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Parathyroid carcinoma and secondary hyperparathyroidism: A case report

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Abstract

Introduction: Parathyroid carcinoma is a rare form of endocrine malignancy which occurs most commonly in patients with primary hyperparathyroidism. Here we report an unusual case of parathyroid carcinoma developing on secondary hyperparathyroidism in a 53-year-old man, with a chronic renal insufficiency on hemodialysis, operated 10 years ago of subtotal parathyroidectomy for secondary hyperparathyroidism with favorable outcome, was hospitalized for bone pain with hypercalcaemia and elevated serum Para Thyroid Hormone (PTH). Explorations showed hyperplasia of lower right parathyroid gland. Surgical exploration revealed polylobulated mass of 4 cm adherent firmly to the oesophagus and to recurrent nerve. Lower right parathyroidectomy was so performed. Histological examination concluded to parathyroid carcinoma. Patient normalized calcium and PTH levels. No recurrence was observed after 36 months of follow up.

Keywords: Parathyroid carcinoma; Hyperparathyroidism; Surgery

Introduction

Parathyroid carcinoma is a rare endocrine malignant tumor accounting for less than 1% of all cases of primary hyperparathyroidism [1]. Preoperative diagnosis and management stills a real challenge.

Here we report an unusual case of parathyroid carcinoma developing on secondary hyperparathyroidism and we identify diagnostic and management challenges of parathyroid carcinoma.

Case report

We report the case of a 53-year-old man with a history of hypertension, chronic renal failure since 22 years in hemodialysis. He was operated 10 years ago of subtotal Para thyroidectomy, keeping the lower right parathyroid gland, for secondary hyperparathyroidism. Post-operative Para Thyroid Hormone (PTH) level was clearly reduced from 1579 pg/ml to 370 pg/ml with normal calcemia. Histological examination has revealed hyperplasia of all parathyroid glands.

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He reported recurrence of bone pain. Physical examination does not reveal any mass palpable in the neck.

Laboratory findings showed serum calcium levels of 2,5 mmol/l and PTH levels of 1160 pg/ml. Cervical Ultrasonography revealed hypoechoic right parathyroid mass of 24 mm heterogeneous and hypervascularized. A 99mTc sestamibi parathyroid scintigraphy showed hyperplasia of lower right parathyroid gland.

Intraoperatively, the mass was polylobulated of 4 cm adherent firmly to the oesophagus and to recurrent nerve. Lower right parathyroidectomy was performed.

Histologic examination concluded to parathyroid carcinoma. Macroscopically, the tumor measured 4cm. On section, it was multinodular with brown color and haemorrhagic features. Histology revealed large cells with a pseudonodular appearance, separated by dense fibrous trabeculae with a thick fibrous capsule invaded by the tumor. Images of vascular emboli were observed on the periphery of the tumor (Figure 1).

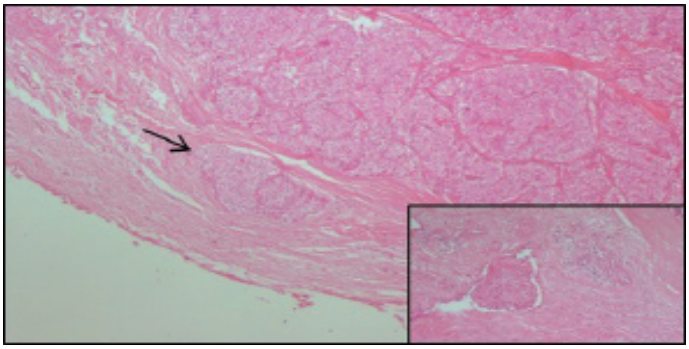

Figure 1: Parathyroid carcinoma showing multinodular growth and capsular invasion (arrow). Inset: Vascular emboli on the periphery of the tumour

Calcium and PTH levels were within the normal range at three months after surgery (calcium: 2, 5 mmol/l, PTH 25,8 pg/ml). No recurrence was observed after 36 months of follow up (the last PTH level: 18pg/ml).

