VOLUME - 13, ISSUE - 07, JULY - 2024 • PRINT ISSN No. 2277 - 8160 • DOI : 10.36106/gjra

Original Research Paper

General Surgery

A DOUBLE PARATHYROID ADENOMA & ONE BEING INTRATHYROIDAL - A RARE ENTITY

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ABSTRACT A rare case report on Double Parathyroid ADENOMA & one being Intrathyroidal with Primary Hyperparathyroidism & hypercalcemic crisis.

KEYWORDS :

INTRODUCTION:

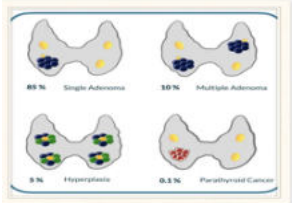
Parathyroid adenomas are the most common benign parathyroid tumors leading to Primary Hyperparathyroidism & hypercalcemia, which in turn can cause major morbidity to the patients. These patients can present with myriad manifestations in the form of muscle weakness, nausea, vomiting, pathological fractures, and neuropsychiatric disturbances.

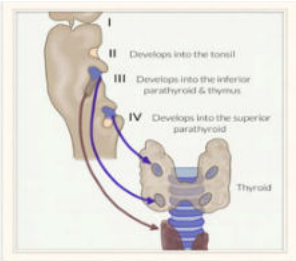
The diagnosis of this entity is not straightforward and requires a strong suspicion on the part of the primary clinician. Diagnosis is reached by a combination of biochemical and radiological investigations.

Technetium 99m SestaMIBI is the diagnostic imaging of choice, as the dye is actively concentrated in the parathyroid glands.

The mainstay of treatment is surgical excision, which can be challenging due to difficulty in identifying the offending gland and also the close proximity of the gland to the recurrent laryngeal nerve.

Primary hyperparathyroidism (PHPT) is an endocrine disease that affects 0.1–0.7% of the general population. After the establishment of diagnosis of PHPT, the localization of the pathological parathyroid gland should be investigated. Parathyroid tumours are usually located in the neck region especially on the posterior capsule of the thyroid or in ectopic locations. Only a few of them are ectopically occurred in the mediastinum due to the migratory pathways of embryologic parathyroid tissue.





An intrathyroidal parathyroid adenoma (IPA) is an ectopic variant where the adenoma is either partly (>50%) or completely enveloped by the thyroid gland. The incidence of IPAs ranges from 0.7% to 6%. 1 , 2 , 3 IPAs can be challenging to manage for a range of reasons. For instance, on imaging, they can appear similar to other structures in the thyroid, including benign thyroid nodules, making differentiation difficult. In the operating room, they can be challenging to find, being embedded in the thyroid. As a result, a thyroid lobectomy is often performed, sometimes with inconclusive prehistological evidence that an IPA is present and can lead to high failure rates.

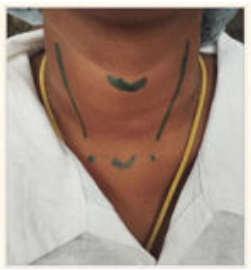
Case Presentation:

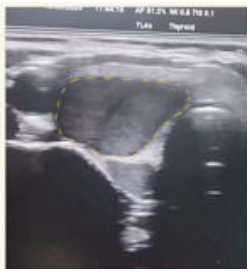
A 30 yr Female Homemaker came with chief complaints of Abdominal pain,Vomitting & loss of appetite for past 2 months. Recently Undergone ERCP stenting & Laparoscopic Cholecystectomy 45 days before admission in private sector,Later presented with B/L HUN (Grade 1)presented with Acute kidney Injury & Hypercalcemic crisis(S.calcium -17.1) & Diagnosed as Primary Hyperparathyroidism(PTH-1517 pg/mL)& Vitamin D-11 ng/mL ,S.Po4-2.0 mg/dL & S.Mg-1.1 mg/dL & Alakaline Phosphatase-1076IU/L

Clinically swelling palpable of size 3*2cm noted over Rt side of thyroid, smooth surface & soft in consistency,cervical Lymph Node not palpable & B/L Carotid palpable & Swelling moves with Deglutition & doesn't Moves with protrusion of tongue,No other swelling present.

Patient treated with Inj Zoledronate 4mg consecutively & Serum Calcium brought down to 9.1 mg/dL & Screening for MEN 1 syndrome done. S. Prolactin -21ng/mL, no pancreatic NET in CECT abdomen

Radiologically **USG NECK** shown -A well defined Hypoechoic lesion of size 4*1.5*2.3cm noted posterior to upper part of RT lobe of thyroid with Internal Vascularity & In Lower part of Left lobe hypoechoic lesion of size 1.5*1cm seen, thyroid nodule/ Suspicious Intrathyroidal parathyroid. Rest of the thyroid lobes appear Normal .. Bone Scan Shown OSTEOPENIA.



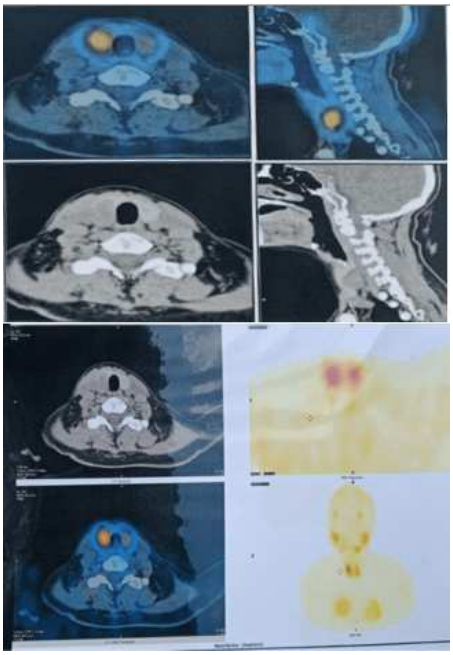


99mTc- Sestamibi Parathyroid SPECT/CT Scan shown Well defined soft tissue density lesion with abnormal increased 99mTc-sestamibi uptake measuring ~ 25 x 15 x 24 mm noted posterior to the right lobe of thyroid gland indenting it anteriorly with indistinct fat plane.

