European Journal of Molecular & Clinical Medicine
ISSN 2515-8260 Volume 07, Issue 06, 2020

Soft and hard tissue giant cell lesion as an expression of parathyroid adenoma – A clinical case presentation

Dr Sanat kumar Bhuyan [1], Dr Ruchi Bhuyan[2], Dr Bikash Bishwaddarshree Nayak[3], Siddhant Bhuyan[4], Gautam Ghosh[5], Akankhya Sahu[6]

[1]*Professor, Department of Oral Medicine & Radiology, Institute of Dental Sciences, Siksha O Anusandhan Deemed to be University, Bhubaneswar*

[2]*Professor, Department of Oral Pathology and Microbiology, Institute of MEDICAL Sciences SUM HOSPITAL , Siksha O Anusandhan Deemed to be University, Bhubaneswar*

[3]*Assistant professor, Department of Oral Medicine & Radiology, Hi-Tech Dental College, Hansapal, Bhubaneswar*

[4]*Undergraduate Student, Nepalganj Medical College,Nepalganj, Nepal*

[5]*Professor ,school of pharmacy , Siksha O Anusandhan Deemed to be University Bhubaneswar*

[6] *phd scholar , Siksha O Anusandhan Deemed to be University, Bhubaneswar*

Email id: [1]drsanatkumar68@gmail.com, [2]drruchi30@gmail.com, [3]drbikas@gmail.com, [4]sidhantbhuyan10@gmail.com

Abstract: Brown's tumour is a special rare localized giant cell related tumour which arises as a consequence of the raised parathyroid hormone. Apart from other bones in body, in facial aspect the mandible is the most predominant site in the compared to maxillary involvement. Brown's tumour, usually manifest as osteolytic activity and gross distortion in the bones including maxillofacial bone,this suggests the necessity of timely diagnosis and timelyaccurate management. Here we report afemale patient of 37 years presented with a soft tissue swelling in the gum of rightside of the mandible along with hard tissue involvement of left side as the foremost appearance of main hyperparathyroidism, because of underlying parathyroid adenoma.

Keywords: Brown tumor, multiple bone involvement, Mandible, Parathyroid Adenoma

1. INTRODUCTION:

Hyperparathyroidism (HPT) is a illness happening because of enlarged excretion of parathormone subsequenting toa physical and biological modifications. HPT is classified into four different categories mainly:primary HPT which is mostly initiated through parathyroid adenomas (85%), hyperplasia (10%), besides carcinomas (5%). Secondary HPT follows due to compensatoryrise in parathyroid hormone tohypocalcaemia or vitamin D lack. Tertiary HPT usually results in indivisuals of chronic secondary HPT leading to the dysfunctioning of a parathyroid gland. The last is an ectopic type perceived in individuals through additional malignancies.[1, 2]several periods, hyperparathyroidism is revealedby chance on tedious biochemical investigations or radiological examinations. Clinical manifestation of abnormally high parathyroid hormone levels are giant-cell lesions which is more popularly known as brown tumor. Thestated prevalence of brown tumor is 0.1% and its frequency in secondary hyperparathyroidism is 1.5 to 1.7%. The mandible ismostly commonly involved in the maxillofacial region.[3] The manifestation of the lesion, initiated by a Brown's tumor,

may lead to apparent osteolysis and significant abnormality in the maxillofacial area, which recommends the necessity for building aprimary treatment and administration. We presents a situation of a woman individual who shows by a small painless bulge in the gums of the mandible as the first sign of primary hyperparathyroidism, because of parathyroid adenoma. This case should appeal the responsiveness of oral clinicians to be alert to the potentialincident of Brown's tumors in the jaws who may or may not be having any history of hyperparathyroidism .[4, 5.]

