Recurrent Postoperative Parathyroid Carcinoma: FDG-PET and Sestamibi-SPECT Findings

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Parathyroid carcinoma is a rare cause for primary hyperparathyroidism, accounting for between 0.5% and 5% of all cases (1). Along with laboratory evaluation, preoperative localization studies are useful. Reports of successful preoperative imaging of parathyroid carcinoma with ultrasonography, CT, MRI and several radionuclide techniques have been reported (2–4). Among these scintigraphic methods, parathyroid carcinoma has been successfully imaged with thallium-technetium subtraction, gallium and 99mTc-sestamibi (5–8).

We present a case of postoperative recurrence of parathyroid carcinoma with misleading findings on both MRI and 99mTc-sestamibi scintigraphy, which was correctly localized before reoperation by PET using [18F]fluorodeoxyglucose (FDG).

CASE REPORT

A 65-yr-old man presented with a 4-mo history of increased arm and leg pain, weakness, fatigue and visual blurring. Physical examination was unremarkable; in particular, no palpable neck masses were seen.

Initial laboratory evaluation demonstrated elevated serum calcium (16.0 mg/dl) and serum parathyroid hormone levels (130 pg/ml, normal range of 10–60 pg/ml). The working diagnosis based on these findings was primary hyperparathyroidism. A surgical neck exploration was then performed. A mass infiltrating the left lobe of the thyroid was found invading the adjacent esophagus and the left recurrent laryngeal nerve.

The resected pathological specimen stained negative for thyroglobulin, thyrocalcitonin and immunoperoxidase for chromogranin but positive with CAM 5.2 staining keratin found in parathyroid tissue. The surrounding thyroid tissue also stained positive with CAM 5.2 and, thus, was not felt to be significant. The histopathologic examination was initially interpreted as metastatic undifferentiated carcinoma, probably of pulmonary origin. The patient’s hypercalcemia resolved postoperatively without further treatment.

Further work-up for a primary neoplasm, including chest radiograph, bronchoscopy with biopsies and bronchial washings and CT of the chest were negative.

Eleven months after the initial surgery, a CT scan of the neck remained negative for tumor recurrence, but MRI revealed the presence of a mass lesion posterior to the trachea and esophagus (Fig. 1). The serum-intact parathyroid hormone level was 160 pg/ml and serum calcium was 13.5 mg/dl. The original pathological specimen was re-evaluated at this time and interpreted as primary carcinoma of the parathyroid.

A double-phase SPECT study of the neck and chest, obtained immediately and 2 hr following intravenous injection of 99mTc-sestamibi, demonstrated activity associated with the right thyroid lobe but was otherwise negative (Fig. 2).

After intravenous administration of [18F]FDG, regional-body PET imaging of the neck and chest demonstrated intense FDG accumulation in the left thyroid bed that extended posteriorly across the midline in the prespinal region (Fig. 3). The patient then underwent a second surgical neck exploration. At surgery, a palpable lesion in the left thyroid bed was found. This hard tumor was adhered densely to the left internal jugular vein and left carotid artery. Further exploration revealed hard tumor between the left carotid artery and the esophagus, which extended posteriorly to the cervical spine and across the midline, and was felt to be nonresectable in this region. An en bloc resection of tissue from the left thyroid bed was performed. Histopathologic evaluation of the specimen was positive for parathyroid carcinoma.

One day postoperatively, the serum calcium level decreased to 8.3 mg/dl. The patient is currently receiving external radiation therapy to the neck for recurrence of parathyroid carcinoma.

DISCUSSION

Parathyroid carcinoma is a rare cause for primary hyperparathyroidism, accounting for between 0.5% and 5% of all cases. Patients may present with symptoms related to hypercalcemia, such as lethargy, fatigue, mental depression or psychosis, weight loss, muscle weakness, bony pain or renal colic (1,8–11). Laboratory abnormalities include marked hypercalcemia,

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FIGURE 1. MRI of the neck demonstrates a mass lesion posterior to the trachea and esophagus. The mass (arrow) is isointense on the sagittal image (left). Transverse image (right) shows no abnormal signal in the left peritracheal region (arrowhead) and abnormal hyperintensity (arrow) posterior to the trachea.
increased urinary calcium and elevated levels of serum parathyroid hormone (12).

The diagnosis of parathyroid carcinoma is often made during surgery or histopathologic examination. The presence of fibrous bands, mitotic figures and vascular invasion have been included in the histopathologic criteria of parathyroid malignancy (1). Unfortunately, the classic histopathologic criteria alone are unreliable and the diagnosis of parathyroid carcinoma is often established by recurrence and clinical presentation (3).

Many clinical PET studies have reported that [18F]FDG, a positron-emitting analog of 2-deoxyglucose, accumulates in many types of malignant neoplasms, including brain tumors, lymphoma, bronchogenic carcinoma, breast carcinoma, musculoskeletal sarcomas and colorectal carcinoma (13–15). Significant accumulation of FDG by parathyroid carcinoma, as demonstrated by PET imaging, has previously been reported (16).

Our present report also demonstrates the avidity of this parathyroid carcinoma for FDG and demonstrates the superiority of FDG-PET compared to 99mTc-sestamibi SPECT and MRI in this case.

Technetium-99m-sestamibi has been shown to be a useful parathyroid imaging agent (17). Several reports have demonstrated the ability of 99mTc-sestamibi to image parathyroid adenomas successfully in patients with primary hyperparathyroidism. It has been observed that parathyroid adenomas tend to demonstrate preferential retention of sestamibi over time, compared to normal thyroid tissue, which typically demonstrates progressive reduction in tracer concentration over time (18). A case of significant sestamibi accumulation by a parathyroid carcinoma has also been reported (5).

Our case, however, is contradictory to this previous report, and failed to demonstrate any appreciable uptake of 99mTc-sestamibi by our patient’s parathyroid carcinoma. In addition, the MRI findings were misleading and failed to demonstrate the entire extent of the parathyroid carcinoma in this patient. With the additional information provided by the FDG-PET study in this patient, a resection of the bulk of the parathyroid carcinoma was performed successfully.

CONCLUSION

FDG-PET provided the most accurate information related to the location and extent of parathyroid carcinoma, compared to the misleading findings of CT, MRI and 99mTc-sestamibi. This case suggests potential benefits provided by preoperative imaging with FDG-PET in parathyroid carcinoma, including replacement of several noncontributory localization procedures and assistance in preoperative surgical planning.

REFERENCES