IMAGE

Parathyroid lipohyperplasia – a rare and difficult to localize entity

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INTRODUCTION

Primary hyperparathyroidism (PHPT) is estimated to be the third most commonly diagnosed endocrine disorder.[1] The clinical presentation is usually variable and ranges from an asymptomatic presentation to florid manifestations of hypercalcemia such as renal calculi, bone disease and pancreatitis.[2] Parathyroid lipohyperplasia is an extremely rare variant of hyperparathyroidism with just about 10 cases reported till date.[3-6] Pre-operative localization of parathyroid adenoma is important for planning surgery – minimally invasive focused parathyroidectomy vs. cervical exploration. We report a case of parathyroid lipohyperplasia which posed difficulty in pre-operative localization.

CASE HISTORY

A 22-year-old gentleman presented with complaints of abdominal pain. On evaluation, he was found to have moderately severe acute pancreatitis. Further evaluation revealed hypercalcemia with high parathyroid hormone (PTH) of 95.4 pg/mL. His body mass index (BMI) was 22 kg/m^2. His highest recorded calcium was 12.5 mg/dL. Bone mineral density showed a well preserved bone mass and imaging of abdomen did not reveal any renal calculi. He underwent a sonogram of the neck and a Sestamibi (technetium-99 m-methoxy isobutyl isonitrile) scan, both of which were reported to be normal [Figure 1]. Further 4D CT (4-dimensional computerized tomography) scan was done to localize the lesion and was inconclusive. Considering his young age and an episode of acute pancreatitis, it was decided to go ahead with cervical exploration and this revealed hyperplastic parathyroid glands. The right superior and inferior glands were larger with both weighing about 200 mg. He underwent excision of three glands. The left inferior was almost normal in size and hence was preserved. The histopathological examination revealed hypercellular parathyroid glands with abundant lipomatous component, and was consistent with parathyroid lipoadenomatous hyperplasia [Figures 2 and 3]. His parathormone level dropped to 12.2 pg/mL and calcium normalized to 9.3 mg/dL.

DISCUSSION

In the reported case, the patient presented with acute pancreatitis and was found to have a PTH dependent hypercalcemia. However, a definite lesion could not be localized by conventional imaging techniques that included a sonogram of the neck, parathyroid scintigraphy and a 4D CT. On cervical exploration, he was found to have hyperplasia of parathyroid glands which was histologically consistent with a parathyroid lipohyperplasia. The clinical presentation of hyperparathyroidism caused by lipohyperplasia is similar to that caused by parathyroid hyperplasia. In the previously reported cases, there was a female predilection (8 of the 10 reported cases were females) and the mean age of presentation was 60 years.[3-6] In contrast, our patient was a young male – this is by far the youngest case of parathyroid lipohyperplasia reported till date. Parathyroid lipohyperplasia usually presents with lower calcium levels, the highest calcium in our case being 12.5 mg/dL. Most of the reported cases presented with non-specific complaints.[7] Our patient, however, presented with acute pancreatitis. Parathyroid lipohyperplasia may be difficult to identify by a sestamibi scan because of the multi-glandular

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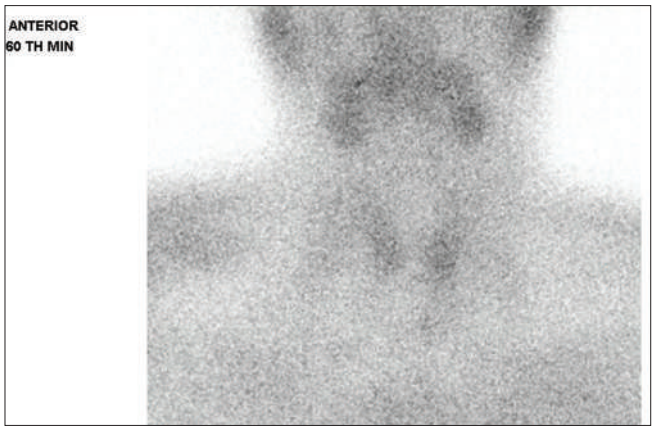

Figure 1: Sestamibi scan of the subject – negative for parathyroid adenoma

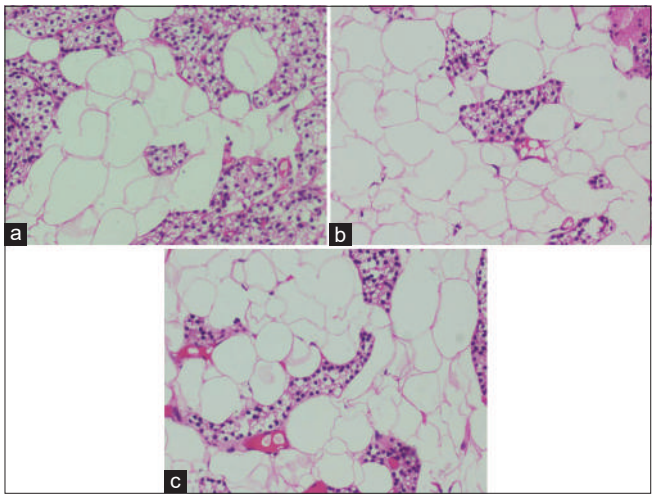

Figure 3: Abundant stromal fat with interspersed islands and cords of chief cells (200 × H and E sections). (a) Left superior parathyroid (b) Right inferior parathyroid (c) Right superior parathyroid

involvement, high adipose content, and relatively small size as compared to parathyroid adenomas. We faced this difficulty in this scenario and the lesion was not localized on sestamibi, ultrasound or a 4D CT.

Parathyroid lipohyperplasia when functional, behaves similar to parathyroid hyperplasia and the line of management is the same. With only 10 cases reported till date, the specific characteristics of lipohyperplasia and their relation to the overall prognosis of the tumor is not known. However, because of high adipose tissue content, it causes difficulty in pre-operative tumor localization and hence warrants a cervical exploration rather than a minimally invasive surgical procedure. In summary, lipohyperplasia are rare variants of primary hyperparathyroidism that may pose difficulties in pre-operative clinico-radiographic localization.

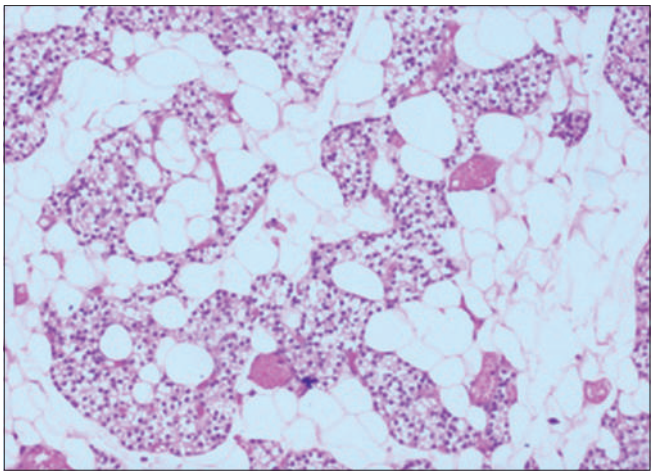

Figure 2: Histopathology of parathyroid lipohyperplasia-40 × magnification. Show sections of a benign lesion composed of proliferation of both epithelial and stromal elements. The epithelial component comprises islands and branching cords of parathyroid epithelial cells, predominantly chief cells and a few oxyphil cells, which are set in a background of abundant (~50%) mature lipomatous tissue, which forms the stromal component

Awareness of this entity is important when PTH dependent hypercalcemia is encountered with negative imaging.

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Conflicts of interest

There are no conflicts of interest.

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