2. CASE REPORT

A patient, who is 37 years old, has noted the major complaint about a gum development since 2-3 months throughout the bottom right mid tooth area. The patient noticed a growth on the right mandibular posterior region since 2-3 months, which was painless and gradually growing in size to attain the present size. Her medical history revealed that she suffered from multiple fractures of forearm and leg (shaft of femur) before 6 years. On external examination, the patient looks of a normalstature and all vital signs with normal limits but she was not able to stand without support due to multiple fractures of forearm and leg. Inextra oral inspection a single diffuse bulge was there on the leftward lower 1/3rd of mouth about 3x2 cm in size extending beyond 1 cm beyond the lower border of the mandible and Dissipate swelling from left cheek, leading to gross asymmetry but overlying skin of swelling appears to be normal. On palpation, a single well circumscribed soft growth was present buccally on the attached gingiva concerning 45, 46 regions about 2 x 1.5 cm in dimensions, Oval in form, superoinferiorly - Spreading from the inter-dental papilla to right gingivo-buccal sulcus Antero-posteriorly - from the mesial side of 45 to the mesial aspect of 46. On palpation, development was solid and – anti-tender, set to the framework of the base. A single diffuse growth was present in the right mandibular alveolus about 3x2 cm in size, bucally from 43 to 47 region.A diffuse swellingnon-tender and bony hard in consistency was also observed on the alveolar mucosa in 33-37 region obliterating left gingivo-buccal sulcus were also palpated. Right side submandibular lymph nodes were tender on palpation, mobile and were not attached to underlying structure. (Fig. 1a-d)

Considering all the clinical findings, site of involvement, history, and characteristics a clinical diagnosis of a benign neoplasm was made.Radiographic examination of intraoral radiographs of molars and orthopantamograph revealed a generalized discontinuity in lamina dura around theteeth, and a diffuse radiolucency above the root of all mandibular molar teeth, sub periosteal erosion of mandibular angle which was suggestive of bony destruction. (Fig 2b-e).A radiographic differential diagnosis of, osteomalacia, and central giant cell granulomaor primary hyperparathyroidism was given. Radiograph (PA SKULL) of head and neck was done, whichrevealed salt pepper appearance with multiple punched out lesions. AP view of hands showed subperiosteal erosion with lytic expansile lesions and multiple fractures with nail placement. (Fig3a-d).Hematological laboratory findings were as follows:), Calcium- 12 mg/dl (8.6-10.3), Phosphorous-2.0 mg/dl, T3 -116.80 ng/dl (84.63-201.08), T4 8.82 micro g/dl (5.13-14.06),TSH 4.54 micro Iu/ml (0.270-4.20), parathyroid hormone- Serum/Plasma- 1061.4pg/ml (14-72), Alkaline Phosphatases (ALKPO4)-428.5 IU/L (40-129.Ultrasonogram (USG) of the neck region revealed a significant , homogenous, small deformity of size, 1 x 1.5 cm, locatedinferior to the lower part of the left thyroid with no involvement of cervical lymph node and it was reported as parathyroid adenoma .(Fig4a)

Excisional biopsy was done of the growth present in the right mandibular attached gingiva concerning 45, 46 and Histopathologically, influential large multinucleated cell to 8–10 nuclei were submitted for analysis, high vascularity with few inflammatory cells, dilated blood vessels with extravasated RBCs. Associating all the Clinical, Radiological,

Biochemical and Histopathological findings brown tumor with Primary Hyperparathyroidism was given as final diagnosis. (Fig2a)

The patient was thanadvised to consult with the Endocrinologist for further intervention and management. Further, to know margins and spread of the tumor, a parathyroid Scintiscan was done with technetium-99m sestamibi, which revealed an abnormal collection of tracers in left lower parathyroid gland suggestiveof Parathyroidectomy. (Fig4b). After one month ofpara thyroidectomy the patient was followed up and she was able to stand with support but after 3 months of follow up, she could walk unsupported with no recurrence of the intraoral lesion. (Fig 5a&b)

