

Ectopic Parathyroid Adenoma: An Exceptional Location- Case ReportGladys Anguezomo^{1*}, Ghizlane El Mghari¹, Nawal El Ansari¹, Aboubakr Matrane²¹Department of Endocrinology, Diabetology, Metabolic Diseases and Nutrition Mohamed VI CHU, PCIM Laboratory FMPM Marrakech Morocco²Département of nuclear medicine Mohamed VI CHU Marrakech Morocco***Corresponding author**

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Article History

Received: 03.05.2018

Accepted: 08.05.2018

Published:30.05.2018

DOI:

10.36347/sjmc.2018.v06i05.010



Abstract: Parathyroid adenoma is the leading cause of primary hyperparathyroidism. Parathyroid ectopia accounts for 20% of cases. A 44-year-old woman with primary hyperparathyroidism which was revealed by pathological fractures. Methoxyisobutylisonitrile (MIBI) scintigraphy exposed a typical image of parathyroid ectopia in mediastinal position. The surgical removal confirmed the parathyroid adenoma allowing the clinical and biological improvement of the patient. Mediastinal localization represents 10% of ectopic parathyroid adenomas. The value of MIBI-Tc 99m scintigraphy in locating parathyroid abnormalities before any surgical procedure remains crucial.

Keywords: primary hyperparathyroidism; ectopic parathyroid adenoma; brown tumors; parathyroidectomy.

INTRODUCTION

Benign parathyroid adenoma, unique and sporadic, accounts for 75 to 85% of pathologies of the parathyroid gland [1]. It is most often suspected in front of severe clinical and biological primitive hyperparathyroidism, with very high levels of calcemia and normal levels of parathormonemia. This primary malignant hyperparathyroidism may be complicated by recklingchaussen fibrocystic osteitis and extra-bones calcification secondary to parathyroid hormone (PTH) excess.

In 20% of the cases, the parathyroid gland is in an ectopic situation, 13% of which are at mediastinal level. Finding the ectopia is a challenge mainly for morphological and functional explorations [2]. We report the case of a patient admitted for primary hyperparathyroidism which was revealed by multiple pathological fractures together with a decline of her general state, revealing an ectopic parathyroid adenoma located at mediastinal level with multiple scattered brown tumors; having evolved positively after surgical removal of the tumor.

CASE REPORT

44-year-old patient hospitalized for primary hyperparathyroidism revealed by pathological fractures together with a decline of her general state, history intake was without specificities; no hematopathy, no long-term drug intake, no history of radiation or gynecological disease. The onset of symptomatology seems to have been traced back to two years by progressive pelvic and lower-limb bone pain with the appearance of swelling in the left leg a year later, requiring a biopsy of the lower left tibia extremity with the histology of giant cells grade I. This was followed by fast worsening of her general state, weight loss (not quantified but considered important), physical asthenia, an absolute functional impotence, as well as spontaneous fractures of the pelvis and left femoral

diaphysis discovered one week before her admission. Clinical examination revealed a cachectic patient, normal BP and PR, hard painless swelling at the nose, thyroid was homogeneous in shape and normal size, without cervical lymphadenopathy, left lower limb was immobilized in traction. The biological exams found: hypercalcemia with corrected Calcium at 162 mg / L; normal PTH (1-84) at 1200 µg / L; hypophosphatemia at 22mg / L; vitamin D deficiency at 15 IU / L; a correct renal evaluation with serum creatinine at 7.8 mg / l (DFG: 85 ml / min); normal TSH at 3.96 mIU / L; alkaline phosphatases raised to 706 IU / L; hypochromic microcytic anemia at 11 g / dL. In addition, searching for multiple endocrine neoplasias was negative including a normal calcitonin (at 4ng / L) and urinary methoxylated derivatives which was negative. Morphological assessment found; cervical ultrasonography : infra-centimeter bilateral thyroid nodules classified TIRADS 3; parathyroid scintigraphy showed focused retention of MIBI-Tc99, at mediastinal level in favor of ectopic parathyroid adenoma, several brown tumors in the femur, left tibia; and left maxillary lesion. The radiological assessment: Brown tumor at the right and left legs, multiple left femoral osteolytic lesions. These lines of evidence argue for a parathyroid adenoma with bone impact. Preoperative medical management for the correction of acute hypercalcemia consisted of 0.9% saline rehydration with forced

