



Contents lists available at ScienceDirect

International Journal of Surgery Case Reports

journal homepage: www.casereports.com

Parathyroid lipoadenoma: The particularities of a rare entity a case report and review of the literature

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ARTICLE INFO

Article history:

Received 14 December 2020
Received in revised form 23 January 2021
Accepted 23 January 2021
Available online 28 January 2021

Keywords:

Parathyroid
Lipoadenoma
Primary hyperthyroidism
Case report
Surgery

ABSTRACT

Parathyroid lipoadenoma is a rare and unusual cause of primary hyperparathyroidism. The clinical presentation usually resembles other causes of primary hyperparathyroidism and the imaging is not always contributory considering its location. However, the histologic criteria are specific.

We present a case that supplements and supports the rare literature data concerning the clinical and therapeutic aspects of parathyroid lipoadenoma.

The case is about a 73 years old female with a right inferior parathyroid lipoadenoma that caused biological primary hyperparathyroidism. Initially followed and treated in Rheumatology department for hypercalcemia and osteoporosis, she was sent to our structure to diagnose and possibly treat the causal etiology. After non-contributory clinical examination and ultrasound imaging, the tumor was diagnosed in the cervical CT scan. The patient underwent successful surgical removal of the lipoadenoma, confirmed postoperatively on histological analysis. The follow up showed rapid normalization of the parathormone level.

Even if it's a rare condition, the diagnosis of lipoadenoma should always be considered in front of primary hyperparathyroidism with a parathyroid lesion.

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1. Introduction

Parathyroid lipoadenoma is a rare and unusual cause of primary hyperparathyroidism, described as a single parathyroid adenoma with more than 50% fat on histologic examination, primary hyperparathyroidism and postoperative resolution of hypercalcemia [1].

Not more than few cases were reported on the literature, only diagnosed after surgical resection. The clinical presentation and biological features resemble the habitual forms of primary hyperparathyroidism [2–4]. Its microscopic aspect mimicking normal parathyroid tissue and difficult location to observe on imaging makes the diagnosis difficult [5].

We report a case treated in our Head and Neck surgery department of the 20 August 1953 Hospital, that supplements and supports the rare literature data concerning the clinical and therapeutic aspects of parathyroid lipoadenoma.

2. Presentation of the case

We report the case of a 73 years old female with a medical history of breast cancer surgically treated 4 months ago by a mastectomy and radio-chemotherapy. The patient was diagnosed with

hypercalcemia and osteoporosis, followed and treated since 2013 in the Rheumatology department. She did not present any signs of kidney stones, fractures or bone pain.

The first line explorations were skeletal scintigraphy that didn't show any hyperfixation area that could be related to a secondary bony localisation while laboratory investigations found a biological hyperparathyroidism up to $17.49 \mu\text{mol/l}$ with normalized calcemia rate at 2.48 mmol/l and calciuria at 201 mg/24 h , in association to normal renal function tests and a mildly decreased glomerular filtration rate (GFR) at 88.75 mL/min .

The patient was referred to our department for further evaluation. The clinical cervical examination didn't find any palpable mass with no other abnormality.

The cervical ultrasound (US) showed moderate goiter with infracentimetric nodules classified as EU TIRADS II with no visible parathyroid lesion or mass, while the cervical computed tomography (CT) scan described a $16 \times 12 \text{ mm}$ soft-tissue mass lesion related to a parathyroid nodule in contact with the lower pole of the right thyroid lobe (Fig. 1).

The parathyroid gland imaging by $[^{99m}\text{Tc}](\text{technetium})$ -sestamibi scintigraphy showed a discrete area of high uptake under the thyroid right lobe fixing electively the MIBI-Tc99m compatible with a parathyroid origin (Fig. 2).

The patient was thus, programmed for a total right inferior parathyroidectomy. The surgery was performed under general anaesthesia, by a senior resident with 4 years of specialised train-

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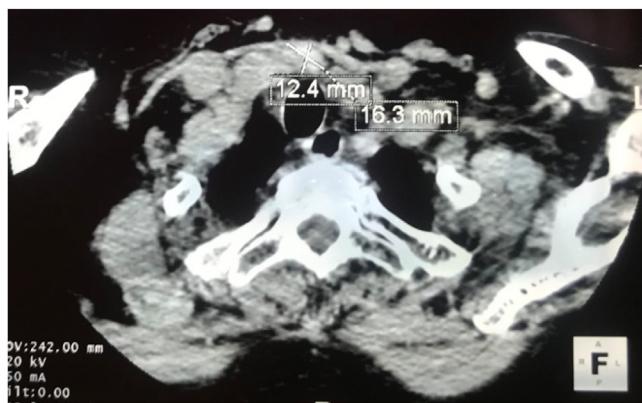


Fig. 1. Cervical axial CT scan showing the parathyroid lesion.

ing. The surgical exploration found a yellow, lobulated, soft-tissue mass, measuring 15 mm, enlarging the right inferior parathyroid gland. The procedure consisted on a complete monobloc excision with a total respect of the recurrent laryngeal nerve.

