Case Report

A rare case of giant parathyroid adenoma presenting with recurrent episodes of pancreatitis

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ABSTRACT

Parathyroid adenomas are usually small in size; (weighing 70 mg–1 g) those weighing more than 2–3 g are classified as giant parathyroid adenomas. Giant parathyroid adenomas are in fact rarely encountered among patients with primary hyperparathyroidism. They are believed to have distinct clinical and biochemical features related to specific genomic alterations. We chanced to manage a unique and possibly the first case of giant parathyroid adenoma (6 cm diameter and weighing 20 g) presenting with recurrent episodes of pancreatitis and discuss its surgical management with an added emphasis on the role of nuclear imaging in its preoperative localization. Our case demonstrates that clinicians should have a high index of suspicion of primary hyperparathyroidism in patients presenting with recurrent episodes of pancreatitis. Timely diagnosis, appropriate preoperative localization techniques, which would include a parathyroid scintigraphy and a focused surgical intervention are crucial to resolve complications and improve outcomes.

Keywords: Giant parathyroid adenoma, pancreatitis, primary hyperparathyroidism, Tc99m sestaMIBI scintigraphy

INTRODUCTION

Primary hyperparathyroidism is most commonly caused by a single parathyroid adenoma (85–90%) and multi-gland disease in approximately 10% of patients. Patients may rarely develop primary hyperparathyroidism due to a parathyroid carcinoma. The size of abnormal parathyroid glands in patients with primary hyperparathyroidism is highly variable, those weighing more than 2–3 g are classified as "giant parathyroid adenomas" that are very rarely reported in literature.[1,2] We report a rare case of giant parathyroid adenoma presenting with recurrent episodes of pancreatitis and discuss its management with an emphasis on the role of preoperative localization.

CASE REPORT

A 50-year-old male without any comorbid illnesses was being evaluated and conservatively managed for recurrent attacks of acute pancreatitis at an outside canter for 9 months. His biochemical evaluation incidentally showed hypercalcemia (11.2 mg/dl) and further investigations revealed elevated levels of serum paratharmone (669 pg/ml). A computed tomography (CT) neck and chest done at the referring center revealed a 6 cm × 4 cm mass in the left paratracheal region with an extension to the superior mediastinum. An ^{18}F-fluorodeoxyglucose (^{18}F-FDG) positron emission tomography/CT (PET/CT) scan was additionally done which confirmed the isolated uptake standard uptake value: 4.3 in the left paratracheal region [Figure 1a and b]. He was subsequently referred to our center for further management. Clinical evaluation at our center was unremarkable except for a vague fullness in the left paratracheal region. The left paratracheal mass lesion was localized to the left inferior parathyroid gland using Tc99m SestaMIBI scintigraphy, which demonstrated a persistent retention of the sestamibi tracer in the left inferior parathyroid gland [Figure 2]. An ultrasound guided fine-needle aspiration cytology from the lesion revealed a benign epithelial lesion that could not be further characterized. A provisional diagnosis of primary hyperparathyroidism due to a giant parathyroid adenoma was made and the patient was planned for a definitive surgery.

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The patient underwent an uneventful resection of the tumor-bearing left inferior parathyroid gland through a focused trans-cervical approach after carefully safeguarding the left recurrent laryngeal nerve [Figure 3a-d]. The patient made an uneventful recovery and was discharged on oral calcium supplementation for biochemical hypocalcemia that normalized in a couple of weeks. The postoperative serum parathormone levels also normalized. The final histopathology confirmed the diagnosis of a giant parathyroid adenoma [Figure 4] and the patient is on regular follow-up.

DISCUSSION

Giant parathyroid adenomas are in fact rarely encountered among patients with primary hyperparathyroidism. They are believed to have distinct clinical and biochemical features related to specific genomic alterations.[1] Parathyroid adenomas are usually located on the posterior capsule of the thyroid but may be found in other ectopic locations. An enlarged parathyroid gland, as seen in our patient generally descends into the mediastinum because of its weight and as a result of the negative intra-thoracic pressure.

