

Case Report

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Undescended Parathyroid Gland Mimicking Salivary Gland Uptake

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Abstract

Undescended glands are a rare cause of primary and secondary hyperparathyroidism.

Patient and method: We report the case of a 35 year-old female with a 14 year-old history of chronic renal failure who presented a severe and persistent secondary hyperparathyroidism despite subtotal parathyroidectomy and removal of mediastinal ectopic parathyroid. The patient remained symptomatic with high iPTH levels at 1700 pg/ml.

Result: A 99mTc-Sestamibi parathyroid scintigraphy was performed. Cervical SPECT/CT localized the pathological uptake in the fused images behind the right submandibular salivary gland, in the right jugulo-carotid bifurcation. Minimally invasive surgical intervention was successfully performed. The postoperative iPTH level was within the normal range.

Keywords: Undescended parathyroid gland; SPECT/CT; Secondary hyperparathyroidism

Introduction

Undescended glands are a rare cause of primary and secondary hyperparathyroidism (HPT). They are more common, in case of parathyroid reoperations for recurrent or persistent HPT after a failed initial cervical exploration. Tc-99m MIBI SPECT is an imaging approach with accurate preoperative localization recommended by many authors in patients with persistent or recurrent secondary hyperparathyroidism to permit the surgeon to minimize the surgical trauma for those patients.

Case Report

We report the clinical case of a 35 year-old female with a 14 year-old history of chronic renal failure undergoing hemodialysis who presented a severe and persistent secondary hyperparathyroidism despite a successive surgical management. She was referred to the Department of Nuclear Medicine in July 2015 for a parathyroid scintigraphy.

With regard to her clinical history, our patient had undergone surgery on 2 occasions. The first intervention was in 2004. A subtotal parathyroidectomy was performed. Three glands were removed and the histological examination confirmed the parathyroid gland hyperplasia. Postoperative intact parathyroid hormone (iPTH) serum level remained high (400 pg/ml). A dual phase parathyroid scintigraphy (the SPECT was not available in our country) were requested and had reveal a mediastinal parathyroid. The ectopic gland

was removed by an opensurgery in 2008. The patient remained symptomatic with high iPTH levels at 1700 pg/ml (range of normality 15-65 pg/ml).

In January 2015, the patient remained symptomatic with aggravation of her renal failure, very high iPTH serum levels (iPTH serum level between 1465-1700 pg/ml; range of normality 15-65 pg/ml) with normal serum calcium levels (serum calcium values between 2, 21 and 2, 4 mmol/l; range of normality 2, 15-2, 50 mmol/l). She also presented multiples brown tumor, the most important were in the left maxillary bone and of the right iliac bone. She was lost to follow-up during 6 years. The patient was referred for parathyroid scintigraphy; planar face images and right oblique incidence were performed completed with a single photon emission tomography coupled to a computed tomography (SPECT/CT) after intravenous injection of 592 MBq (16 mCi) of 99mTc-Sestamibi. The examination was carried with a parallel collimators focused on the neck, including salivary glands in the field of view, and the thorax. Planar subtraction images showed a moderate pathological uptake at the height of the right submandibular salivary gland. There was no cervical or mediastinal fixation (Figure 1). Cervical SPECT/CT localized the pathological uptake in the fused images behind the right submandibular salivary gland, in the right jugulo-carotid bifurcation (Figures 2 and 3). With these results, another minimally invasive surgical intervention was performed, localizing a 2 cm hyperplastic parathyroid gland in the right carotid sheath. The post-operative iPTH levels was within the normal range (15 pg/ml) with normal serum calcium level (2, 4 mmol/l).