3. DISCUSSION

Brown tumors are as a result of hyperparathyroidism show bonylesions with erosion commenced by hasty osteoclastic activity. These lesions are generally benign and theirexpression is like a mass with discreetly cystic and to a certain degree solid regions. They aretypically slow lesions that may be locally destructiveexhibiting in the disparityof symptoms such as noticeable bonyswelling, discomfort and pathological fracture. The typical "brown tumor" is frequentlyappreciated in ends of bones of limbs, ribs and the pelvis.Maxillofacial involvement is uncommonand sometimes may involves the mandible. Parathyroid adenoma is the commonest source of primary hyperparathyroidismand typically presents with warning sign of hypercalcemia.[6,7]Circulating parathyroid hormone elevates serum calcium by the sequential process that is by first stimulatingosteoclastic resorbtion of osseous tissue which raises conversion of vitamin D to active Vitamin D(1,25-dihydroxycholecalciferol).which subsequently increases distal tubular reabsorption of calcium.[8]Maximum individuals through Over 60 years of age are main hyperparathyroidism. Female have this disorder 2 to 4 times as many as males. Throughout a regularly scheduled, biochemical inquiry, the dysfunction is commonly presented as well as the most number of the patients are somewhat symptomless 9. A radiographic investigation, usually involving subperiosteum and swelling including its index phalanges as well as the center digits, is perhaps one of the early clinical symptoms of the condition. The systematic irregularity of the lamina dura anterior to all the teeth is often known as an abimal symptom of the condition. Modifications throughout trabecular design typically grow subsequently. A “ground glass” appearance in some cases, due to decrease in trabecular density.[10] In this reported case patient was 37 years which is rare to have HPT who presented with soft and hard tissue lesion in oral cavity and had history of multiple fractures of legs and hands and had all comparable radiographic features as statedbefore. It is perplexing to discriminate Brown’s tumours since additional huge cell tumours only by the base of histopathological and radiological considerations, the positive diagnosis isalways prepared on the basis ofthorough biochemicalinformation. [11] Foremost treatment for this tumour is surgical elimination of allied parathyroid pathology. Brown’s tumours commonly regress instinctively over time and other treatment choicesconsist of enucleation and curettage, thorough resection with reconstruction, followed by chemotherapyand radiotherapy.[12,] though, many authors suggest to resect any remaining part of tumorafter resolving HPT. Someauthors alsorecommend pharmacotherapy as an adjunct for maintaining the hormone level and prevent recurrence of brown tumor usually vitamin D is given as maintained therapy.[13] Some suggest use of clodronate as an effective drug therapy for hypocalcaemia in management of primaryhyperparathyroidism.[14] In present case surgical excision of tumor was done along with parathyroidectomy and patientwas followed up for a year with no recurrence. Recent studies suggest and recommends the use of methylene blue in felicitating intraoperative

localization and preservation of parathyroid glands by fluorescent imaging which in turn improves outcomes following thyroid and parathyroid surgery[15].

4. CONCLUSION

This case is a unique presentation of primaryhyperparathyroidism due to parathyroid adenoma presenting as brown tumor with soft and hard tissue involvement in jaw as first clinical manifestation. This case emphasizes the oral clinicians to be accustomed with the distinguishing radiographic and clinical presentations in patients with hyperparathyroidism which may represent in jaw boneand gums of maxillofacial region Final diagnosis of HPT can beonly the analysis of both surgical, X-ray, biological and histopathological variables. hugr cell lesions identified histopathologically should be believed and investigated to rule out hyperparathyroidism for appropriate management and averting the reappearance.

