Clinical Case Reports

Open Access

CASE REPORT

Concomitant occurrence of primary hyperparathyroidism (PHPT) due to mediastinal parathyroid adenoma and sublingual thyroid gland: the role of parathyroid technetium-99m-MIBI scintigraphy

Sara Choukry[1], Jaafar Benouhoud[1], Ghofrane Cherkaoui Salhi[1], Sara Taleb[1], Amal Guensi[2] & Karim Choukry[3]

[1]Nuclear Medicine Department, CHU Ibn Rochd, Casablanca, Morocco
[2]Faculty of Medicine and Pharmacy of Casablanca, Nuclear Medicine Department, CHU Ibn Rochd, Casablanca, Morocco
[3]Department of Otorhinolaryngology, 20 August Hospital, Casablanca, Morocco

Correspondence
Sara Choukry, CHU Ibn Rochd, 1 Rue des hopitaux, Casablanca, Morocco. Tel: 00212 522 48 40 40; Fax: 002125976222;
E-mail: direction@chu.ma

Funding Information
No sources of funding were declared for this study.

Received: 30 April 2016; Revised: 24 August 2017; Accepted: 26 August 2017

***Clinical Case Reports* 2017; 5(11): 1882–1886**

doi: 10.1002/ccr3.1204

Key Clinical Message

The concomitant appearance of a sublingual thyroid and primary hyperparathyroidism due to parathyroid mediastinal adenoma is not common. This co-occurrence can lead to a misdiagnosis by morphological imaging methods alone. This case emphasizes the role of 99mTc-MIBI scintigraphy in the detection of parathyroid ectopic adenoma in a patient with an ectopic thyroid gland. This more accurate location of parathyroid scintigraphy is of great benefit to the surgeon for surgical excision.

Keywords

Ectopic parathyroid adenoma, ectopic thyroid, hyperparathyroidism, mediastinal parathyroid adenoma, parathyroid scintigraphy.

Introduction

Ectopic thyroid gland (ETG) is defined as thyroid tissue not located in its normal region, in the base of the neck between the second and fourth tracheal cartilage. This is a rare condition caused by an abnormal embryonic development. The thyroidal tissue can stop migrating at lingual or sublingual level [1].

Primary hyperparathyroidism (PHPT) is a common endocrinopathy. It is more often caused by a solitary adenoma (90%) than other pathologies such as nodular hyperplasia (6%), double adenomas (2–5%), and parathyroid carcinoma (<1%) [2]. In about 6–16% of PHPT, parathyroid adenomas may be ectopic, which makes treatment by surgical resection a very challenging task.

The treatment of PHPT is surgical, and it was traditionally based on the excision of the adenoma currently using minimally invasive approaches (depending on the location).

The concomitant occurrence of sublingual thyroid and primary hyperparathyroidism (PHPT) due to mediastinal parathyroid adenoma is uncommon.

Indeed, to date, no such co-occurrence has been described in the literature. This case emphasizes the role of 99mTc-sestamibi parathyroid scintigraphy in localizing accurately an ectopic parathyroid adenoma in a context of an abnormal migration of the thyroid bud and hence guiding its surgical treatment.

Case Presentation

We report herein the case of a 45-year-old woman, with a history of sublingual thyroid gland discovered accidentally in a craniofacial CT scan performed for a nasolacrimal duct obstruction suspicion.

The anamnesis reported dysphagia, joint pains, asthenia, and muscle cramps.

The clinical examination noted a raucous voice, a left cervical adenopathy, and an empty thyroid lodge, suspecting an ectopic thyroid gland.

The CT scan describes a sublingual mass well delimited with homogeneous enhancement of contrast.

Cervical ultrasonography showed a sublingual ectopic thyroid gland without any hypertrophic parathyroid gland. It also showed left cervical adenopathy (group 2a).

The thyroid scintigraphy acquired in static mode 20 min after intravenous injection of 99mTc of pertechnetate confirmed the thyroid nature of the sublingual tissue (Figs 1, 2 and 3).

The thyroid function was normal (TSH:2.3 UI/L T3: 1.3 nmol/L and T4: 15 pmol/L).

The diagnosis of primary hyperparathyroidism has been confirmed biologically by hypercalcemia (serum calcium value: 110 mg/L) and hypercalciuria (456 mg/24 h) with elevated PTH levels (220 pg/mL).