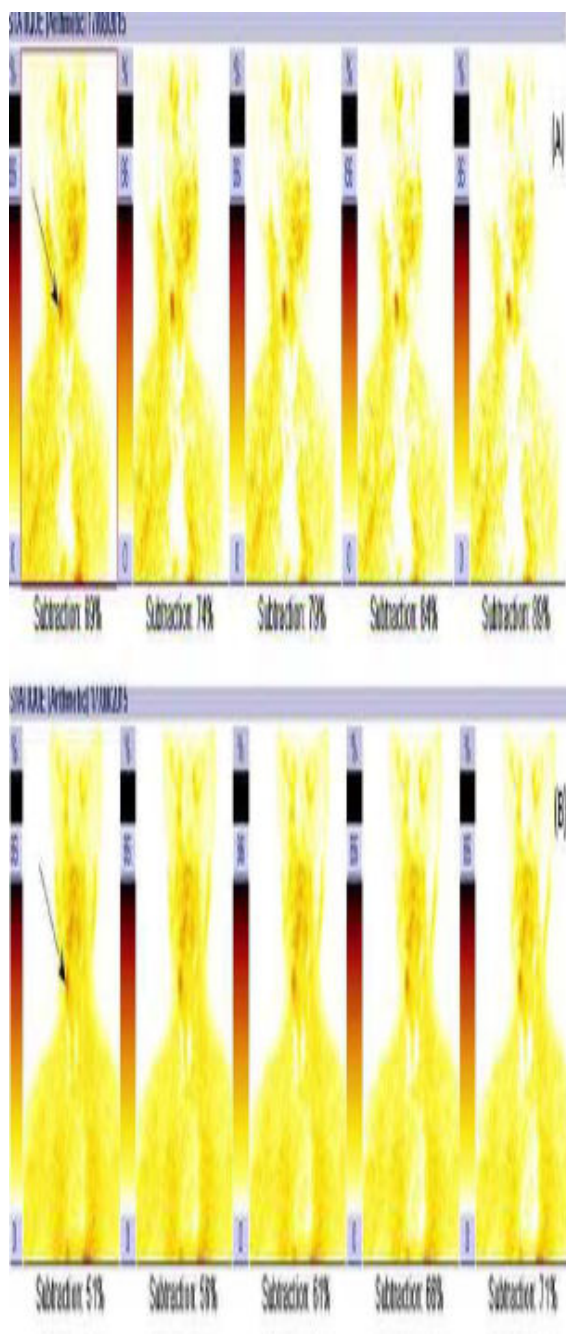


Figure 1: Planar subtraction imaging showing sestaMIBI uptake behind right submandibular salivary gland (A) right anterior oblique incidence subtraction images (B) antero-posterior incidence subtraction images.

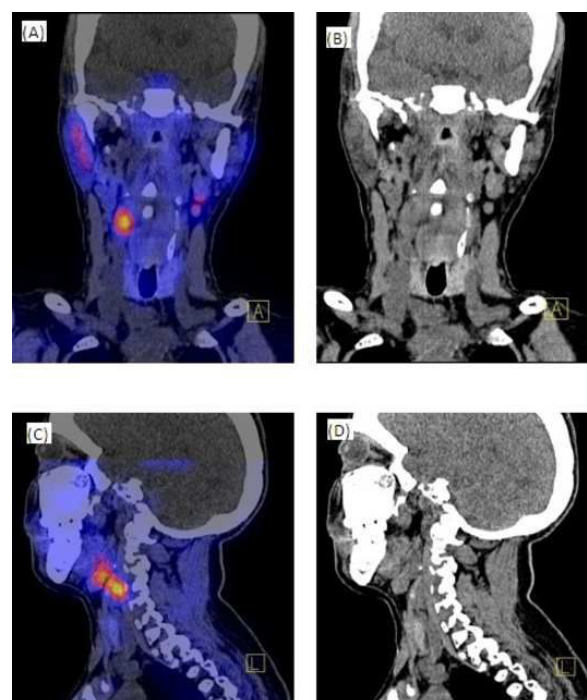


Figure 2: SPECT/CT of the cervical region showing parathyroid gland in the right carotid sheath.