Discussion

To date, the etiology of parathyroid cancer is still unknown [1]. However, some causes are associated with an increased risk of parathyroid malignancy, such as previous external radiation of the neck, hereditary forms of hyperparathyroidism [2]. Parathyroid carcinoma described in patients with secondary and tertiary hyperparathyroidism caused by chronic renal failure, such as the case of our patient are rare.

Parathyroid carcinomas are generally hyperfuctioning. Therefore, symptoms of severe hypercalcemia often dominate clinical presentation such as weight loss, weakness, arthralgia, skeletal involvement [1]. Less than 10% of patients are asymptomatic, related with nonfunctioning tumor.

On physical examination, a palpable neck mass can be present in 40 to 70% of cases which is highly suggestive for malignant tumor [1]. The presence of recurrent laryngeal nerve palsy is rare and indicates invasive disease [1]. Frequently, the diagnosis can only be made at the time of surgery or at final pathological report such as the case of our patient.

Laboratory findings include markedly increased PTH levels (5 to 15 times above normal levels) and serum calcium concentration which is greater than 3,5mmol/l [1,3]. In fact, especially high serum PTH and calcium were predictive for the occurrence of parathyroid carcinoma [4]. However, calcium and PTH levels are within normal range in patients presenting non functioning cancers.

Imaging is essential to confirm diagnosis and localize parathyroid lesions. Ultrasonography is usually the first modality used. Certain sonographic features may be suggestive of malignancy like local infiltration, calcifications, suspicious vascularity, presence of thick capsule and suspect cervical lymph node [1]. The usual technique for detecting parathyroid disease is scintigraphy with ^{99m}TC-sestamibi. However, small lesions and malignant parathyroid tumors may remain undetected [5]. 99m Technicium Sestamibi scanning with Single-Photon Emission Computed Tomography (SPECT), can detect ectopic parathyroid glands and may differentiate benign from malignant parathyroid lesions [1].

In cases where malignancy is suspected, or recurrence of hyperparathyroidism, computed tomography may localize the lesion and reveal invasion of surrounding structures and lymph nodes [1]. Magnetic resonance imaging is useful in identification of metastases and recurrent disease. In our case, it was more prudent to practice an MRI before surgery.

Histological distinction between parathyroid adenoma and parathyroid carcinoma represents a diagnostic challenge. The criteria of malignancy are: Presence of lobular architecture separated by fibrous trabeculae, cytonuclear atypias, mitotic figures, capsular or vascular invasion and the presence of metastases. However, these criteria are inconstantly observed and none of them is pathognomonic [1,6].

The main differential diagnosis is parathyroid adenoma. Parathyroid hyperplasia, anaplastic thyroid carcinoma and metastasis of renal cell carcinoma must also be excluded.

The only curative treatment of parathyroid cancer is surgery with “en bloc” resection including the tumor, the ipsilateral thyroid lobectomy [7] and for certain authors tracheal skeletonization and excision of any adherent muscle [8]. However, the diagnosis is commonly made intra or postoperatively. Therefore, 78,6% of parathyroid cancers were resected by simple Para thyroidectomy [8]. The sacrifice of recurrent laryngeal nerve is not recommended unless it is invaded by the tumor [8].

The incidence of regional lymph node involvement at initial diagnosis is variable ranging between 6,5 and 32,1% [3,9]. Therefore, therapeutic neck dissection is recommended if there is evidence of lymph node involvement [8]. Radiotherapy and chemotherapy is generally ineffective for parathyroid carcinoma [1].

The recurrence rate of parathyroid carcinoma is variable from 30 to 60% in most series [1,6,8]. Recurrence occurs 2 to 5 years after initial surgery in loco regional sites [1].

These recurrences are generally revealed by elevated calcium and PTH levels. Therefore, patients should be followed closely at intervals of 3 months using serum calcium and PTH levels [1,10] to detect an eventual recurrence. The best treatment of recurrent disease is surgical resection.

The most important factor affecting prognosis is the en bloc resection of the primary tumor [1,3,8]. The overall 10-year survival rate is varies from 49 to 77% [8].

Conclusion

Parathyroid carcinoma is a rare malignant tumor which still presents challenges in diagnosis and treatment. Surgery with en-block resection of the tumor and involved surrounding structures is the principal modality of treatment. The prognosis is variable due to frequent recurrences.

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