The histologic examination of the operatory piece concluded to a parathyroid lipoadenoma with parathyroid epithelial cells in a background of abundant fat tissue. No malignancy signs were

observed such as atypia, multiple mitosis or capsular and vascular invasion (Figs. 3 and 4).

Postoperatively, we reported a normalization of the parathormon at day 5. The patient was discharged after a hospital stay of 5 days with no notable incident. The follow up until 12 months, showed a total resolution of the symptomatology reported by the patient.

This case has been reported in line with the SCARE 2020 criteria [6].

3. Discussion

Parathyroid adenoma is the most frequent cause of primary hyperparathyroidism with a rate of 85%. Parathyroid lipoadenoma described for the first time in 1962 [3], is its rare and uncommon variant [7], with a reported incidence from 0,5 to 1,6% [8,9]. No predilection related to sex was found on the literature, with a sex ratio up to 1, and it affects a large range of ages, from 41 to 92 years old [10,11].

The World Health Organization defines parathyroid lipoadenoma as a “hamartoma-like benign neoplasm containing both chief cells and prominent stromal elements” [12]. Indeed, the histopathology examination describes intermingling of chief and oxyphil cells with abundant adipose cells [7].

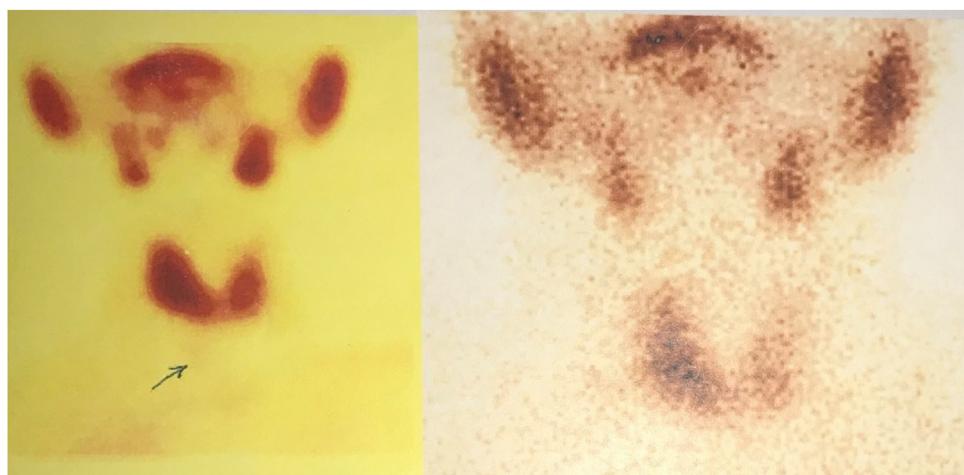


Fig. 2. Discrete lesion fixing electively the MIBI-Tc99m compatible with a parathyroid origin.

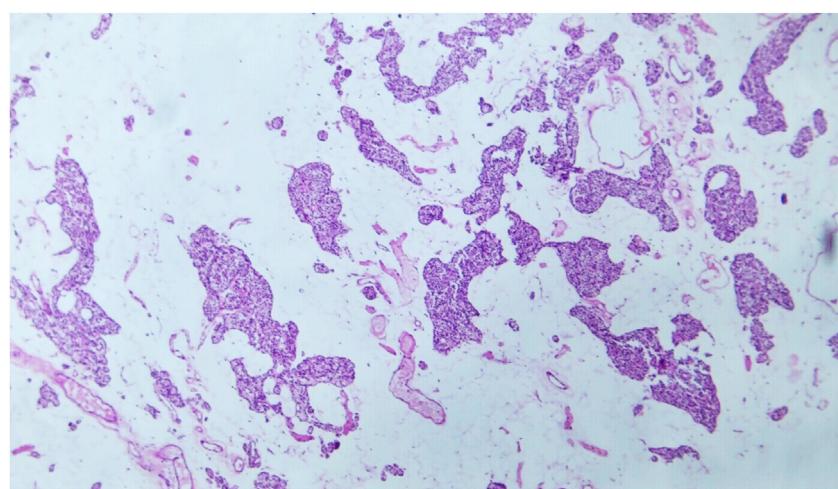


Fig. 3. Microscopic aspect ($\times 40$) of parathyroid tissue within stromal adipose tissue.