REFERENCES

[1] Zou H, Song L, Jia M, Wang L, Sun Y. Brown tumor of multiple facial bones associated with primary hyperparathyroidism: A clinical case report. Medicine. 2018; 97(33).
[2] Farzaneh Amini Nezhada, Moloud Payabb, Sara Nayebandic, Shirin Hasani-Ranjbara,b,. Jaw tumor in recurrent primary hyperparathyroidism: A case report. International Journal of Surgery Case Reports 2018; 52(54-58).
[3] Gulati D, Bansal V, Dubey P, Pandey S, Agrawal A. Central giant cell granuloma of posterior maxilla: first expression of primary hyperparathyroidism. Case reports in endocrinology. 2015; 2015.
[4] Kalapala L, Babburi S, Venigalla A, Pinisetti S, Kotti AB, Ganipineni K. An Endocrine Jaw Lesion: Dentist Perspective in Diagnosis. Case reports in dentistry. 2016; 2016.
[5] Nair PP, Gharote HP, Thomas S. Rare disease: Brown tumour of the jaw. BMJ case reports. 2011; 2011.
[6] Di Fede V, Di Fede O, Rodolico V, Campisi G, Cordova A. Mandibular brown tumor as the first manifestation of primary hyperparathyroidism: a case report. Annali di stomatologia. 2013; 4(2):14-5.
[7] Gadhia T, Adegun OK, Fortune F. Brown tumours: widespread involvement of multiple maxillofacial bones and cervical spine. BMJ case reports. 2014; 2014.
[8] Rai S, Rattan V, Bhadada SK. Giant cell lesions associated with primary hyperparathyroidism. Journal of maxillofacial and oral surgery. 2015; 14(4):930-4.
[9] Azzi L, Cimetti L, Annoni M, Anselmi D, Tettamanti L, Tagliabue A. A Giant-Cell Lesion with Cellular Cannibalism in the Mandible: Case Report and Review of Brown Tumors in Hyperparathyroidism. Case reports in dentistry. 2017; 2017.
[10] Jakovljević B, Grubor G, Jakovljević A, Grubor P. primary hyperparathyroidism-case report of a female patient with advanced disease. Acta medica Medianae. 2009; 48(2):52-4.
[11] Yucesoy T, Kilic E, Dogruel F, Bayram F, Alkan A, Akcan AC, Ozturk F. Spontaneous and Excellent Healing of Bilateral Brown Tumors in Mandible after Endocrinal Therapy and Subtotal Parathyroidectomy: Case Report with 4-Year Follow-Up. Case reports in dentistry. 2018; 2018.
[12] Qaisi M, Loeb M, Montague L, Caloss R. Mandibular brown tumor of secondary hyperparathyroidism requiring extensive resection: a forgotten entity in the developed world?. Case reports in medicine. 2015; 2015.

[13] Hussain M, Hammam M. Management challenges with brown tumor of primary hyperparathyroidism masked by severe vitamin D deficiency: a case report. Journal of medical case reports. 2016; 10(1):166.
[14] Douglas DL, Kanis JA, Paterson AD, Beard DJ, Cameron EC, Watson ME, Woodhead S, Williams J, Russell RG. Drug treatment of primary hyperparathyroidism: use of clodronate disodium. Br Med J .Clin Res Ed. 1983; 286(6365):587-90.
[15] Hillary SL, Guillermet S, Brown NJ, Balasubramanian SP. Use of methylene blue and near-infrared fluorescence in thyroid and parathyroid surgery. Langenbeck's archives of surgery. 2018; 403(1):111-8.

FIGURE LEGENDS

Fig 1(a) Patient reported to the outdoor on wheelchair**. (b)** Extraoral swelling on left side of mandible. **(c)**Intraoral presentation on right side of mandible on gingiva. **(d**) Intraoral bony swelling on left side of mandible.

Fig 2(a)H&E section under 40x showing giant cells with extravasted RBCs and blood vessels. **(b)**Occlusal x-ray showing bony expansion of left side on mandible. **(c,d,e)** Showing loss of lamina dura in relation with all teeth.

Fig 3 (a) PA skull view showing punched out lesions. (**b**) Lateral skull showing salt pepper appearance. **(c)** AP view of hands showing sub periostealerosion. **(d)** AP view of legs showing multiple fractures with nail placement.

Fig.4(a) Ultrasonogram (USG) of the neck regionshowing parathyroid adenoma**. (b**) Scintiscan with technetium-99m sestamibi, revealing an abnormal collection of tracers in left lower parathyroid gland.

Fig 5(a) Patient followed up after one month was able to stand with support. **(b)** 3 months of follow up, she could walk unsupported.