Well defined nodule within the left lobe of thyroid gland along its inferior pole with mild increased 99mTc-sestamibi uptake? Thyroid nodule/? Intra-thyroidal parathyroid ADENOMA.

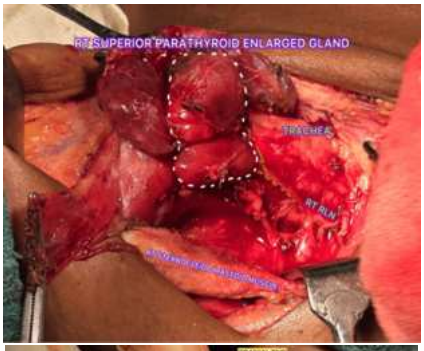
Pre operative planned : for Right Superior Parathyroidectomy

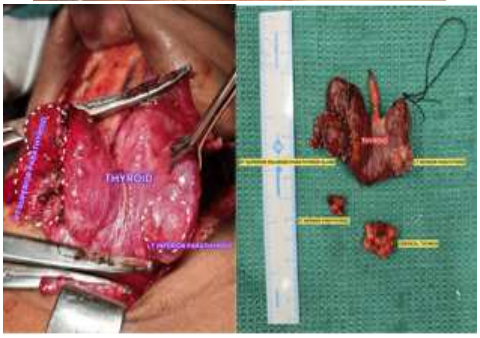
& Left Hemithyroidectomy due to Left Inferior Intrathyroidal Adenoma



Intra-operative Finding:

Intra operatively Right Superior Parathyroid enlarged & adherent to Right thyroid lobe,so proceeded with Total Thyroidectomy & Cervical Thymectomy done in suspicion of MEN 1 leaving behind Left Superior Parathyroid gland.

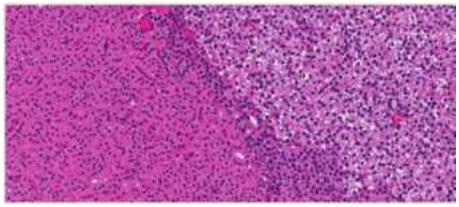




Histopathological Examination :-

Features Suggestive of Rt Parathyroid Adenoma /Hyperplasia & Left Inferior Intrathyroidal Parathyroid Adnenoma.

Postoperatively PTH drastically Dropped to a value of 16 pg/mL.Serum Calcium level dropped to a least value of 7.1 mg/dL (Corrected Calcium) & her Hungry Bone Syndrome managed aggressively with parenteral calcium & Oral Supplement along with Activated Vitamin D3 .At the time of Discharge Serum Calcium was 9.4 mg/dL & Alkaline Phosphatase in decreasing trend at 453 IU/L.



DISCUSSION:

Primary hyperparathyroidism is mediated through hypercalcemia, and this clinical picture includes osteitis fibrosa cystica, nephrolithiasis, and neuropsychiatric symptomatology. Severe hypercalcemia may precipitate alterations in mental status, cardiac arrhythmias, and pancreatitis

Renal manifestations most commonly include nephrolithiasis. Less commonly, acute kidney injury is seen. The role of hypercalcemia in AKI is multifaceted and includes hyposthenuria through the downregulation of aquaporin 2 channels and tubulointerstitial injury mediated by medullary calcium deposition. Prerenal azotemia is elicited through renal vasoconstriction and prostaglandin E2-mediated reduction in NaCl reabsorption

Hungry bone syndrome (HBS) is a rapid, intense, and prolonged hypocalcemia that follows parathyroidectomy. It is caused by a sudden drop in PTH levels and its impact on osteoclastic resorption. Risk factors include age >60, PTH level >1000pg/ml, and alkaline phosphatase level three times the upper limit of normal. The main goal of treatment is replacing calcium deficiency through supplementation with calcium salts and high doses of active vitamin D

Intrathyroidal Parathyroidal Adenomas are the second most common cause of persistent hyperparathyroidism, after intrathymic adenomas.True IPA occurred in only 0.7% of 10,000 primary cases, whereas another 1.2% were closely adherent to or partially within the thyroid substance.IPA occurs in predictable locations within the thyroid gland, with the vast majority occurring in the lower lateral quadrant and a small percentage near the recurrent laryngeal nerve and superior pole. The parathyroid gland occurs as an intrathyroidal variety when it becomes trapped within the thyroid as the lateral and medial lobes fuse. Although this is the common mechanism for both superior and inferior glands, the latter (derived from the 3rd branchial pouch) are pulled by the thymus during its descent, and because of migrating a longer distance, they have an increased chance of becoming entrapped during the fusion of the thyroid lobes.Most useful investigations in the localization of IPA are USG and^{99m} Tc-sestamibi scans.

CONCLUSION:

Symptoms of Primary hyperparathyroidism vary and lack specificity.Parathyroid adenomas are the most common cause of primary hyperparathyroidism. Treating clinicians should have an high index of suspicion for diagnosing this rare entity. Accurate localization of the offending gland is the key to a correct diagnosis and a good surgical outcome.

99mTc- SestaMIBI scan is the gold standard investigation for parathyroid gland with high sensitivity and specificity. Exact

anatomical location and good surgical techniques provide the recipe for a Sucessful Surgery and uneventful postoperative period.

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