around lips. Vital signs were within normal limit. There was generalized enlargement of the tongue with indentation on the lateral border due to pressure from tooth. There was no fasciculation or tongue deviation. The tongue was red on dorsal and ventral surfaces. Dorsal surface was rough; multiple painful small, nodular and plaque-like ulcers were present all over the tongue (Fig. 1). Angular stomatitis was absent. There was no tooth spacing. Shape and

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Fig. 1: Dorsal surface of the tongue showing enlargement, redness and multiple small, nodular and plaque-like ulcers.

movement of the chest were normal and symmetric and trachea was central. Breath sound was vesicular with few bilateral basal crackles. Nervous system examination was normal.

Investigations showed Hb—10.3 g/dL; peripheral blood film (PBF)—normocytic normochromic anemia; liver function tests—normal; renal function tests—normal, except urine routine examination revealed 1 + protein; S. electrolytes and S. calcium level—normal; thyroid function test—normal; C-reactive protein, antinuclear antibody, rheumatoid factor test, anti-cyclic citrullinated peptide—negative; X-ray of knee joint—osteoarthritis; X-ray of skull—normal; echocardiography—septal, anterior and lateral wall hypokinesia (OMI); moderate left ventricular systolic dysfunction. Borderline concentric left ventricular hypertrophy; ejection fraction was 38%. Ultrasonography (USG) of W/A—normal. USG of tongue—macroglossia, where echotexture was homogeneous and multiple tiny hypoechoic nodules were seen in the musculature. Urine for Bence Jones protein was positive. Protein electrophoresis was done on two episodes. On first report, there was significant raised alpha-2, and on both reports no monoclonal band was seen; bone marrow study—6–7% of plasma cells.

QUESTIONS

1. What are the causes of gradual swelling of tongue?
2. What are the possibilities in this case?
3. What should be the next plan of investigations?

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DISCUSSION

Most common causes of acquired macroglossia are: acromegaly, hypothyroidism and amyloidosis.

Clinical features were not suggestive of acromegaly in our case and we ruled out hypothyroidism by thyroid function tests. Bone marrow study and protein electrophoresis reports were not in favor of multiple myeloma. Our patient's positive history was enlarged tongue with cardiac involvement with occasional diarrhea.

We did a tongue biopsy with Congo red stain and it revealed sub-epithelial tissue showing focal hyalinization with deposition of eosinophilic structureless substances morphologically resembling amyloid protein. Congo red stain of biopsy material with polarized light was **positive** (Apple-green birefringence in polarized light.) In free light chain assay Kappa–Lambda ratio was increased (8.80).

Final Diagnosis: Primary Systemic Amyloidosis

Amyloidosis is characterized by abnormal deposit of fibril protein within tissues. Usually amyloidosis is systemic, where different organ involvement is present, but rarely amyloidosis can be localized where one organ or one body part is involved. Systemic amyloidosis is sometimes associated with multiple myeloma. Tongue involvement in amyloidosis is not uncommon and typically tongue amyloidosis is secondary to systemic involvement.[1]

Tongue amyloidosis can be localized though unusual. Localized amyloidosis has better prognosis than the systemic disease as surgical treatment is possible in localized disease.[2]

In case of tongue involvement due to systemic disease, local therapy or role of surgery is not clear as recurrence is very common in systemic involvement.[1]

Treatment for systemic amyloidosis: combination chemotherapy with steroid and alkylating agent is effective.

Chemotherapy (Conventional): Melphalan + Prednisolone, Melphalan + Dexamethasone.

Newer agent: Bortezomib, lenalidomide—combination with steroid and alkylating agent.[3]

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101 Interesting Cases in Clinical Medicine

The 101 Interesting Cases in Clinical Medicine provides basic knowledge of various clinical problems that are faced by physicians. It analyses appropriate approach in diagnosing and management of these problems. It provides real diagnostic and management skills which can be applied by practitioners. The clinical cases are compiled in stepwise pattern for physicians that focus on appropriate scientific approach for suitable diagnosis.

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