Parathyroid scintigraphy was performed in the nuclear medicine department of IBN ROCHD University Hospital.

This functional imaging was acquired using both single-isotope dual-phase technique (99mTc-MIBI) and dual-isotope subtraction imaging technique (99mTc-pertechnetate and 99mTc-MIBI radiopharmaceuticals), and it depicted a focal uptake in the superior mediastinum with a delayed washout, leading to the conclusion of an ectopic mediastinal parathyroid adenoma. On the other hand, no other parathyroid tissue was noticeable by subtraction technique in the sublingual area (Figs 4–6).

A thoracic CT scanner was done for an accurate location of the adenoma. Afterward, the patient has undergone a surgical excision. The anatomopathological examination confirmed the parathyroid origin of the adenoma (cells with clear cytoplasm, without mitosis, without necrosis, without capsular invasion, and no suspect aspect of malignancy).

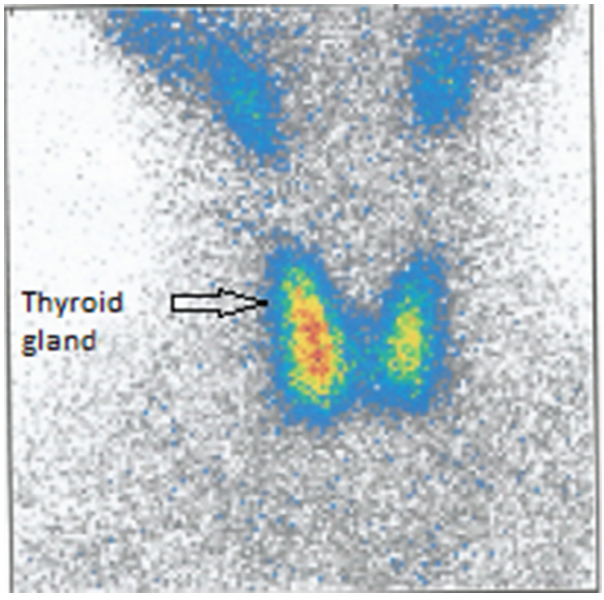

Figure 1. Thyroid scintigraphy acquired by a static mode 20 min after intravenous injection of 99m-Tc pertechnetate: normal location of thyroid gland.

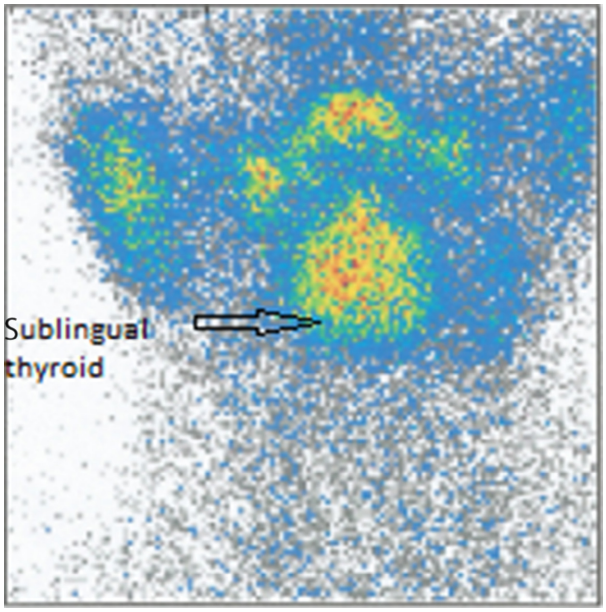

Figure 2. Thyroid scintigraphy acquired by a static mode 20 min after intravenous injection of 99m-Tc pertechnetate: high sublingual uptake.

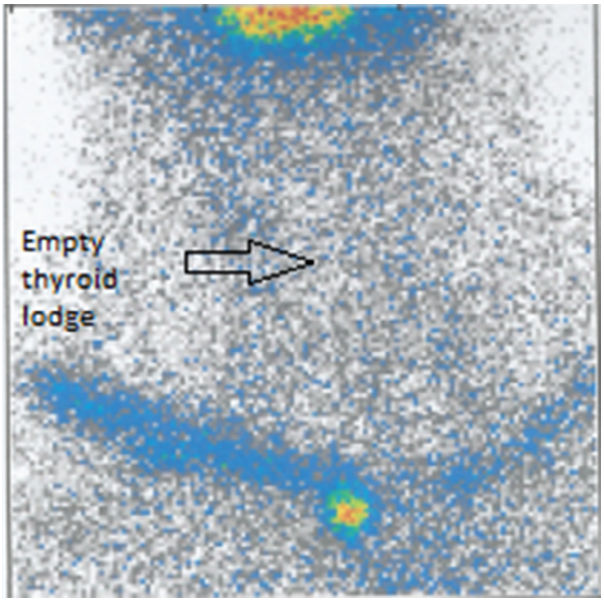

Figure 3. Thyroid scintigraphy acquired by a static mode 20 min after intravenous injection of 99m-Tc pertechnetate: Empty thyroid lodge.