Fig 5a

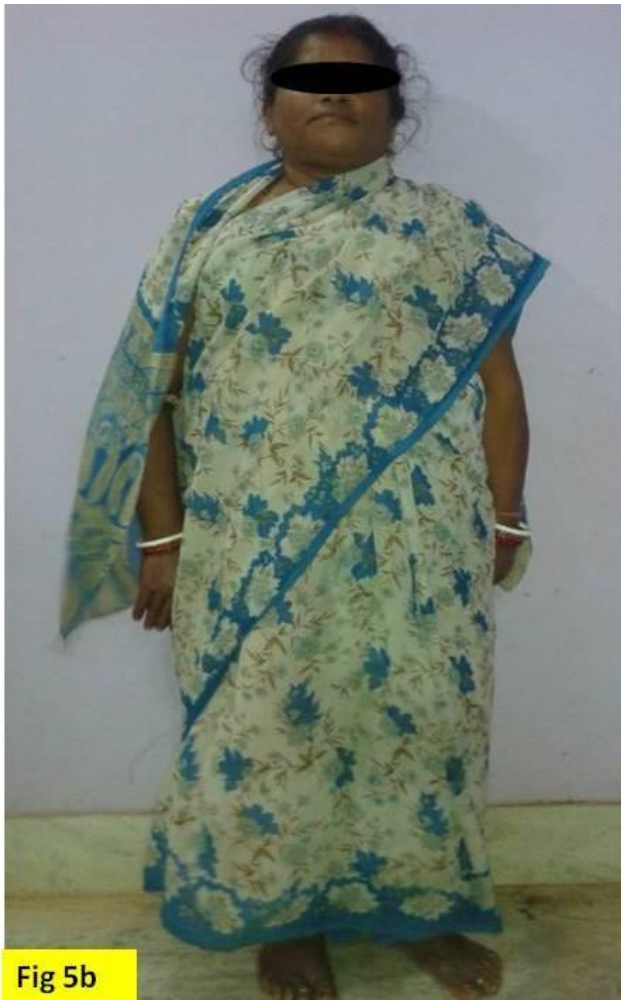

Fig 5b

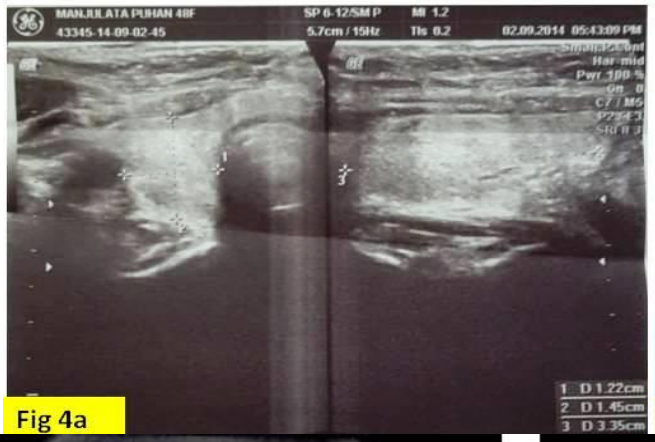

Fig 4a

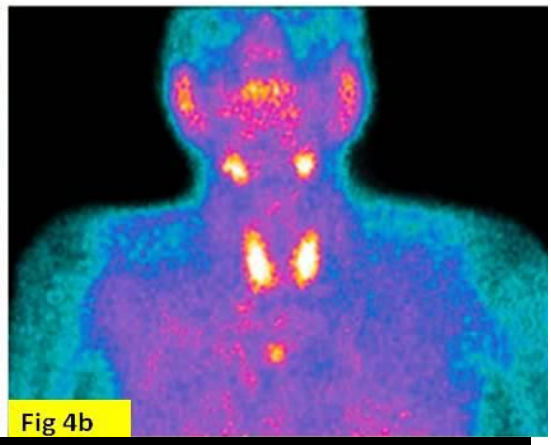
Fig 4b

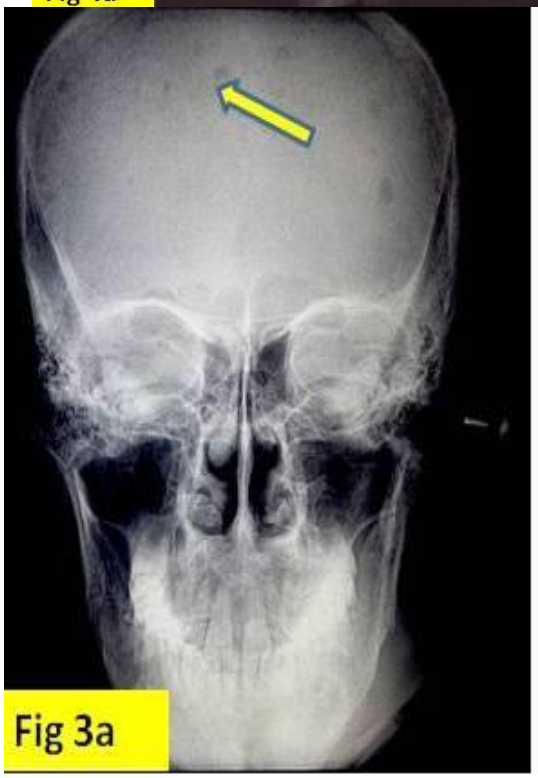