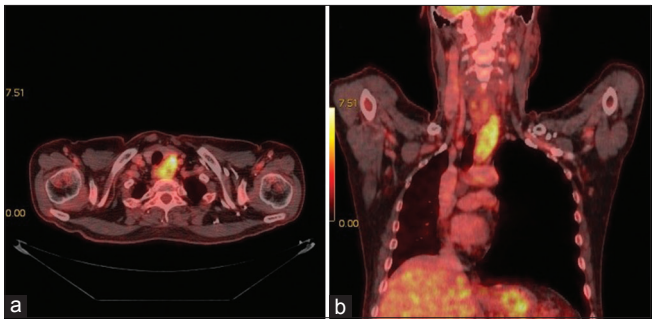

Figure 1: (a and b) ^{18}F-fluorodeoxyglucose positron emission tomography/computed tomography revealed tracer uptake in a 6 cm × 4 cm mass in the left paratracheal region with an extension to the superior mediastinum. (Standard uptake value 4.3) no other significant uptakes were seen

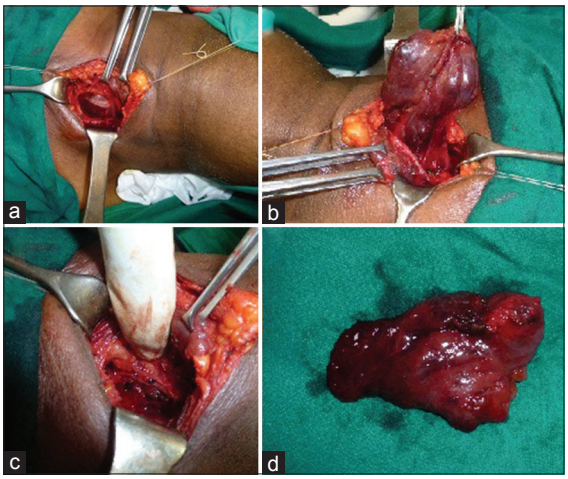

Figure 3: Intraoperative photograph showing the focused neck exploration. (a) The left recurrent laryngeal nerve splayed over the left giant parathyroid adenoma. (b) The left parathyroid adenoma dissected off the nerve and delivered in the neck. (c) Operative bed following the tumor removal. (d) Specimen photograph of the giant parathyroid adenoma

The clinical presentation of primary hyperparathyroidism has changed over the years from a severe, debilitating disease to a disease with subtle symptoms and physiologic derangements.[2] The diagnosis of primary hyperparathyroidism rests on the laboratory confirmation of increased serum calcium levels and inappropriately elevated levels of serum parathormone. Some authors have suggested that the weight of the parathyroid adenoma has a direct correlation with the functional status of the gland and the severity of biochemical abnormalities, but this was not true in our patient. The incidence of acute pancreatitis associated with hyperparathyroidism is reported to be <10%.[3,4] Although the actual causal relationship between hypercalcemia and pancreatitis has been a topic of debate, detection of hypercalcemia points to a differential diagnosis of primary hyperparathyroidism as was encountered in our patient due to a giant parathyroid adenoma.

Preoperative localization of the hyper-functioning parathyroid gland/glands is considered vitally important prior to definitive surgical management.[1] Several diagnostic localization methods, both invasive and noninvasive exists, which range from ultrasound, CT scans, magnetic resonance imaging, Tc99m SestaMIBI scintigraphy and PET scanning in the preoperative setting and gamma probe and intraoperative parathyroid hormone assays in the intraoperative setting. Preoperative localization studies with two concurrent imaging techniques (combined ultrasound and Tc99m SestaMIBI scintigraphy) are increasingly becoming