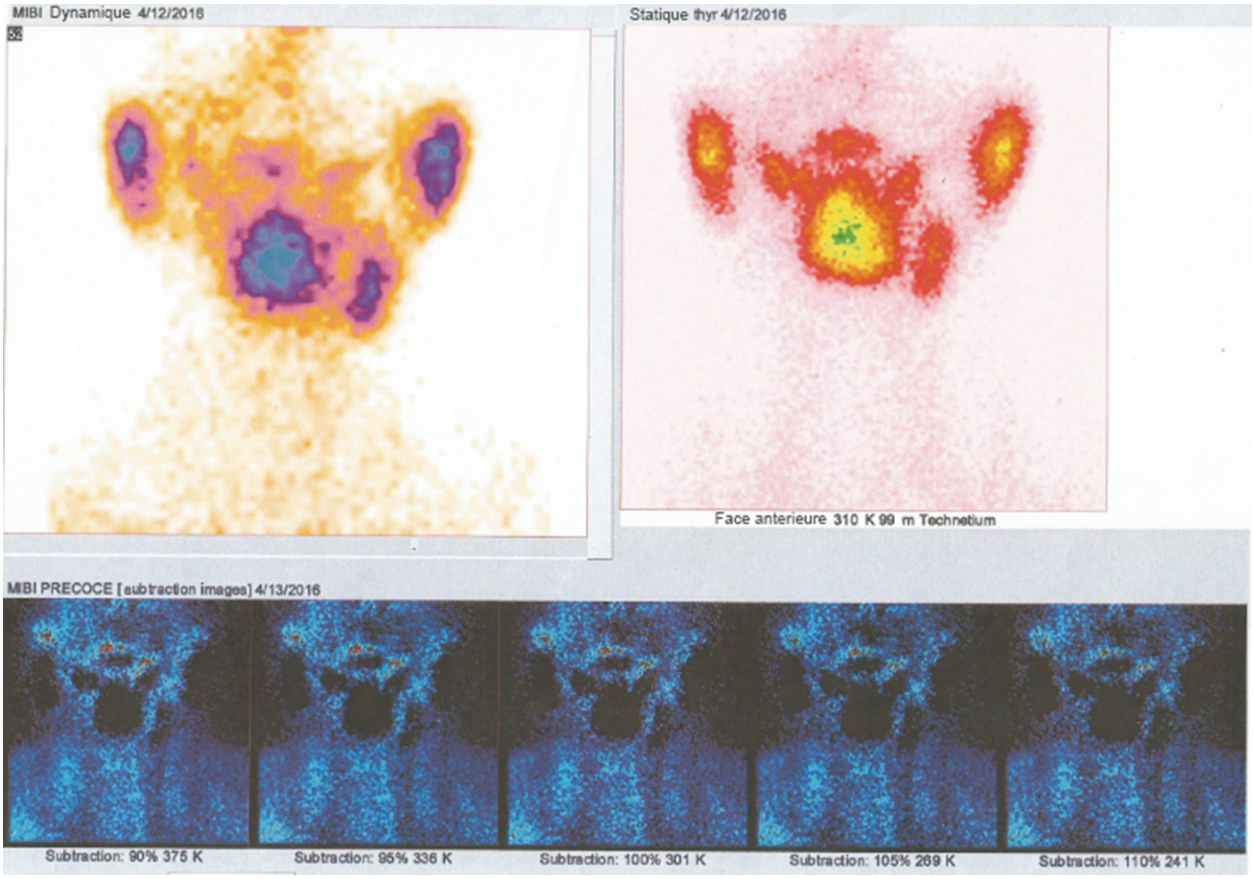

Figure 4. Parathyroid scintigraphy with dual-isotope subtraction imaging technique (99mTc-pertechnetate and 99mTc-MIBI radiopharmaceuticals): High uptake in the salivary gland without any parathyroid tissue.