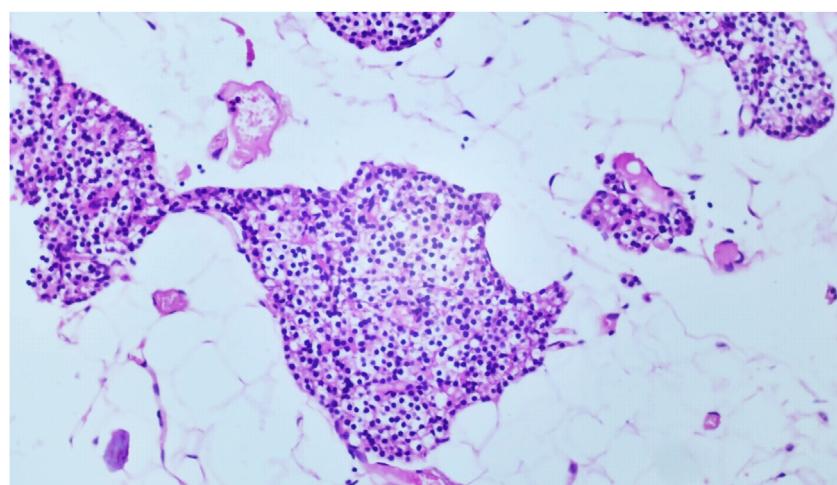


Fig. 4. Microscopic aspect ($\times 100$) of parathyroid cords of chief cells and occasional oxyphilic cells within adipose tissue.

The fat component occupies normally an average of 25% of the parathyroid gland and can be increased with age and obesity. The abundance of adipocytes in parathyroid tissue can be observed in multiple conditions such as, lipohyperplasia, lipoadenoma and infiltrative carcinomas affecting the peripheral nearby fat tissue. To differentiate between these entities and find the precise diagnosis may be difficult for the pathologist at the frozen sections. Besides, the peroperative parathormon test may be the appropriate technic to finalize the surgical procedure of lipoadenomas [13].

The clinical presentation can be asymptomatic on 28% according to previous case reports [14]. Also, contrary to parathyroid adenoma, the lipoadenoma does not belong to multiple endocrine neoplasia or family hyperparathyroidism [7].

The challenges of lipoadenomas are their difficulty to assess by imaging even though they are larger in size (up to several centimeters) because of their high fat content [7].

The US allowed identifying theses tumours in 50% of the cases while the parathyroid gland [^{99m}Tc]-sestamibi scintigraphy was efficient in 71% of the cases in a serie of 11 cases [15]. The role of scintigraphy in the preoperative evaluation of recurrent or persistent hyperparathyroidism is unanimously established and more and more used in the first line exploration of the parathyroid [16].

In our reported case, the US could not detect the parathyroid lesion that was exposed further thanks to CT scan and scintigraphy. We relate this difficulty at the US to the radiologist experience. In parallel, the surgical exploration may be difficult and unusual for surgeons not aware of this particular entity.

4. Conclusion

Parathyroid lipoadenoma is a rare cause of primary hyperparathyroidism.

But, if its histologic criteria are specific, its clinical presentation resembling other causes of primary hyperparathyroidism and its difficult detection on imaging represent true challenges. Thus, being aware of this specific entity will allow a better diagnosis and treatment.

Declaration of Competing Interest

The authors declare having no conflicts of interest for this article.

Funding

None.

Ethical approval

Ethical approval has been exempted by our institution.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

Mennouni Mohamed Amine: Study concept and writing the paper.

Oukessou Youssef: Study concept.

El Bouhmadi Khadija: Corresponding author and writing the paper.

Sami Rouadi: study concept.

Abada Reda: study concept.

Mahtar Mohamed: correction of the paper.

Registration of research studies

Not required.

Guarantor

Dr Mennouni Mohamed Amine.

Provenance and peer review

Not commissioned, externally peer-reviewed.

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