Fig 3a

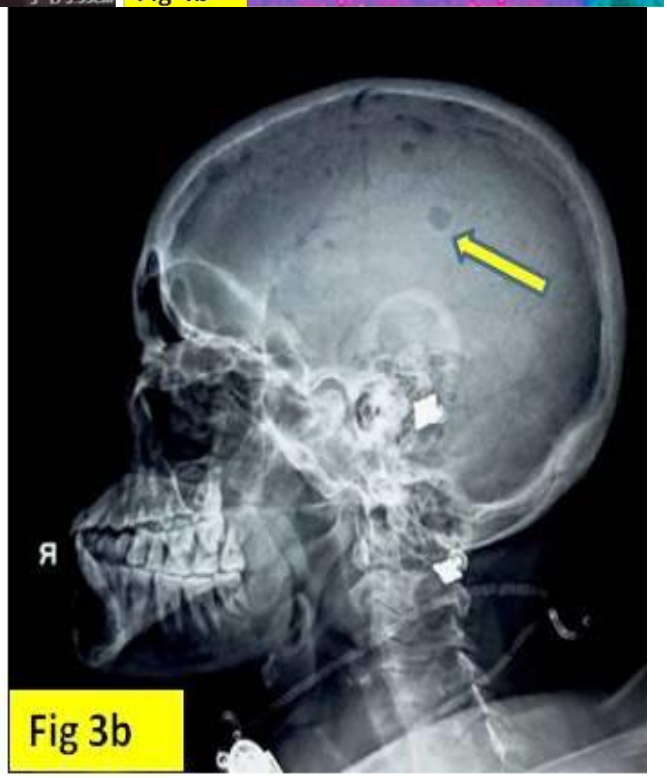
Fig 3b

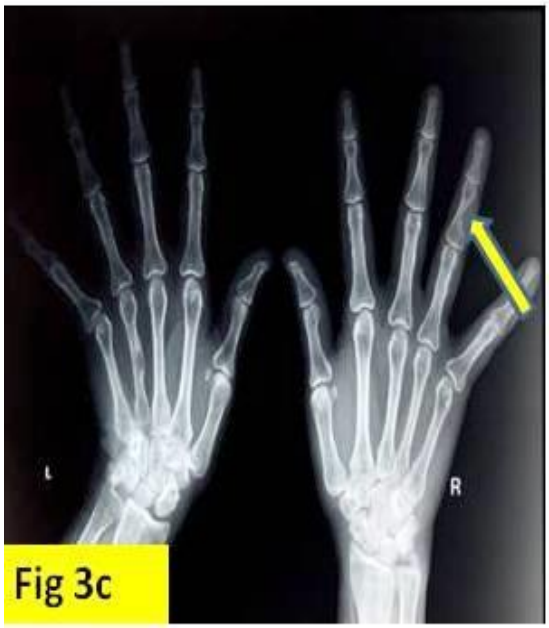
Fig 3c

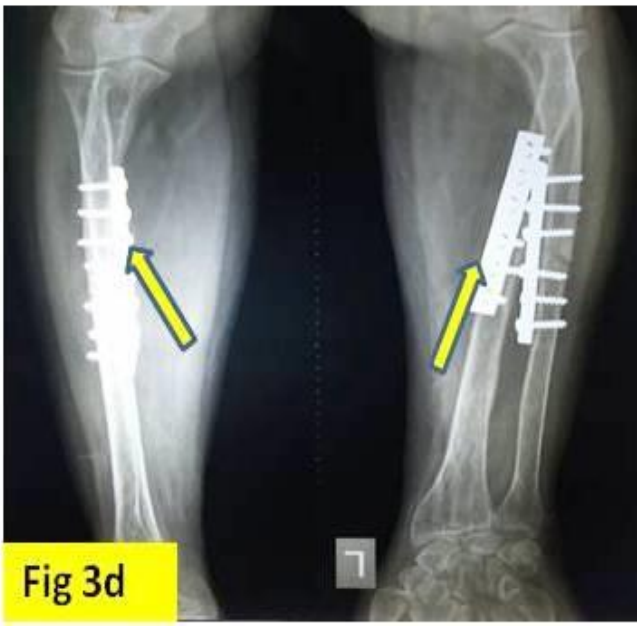
Fig 3d