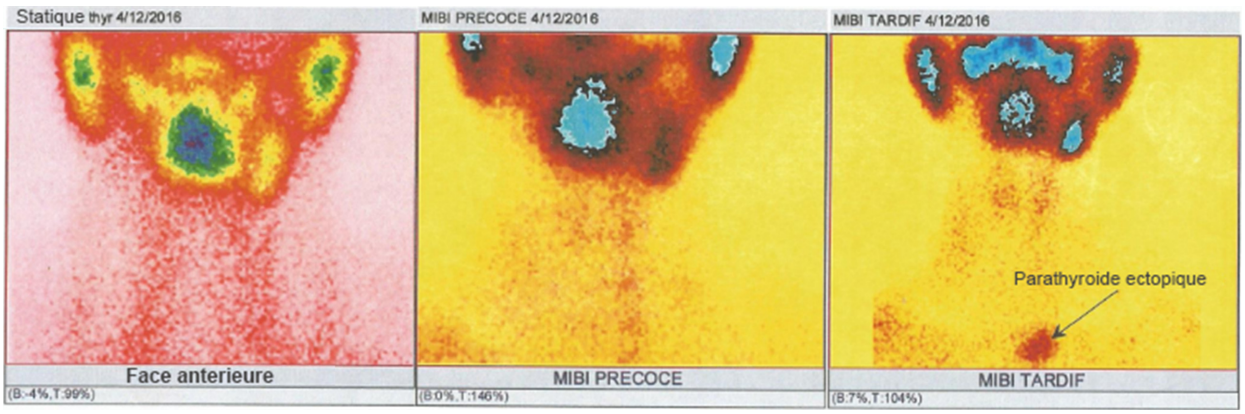

Figure 5. Parathyroid scintigraphy using dual-phase technique with static images taking the neck and the mediastinum: a focal uptake in the superior mediastinum with a delayed washout without any other parathyroid tissue.

The last checkup carried out 10 months after the surgery shows normalization of serum calcium and PTH (serum calcium value: 95 mg/L and PTH:60 ng/mL).

Discussion

Ectopic thyroid gland is an uncommon entity and its prevalence is estimated between one and three per 100,000

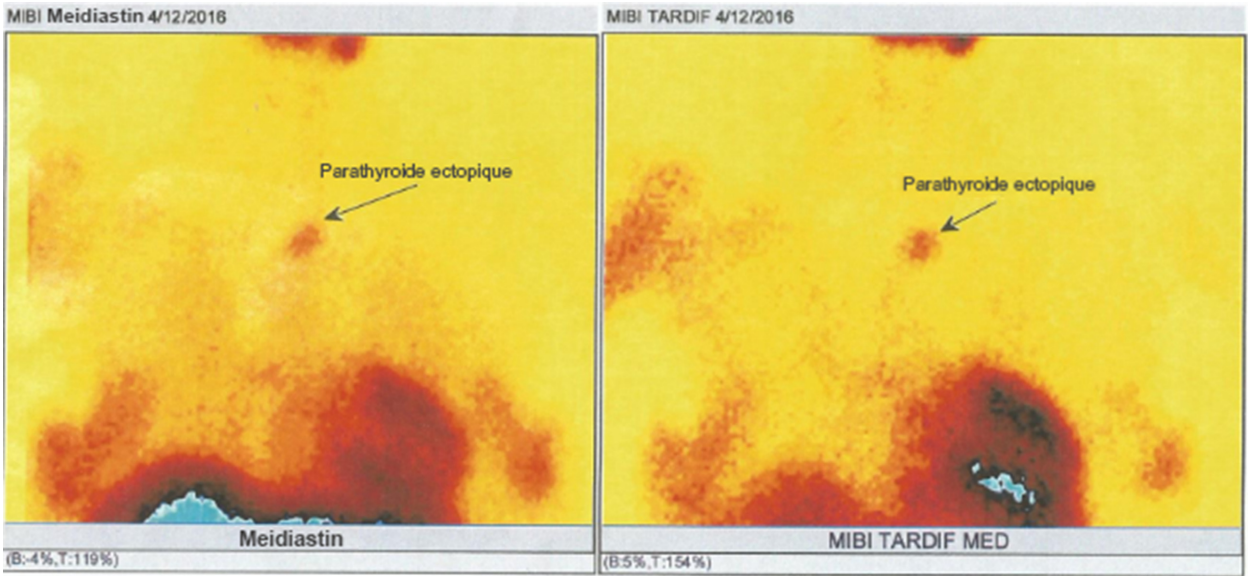

Figure 6. Parathyroid scintigraphy using dual-phase technique with static images taking the neck and the mediastinum: a focal uptake in the superior mediastinum with a delayed washout without any other parathyroid tissue.