Discussion

A major cause of persistent hyperparathyroidism is failure to detect an ectopic parathyroid adenoma. The frequency of ectopic glands in the secondary hyperparathyroidism ranges from 12% to 19% in the surgical series, even higher in autopsy series reaching 42% [1]. Ectopic parathyroid glands may be located anywhere from the base of the tongue to the mediastinum. The superior parathyroid glands derive from the fourth and the inferior glands from the third branchial pouch, the latter closely associated with the thymus. Therefore the inferior parathyroid glands have a longer route of embryologic descent and its final location at or around the lower pole of the thyroid lobe are variable [2,3]. In general case, the most frequent locations are in the thymus or in the anterosuperior mediastinum (35%), in the thyrothymic ligament (12%) and the intrathyroidal gland (18%). Other unusual ectopic locations have been also described in the literature; in the aortopulmonary window, the pericardium, the pyriform sinus, the vagus nerve sheath and the posterior cervical triangle [4]. An undescended inferior parathyroid gland is a rare location (<1%) found most commonly in the carotid sheath at the level of the carotid bifurcation. It is particularly difficult to identify preoperatively, leading often to surgical failure [5,6]. In our case, undescended inferior right parathyroid gland was found in the right carotid sheath. Parathyroid glands can be explored with multiple imaging modalities including ultrasonography, thin section CT and MRI. Subtraction 99mTc-Sestamibi scintigraphy has proved to be an accurate method for locating parathyroid hyperplasia especially in ectopic site. Its sensitivity reaches 83% and it's even higher with the addition of single photon emission computed tomography coupled with CT (SPECT/CT) reaching 100% [7-9]. SPECT/CT has been proven to increase the

sensitivity and the accuracy of the location especially in case of parathyroid reoperations for recurrent or persistent HPT after a failed initial cervical exploration [9]. In the interpretation of the studies, as in our case, it should be taken into account that in presence of an undescended ectopic parathyroid gland, high cervical region must be included in the field of view for planar. The addition of SPECT/CT increases the diagnostic safety and it increases trust of surgeons in radionuclide imaging.

Conclusions

Current development of noninvasive imaging techniques such as SPECT/CT has improved the preoperative localization of UPAs and increased the surgical successful rate. The field of view must include the upper cervical region to avoid missing UPAs.

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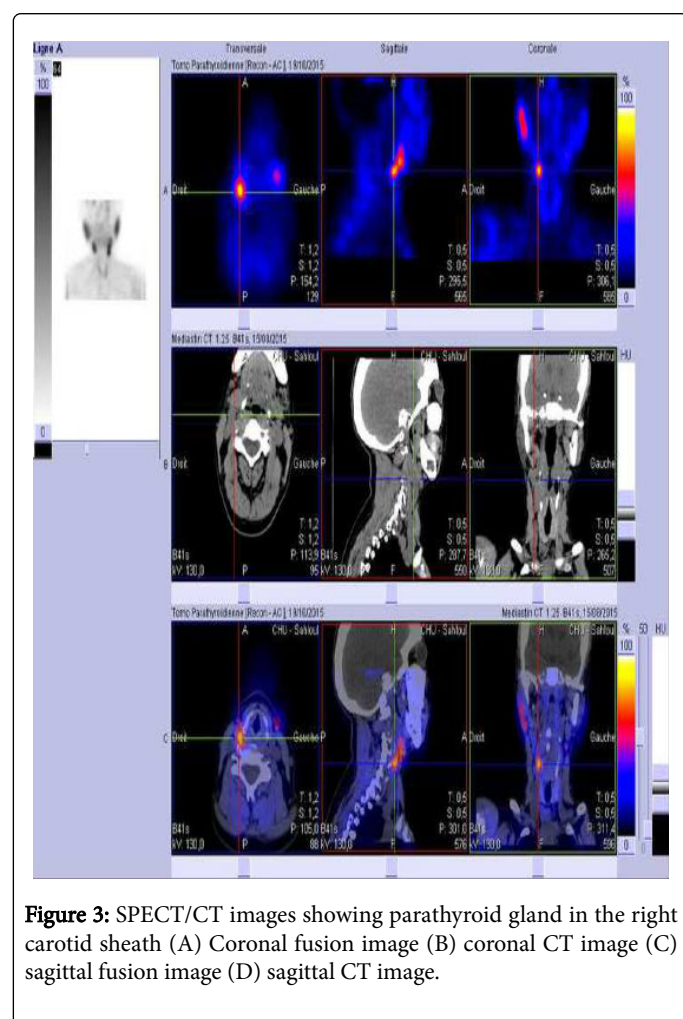


Figure 3: SPECT/CT images showing parathyroid gland in the right carotid sheath (A) Coronal fusion image (B) coronal CT image (C) sagittal fusion image (D) sagittal CT image.