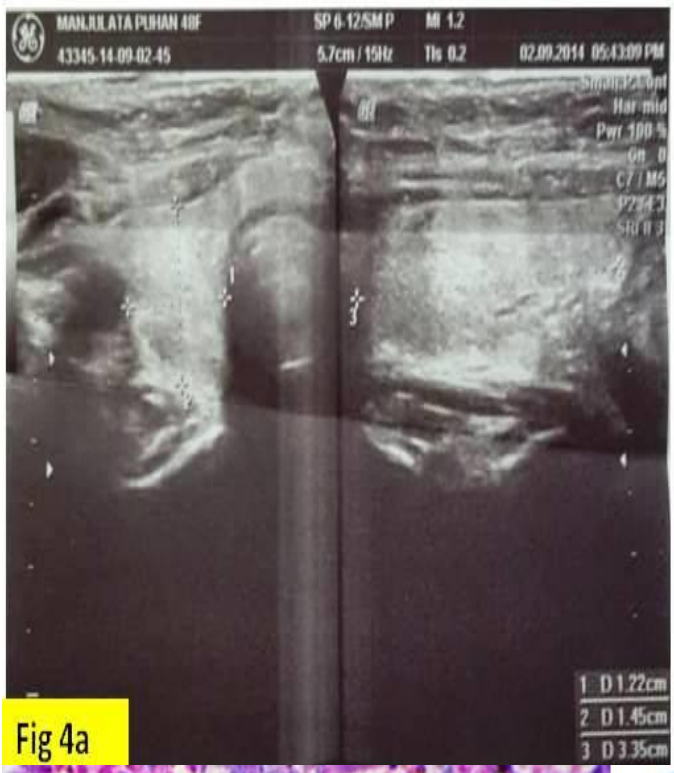

Fig 4a

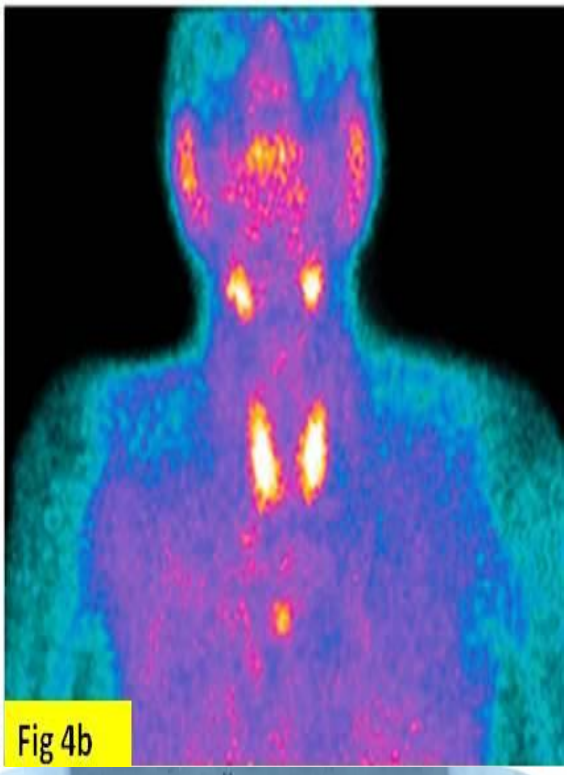
Fig 4b

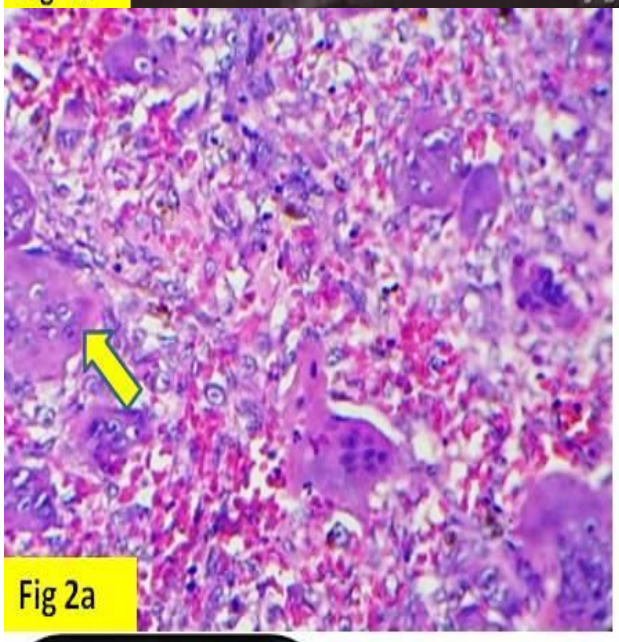
Fig 2a

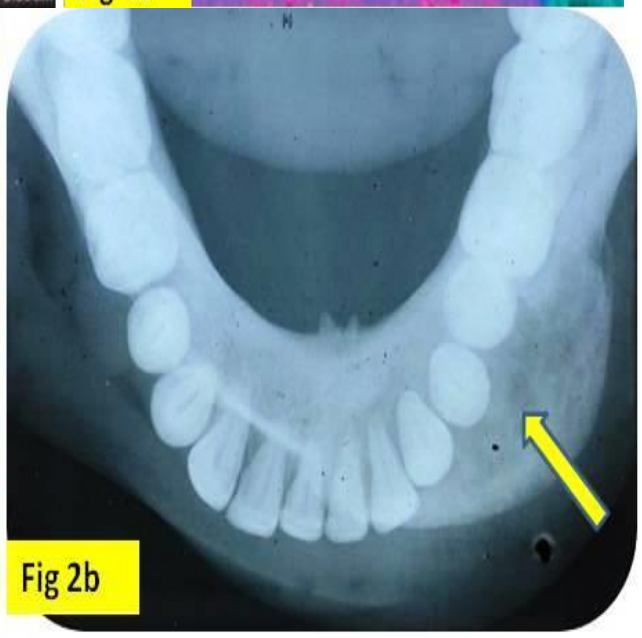