persons and occurs in one in 4000–8000 patients with thyroid disease [1]. Most studies report that women are more likely to have an ETG than men. In Morocco, this is the second reported case of an ETG. However, due to the presence of asymptomatic cases, its prevalence remains an estimate. Lingual thyroid is the most frequent location of ETG, and the sublingual situation is less common and may be classified as suprahyoid, hyoid, or infrahyoid. The etiopathogeny of this abnormal migration is still unclear, but mutation of factors influencing the thyroid morphogenesis and differentiation may be involved [3].

Clinical manifestations usually include dysphagia, a lump sensation in the throat, or growth retardation. Indeed, this latter symptom is due to the high association of ETG with hypothyroidism [1].

Imaging evaluation includes neck ultrasonography, CT scan, and especially thyroid scintigraphy using pertechnetate 99mTc or radioactive iodine, which shows a functional thyroid tissue in an abnormal location.

In our case, even if the patient was symptomatic (dysphagia with a raucous voice), the discovery was accidental at the age of 45 years by a craniofacial CT scan (relatively belated). It was then affirmed by 99mTc-pertechnetate thyroid scintigraphy.

PHPT is a common endocrinopathy. Studies suggest that up to 1% of the population may have a hyperparathyroidism with a tendency to affect the females after the menopause [4]. Hyperparathyroidism is asymptomatic in 83–95%. Indeed, it is usually discovered during a biochemical routine checkup by detecting a hypercalcemia [5]. PHPT diagnosis is based on biochemical data. Imaging studies are only requested after the exclusion of other pathologies that may cause hypercalcemia and preferably before surgical treatment to localize and hence to plan the operative approach [6].

The treatment of PHPT is surgical today, using minimally invasive methods allowing both exploration and resection of adenomas. Currently, most surgeons prefer to be guided by the data of the MIBI-Tc 99m scintigraphy, particularly given the frequency of the glandular ectopy [2].

About 6–16% of parathyroid adenomas may be ectopic. Indeed, parathyroid adenomas can be located above the upper pole of the thyroid lobe, behind the pharynx or esophagus, in the neck, but the mediastinal location remains the most common (about 20% of hyperparathyroidism cases) which makes the surgical resection a very difficult task [2].

Preoperative imaging techniques help localizing parathyroid adenomas and planning the operative approach. Parathyroid scintigraphy using 99mTc-sestamibi is the most widely used imaging technique. 99mTc-sestamibi is taken up in thyroid and parathyroid tissue by the mitochondria. The double-phase protocol is based on the realization of two acquisitions, the first shortly after the radiotracer injection and the second one, two hours later. As the clearance of 99mTc-sestamibi is slower in parathyroid tissue (the presence of the mitochondria-rich oxyphilic cells), foci with high uptake are consistent with hyperfunctioning parathyroid tissue [2].

These acquisitions are commonly acquired in a planar mode. If available, we can use the three-dimensional images by single photon emission tomodensitometry (MIBI-SPECT) and SPECT/CT which give better

resolution. It also estimates the depth and distinguishes parathyroid adenomas from other anatomical structures.

The thyroid subtraction protocol can be used with the double-phase protocol. It uses two tracers: one is taken up in the thyroid tissue and the other in both thyroid and parathyroid glands. The images are then subtracted.

The good visualization of parathyroid adenomas in patients with a large adenoma and an increased parathyroid's hormones suggests a correlation between the weight of parathyroids and their hyperfunction. Nevertheless, the positivity of the scintigraphy MIBITc99m depends not only on the weight of the adenoma but also on the number of oxyphilic cells rich in mitochondria.

The scintigraphy of the parathyroid glands at 99mTc-MIBI in dual phase has a sensitivity of 90% (94% for glands weighing around 200 mg) [7]. Ultrasonography is highly sensitive examination especially if performed by a doctor used to this method. It is also an affordable examination compared to the scintigraphy, but the association of thyroid pathologies with PHPT reduces its accuracy (20–30% of cases) [8]. Most parathyroid experts rely on both ultrasonography and sestamibi scintigraphy (SPECT if available) for preoperative localization [9].

