



CASE

HISTORY

A thin built 60 year old female patient from the rural parts of the western Indian state of Rajasthan was referred from the dental clinic of our hospital. She attended in a wheel chair and had following complaints

- Swelling near the left lower 3rd molar with significant difficulty in eating.
- Difficulty in walking and bony pains over the past 6 months
- Increasing dependency for daily activities



Intra orally single well defined swelling (approximately 4*3 cm) extent: from 41 to 45 and bucco-lingually obliterating buccal and lingual vestibule with normal overlying mucosa.

ON EXAMINATION

- Pallor and general frailty
- Proximal muscle weakness but no other significant neurological abnormalities
- Widespread bony tenderness
- 2 firm non-tender swellings over:
 - Lower half of her right forearm
 - Right hand.



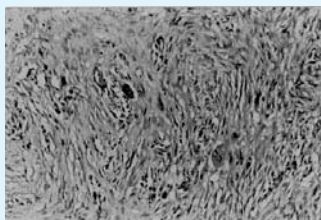
Lab Investigations: table 1

	At presentation	At 4months Follow Up
Total Calcium (2.2-2.6mmol/L)	2.3	2.4
Phosphorus (0.81-1.4mmol/L)	0.63	0.84
Albumin (35-50g/L)	33	
Alkaline Phosphatase (42-98U/L)	124	
Creatinine(44-100µmol/L)	61.9	
Parathyroid Hormone (8-51ng/L)	2842	276
Vitamin D(30-100ng/ml)	14.6	24.6

- Skeletal x-rays showed multiple radiolucent oval lesions (arrows) with appearances suggestive of Brown tumours



- Biopsy of the swelling in mouth demonstrated giant granuloma with an overall appearance of a brown cell tumor.



DIAGNOSIS

- Secondary hyperparathyroidism due to long standing vitamin D deficiency

MANAGEMENT

- 60,000 units of cholecalciferol weekly
- Calcium salts

4-MONTH FOLLOW UP

- Significant improvement in mobility, bony pains and sense of well-being.
- Biochemistry: table 1
- Persistence of oral symptoms including swelling

UNUSUAL FEATURES

- A Brown tumour is a recognised sequelae of long-standing **primary hyperparathyroidism** but the presence of **multiple tumours** has not been reported in a patient with **vitamin-D deficiency**. The persistence of **symptomatic oral tumour requiring excision** is also unusual.

RARE MANIFESTATIONS

- **Brown tumours are rare in a patient** with secondary hyperparathyroidism and **unusual in vitamin D deficiency**. Most previous reports¹ describe them as a sequelae of long-standing **primary hyperparathyroidism**
- Such lesions are rare because hyperparathyroidism is now usually diagnosed and treated before they develop
- This is well illustrated by a study published by Rosenberg and Guralnick

PRESENTATION WITH CONCURRENT MULTIPLE BROWN TUMOURS

- There have been cases³ reported on diverse presentations of vitamin D deficiency including rare case reports of solitary brown tumor but **presence of multiple brown tumors is a rarity**

BROWN TUMOR REQUIRING EXCISION IS UNUSUAL

- Most of such tumors regress with treatment
- However this patient requiring excision of the oral swelling despite treatment makes it an infrequent finding
- ²[Bereket A et al published "Brown tumour can develop in severe, long-standing vitamin D deficiency rickets and responds to vitamin D treatment"](#)

DELAY IN PRESENTATION DESPITE INCREASING DEPENDENCY

- Thus it was striking that whilst painful oral lesion with eating difficulty promptly ensured a visit to a major hospital, progressive dependency due to muscle weakness failed to do so!!!. There are several cultural and economic issues which probably governed this behaviour. This adds to the delay in diagnosis. These constraints governing patients' management in India often vouches for the empirical use of vitamin D therapy
- Patient was also fearful that she had developed oral cancer and this is often the reason for presentation
- The study⁴ by nezih meydan et al mentioned about brown tumor mimicking cancer metastasis in primary hyperparathyroidism.
- The studies and case reports mentioned in literature citing brown tumor as cancer mimetic generally have been in context of primary hyperparathyroidism

TAKE HOME MESSAGE

- **Multiple brown tumours** can be yet another presentation of **long-standing vitamin D deficiency**.
- Rarity of this situation despite such a high prevalence of vitamin D deficiency⁵ in India of the magnitude as high as 50-76% and **not amenable to vitamin D therapy** and necessitating **surgical excision** makes it a noteworthy feature and a **learning experience**
- Common presentations of a common disease is more common than the commonest presentation of a rare disease!
- However uncommon presentation of a common disease can complicate the scenario and can often lead to delayed diagnosis.

REFERENCES

- Rosenberg and Guralnick
- Bereket A, Casur Y, Firat F, Yordam N. Department of Paediatrics, Hacettepe University, Ihsan Dogramaci Children's Hospital, Ankara, Turkey
- K. V. Arunkumar, Sanjeev Kumar, and D. Deepa
- Damodharan Suresh, Chithambaram Nethaji, Penny Hyatt & Neil Halliday
- Erdinc Erturk, Murat Keskin, Canan Ersoy, Tufan Kaleli, Sazi Imamoglu, Gulaydan Filiz
- Nezh Meydan, Sabri Barutca, Engin Guney, Sukru Boylu, Oner Savk, Nil Cullhaci, and Mediha Ayhan
- Cv Harinarayan et al in south India. prevalence of vitamin D