Fig 2b

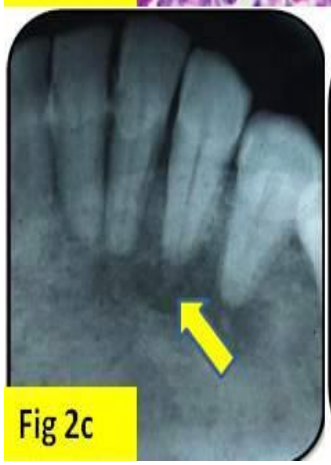
Fig 2c

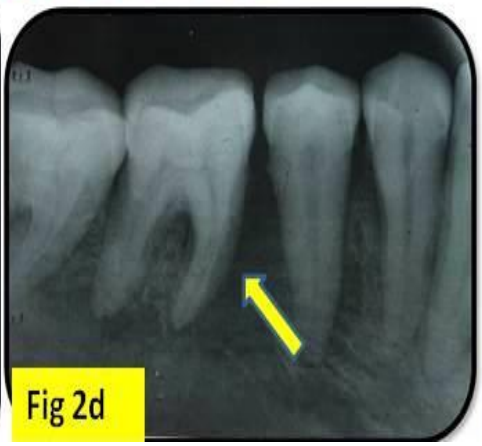
Fig 2d

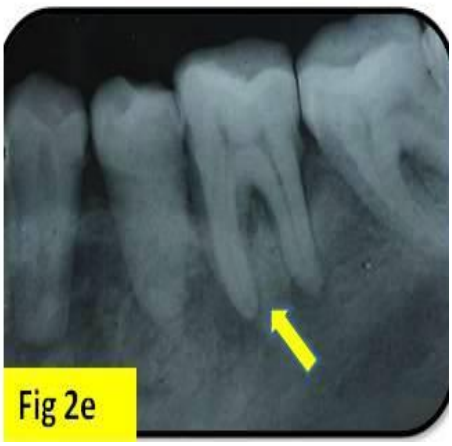
Fig 2e

Fig 1a

Fig 1b

Fig 1c

Fig 1d