However, some small parathyroidal adenomas may go unnoticed in parathyroid scintigraphy because of its poor spacial resolution and the masking effect of retained radiopharmaceutical in the adjacent thyroid or submandibular glands [10].

In this case report, the concomitance of ETG and PHPT made the interpretation of morphological images a very challenging task for radiologists. Indeed, the coexistence of a parathyroid adenoma in a context of an abnormal migration of thyroid bud lowers the sensitivity and specificity of ultrasonography and CT scans, especially when the parathyroid adenoma is in an ectopic location. In this case, functional imaging using both "single-isotope dual phase" and "dual isotope subtraction imaging techniques" showed the hyperfunctioning ectopic parathyroid tissue located in the upper mediastinum. As a result, incision placement is determined, and minimally invasive surgery might be considered.

Conclusion

This case report depicts the first case with a concomitant ETG and an ectopic parathyroid adenoma. Among localization studies undergone, parathyroid scintigraphy allowed to locate the adenoma and hence to guide the surgical treatment.

Authorship

SC: was the principal author of the article, who realized the thyroid and parathyroid scintigraphy for the patient, wrote and submitted the article after making all the corrections requested by the reviewers. JB: contributed to the interpretation of the thyroid and parathyroid scintigraphy and the follow-up of the patient. GCS: participated in the writing of the article mainly through the bibliographic search. ST: participated in the correction of the article. AG: followed closely all the stages of the drafting of the article. KC: participated in the bibliographic research and postoperative follow-up of the patient.

Conflict of Interest

None declared.

References

1. Yoon, J. S., K. C. Won, I. H. Cho, J. T. Lee, and H. W. Lee. 2007. Clinical characteristics of ectopic thyroid in Korea. Thyroid 17:1117–1121.
2. Eslamy, H. K., and H. A. Ziessman. 2008. Parathyroid scintigraphy in patients with primary hyperparathyroidism: 99mTc sestamibi SPECT and SPECT/CT. Radiographics 28:1461–1476.
3. Abramowicz, M. J., G. Vassart, and S. Refetoff. 1997. Probing the cause of thyroid dysgenesis. Thyroid 7:325–326.
4. Bandeira, F., L. Griz, N. Chaves, N. C. Carvalho, L. M. Borges, et al. 2013. Diagnosis and management of primary hyperparathyroidism–a scientific statement from the Department of Bone Metabolism, the Brazilian Society for Endocrinology and Metabolism. Arq. Bras. Endocrinol. Metabol. 57:406–424.
5. El Oueriachi, F., A. Arsalane, and H. el Kabiri. 2015. Uncommon ectopic parathyroid adenoma. Arch. Bronconeumol. 51:301–302.
6. Yip, L., D. A. Pryma, J. H. Yim, M. A. Virji, S. E. Carty, and J. B. Ogilvie. 2008. Can a lightbulb sestamibi SPECT accurately predict single-gland disease in sporadic primary hyperparathyroidism? World J. Surg. 32:784–792; discussion 93–94.
7. Giron, J., E. Ouhayoun, M. Dahan, J. Berjaud, J. P. Esquerre, J. P. Senac, et al. 1996. Imaging of hyperparathyroidism: US, CT, MRI and MIBI scintigraphy. Eur. J. Radiol. 21:167–173.
8. Bentrem, D. J., P. Angelos, M. S. Talamonti, and R. Nayar. 2002. Is preoperative investigation of the thyroid justified in patients undergoing parathyroidectomy for hyperparathyroidism? Thyroid 12:1109–1112.
9. Udelsman, R., G. Akerstrom, C. Biagini, Q. Y. Duh, P. Miccoli, B. Niederle, et al. 2014. The surgical management of asymptomatic primary hyperparathyroidism: proceedings of the Fourth International Workshop. J. Clin. Endocrinol. Metabol. 99:3595–3606.
10. Galvin, L., J. D. Oldan, M. Bahl, J. D. Eastwood, J. A. Sosa, and J. K. Hoang. 2016. Parathyroid 4D CT and scintigraphy: what factors contribute to missed parathyroid lesions? Otolaryngol. Head Neck Surg. 154:847–853.