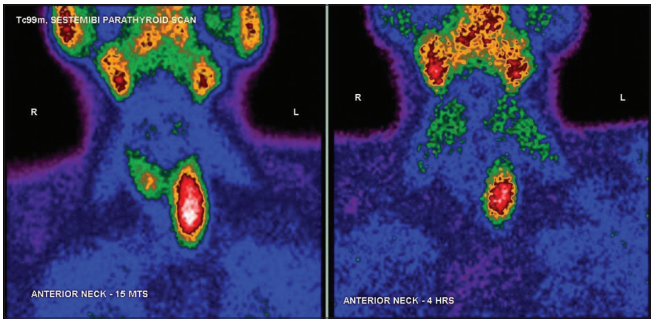

Figure 2: 25 mCi of 99mTc-SestaMIBI was given intravenously followed by the acquisition of early and delayed images of the neck and upper thorax and/or mediastinum. The initial early image is obtained at 15 min after the injection, and delayed imaging is obtained at 1, 2 and 4 h after the injection. The scan demonstrated a persistent retention of the SestaMIBI tracer in the left inferior parathyroid gland at 4 h

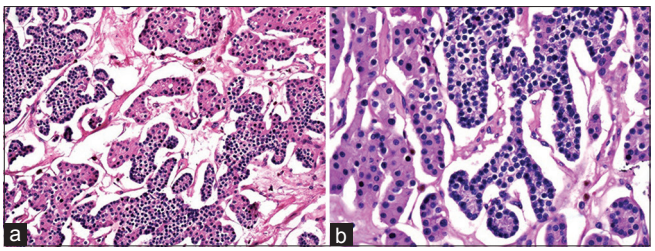

Figure 4: (a and b) H and E, ×20 section shows tumor cells which are large with moderate eosinophilic granular cytoplasm, fine nuclei and tiny nucleoli, oncocytic type of cell clusters are seen, with areas of hemorrhage, no capsular or vascular invasion are seen, the findings are suggestive of a diagnosis of parathyroid adenoma

increasingly popular as they enable minimally invasive surgical approaches.[5,6]

Nuclear imaging with parathyroid scintigraphy is the primary and standard method used for preoperative localization. The sensitivity of Tc99m SestaMIBI scintigraphy in detecting parathyroid adenomas has been reported to range from 70% to 100%. The use of this single-isotope dual-phase scintigraphic technique, is based on their observation that 99m Tc-sestamibi washes out more rapidly from the thyroid gland than from hyper functioning parathyroid glands[7] hybrid Tc99m SestaMIBI single-photon emission CT/CT imaging is in fact superior to CT and Tc99m SestaMIBI scintigraphy alone in the preoperative localization of the hyper-functioning pathologic glands, especially in patients suffering from multiglandular disease with primary hyperparathyroidism.[8,9]

Although the role of ^{18}F-FDG PET/CT scans in identifying parathyroid carcinomas is well established, its use in the localization of parathyroid adenomas has yet to be widely utilized.[10] A recent study reported that clinicians should possibly consider PET-CT as a second line modality when conventional imaging fails to localize the hyper functioning parathyroid gland.[11] There have been a few reports for the use of ^{11}C-methionine PET as a promising imaging modality for localizing parathyroid adenomas.[12]

Surgical excision of the hyper functioning gland is the treatment of choice for parathyroid adenomas.[13] The surgical approach for an inferior giant parathyroid adenomas extending on to the mediastinum can be cumbersome requiring a full collar incision or occasionally a median sternotomy. Our case further demonstrates that a focused trans-cervical excision of giant inferior parathyroid adenoma is a viable approach for resection and should be considered prior to a formal neck exploration or sternotomy. Further it is believed that following aggressive medical management of acute pancreatitis, parathyroidectomy can improve the clinical outcome and prevent further recurrences of pancreatitis.[4]

CONCLUSION

Clinicians should have a high level of suspicion of primary hyperparathyroidism in patients presenting with recurrent episodes of pancreatitis. Timely diagnosis, appropriate preoperative localization techniques that would include a parathyroid scintigraphy and a focused surgical intervention are important to resolve complications and improve outcomes.

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Conflicts of interest
There are no conflicts of interest.

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