diuresis, the use of bisphosphonates including ibandronic acid (Brondronat 6mg IVL) and also vitamin D supplementation. Parathyroidectomy consisted of block removal of the tumor. Pathological examination of the surgical specimen, weighing 15 g and measuring 4x2.5x2 cm, showed a histological appearance compatible with parathyroid adenoma including oxyphil

and intermediate cells with no capsular invasion neither pulmonary embolism. The postoperatively; after one week, was marked by hypocalcemia at 64 mg / l on day 5 rapidly corrected by a vitamin-calcium substitution; normal PTH (1-84) increased from 1200 to 9.9pg / L on day 1 postoperatively.

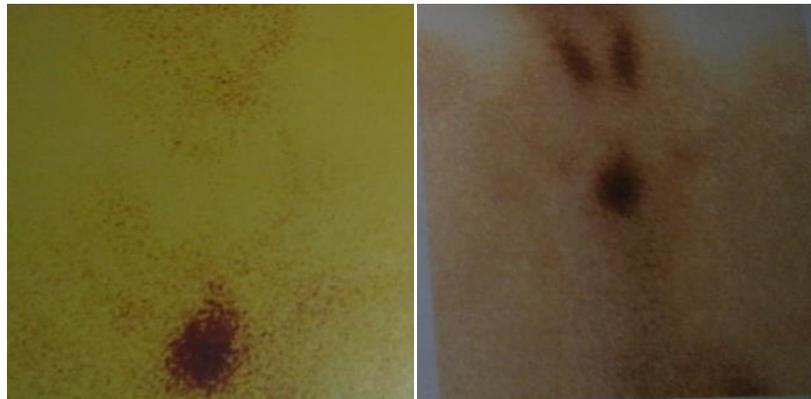


Fig-1: Scintigraphy images of ectopic parathyroid

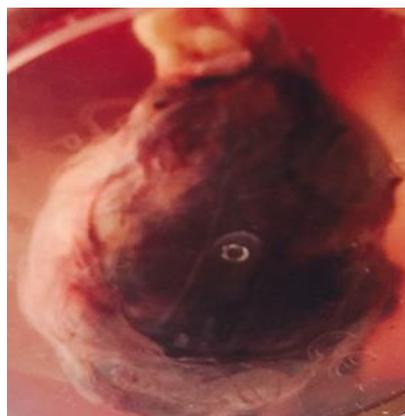


Fig-2: Image of parathyroid adenoma

DISCUSSION

Cervical or mediastinal parathyroid ectopia can be explained by either a defect or excess of embryological migration, or by migration acquired by gravity of a pathological gland [3-5]. Approximately in 10% of cases, the parathyroid gland is in ectopic paraesophageal or paratracheal high because of abnormal embryologic migration of the lower or higher parathyroid glands. The slightly less frequent mediastinal localization represents 10% of ectopic parathyroid adenomas [3, 4]. The frequency of parathyroid ectopia varies according to the series, Lappas and al. reported 8.5% of ectopic parathyroid glands on 942 autopsies; According to Andrade and al, 13.6% of ectopic parathyroid glands were found in a series of 664 operated parathyroid glands [5]. The preoperative diagnosis of the ectopic gland localization is important. According to many authors, MIBI-Tc 99m scintigraphy remains the examination of choice for the exploration of parathyroid glands and more particularly in their ectopic locations, especially mediastinal as it

was in the case of our patient. The difficulty of finding an ectopic parathyroid adenoma by the current only morphological imaging methods is related to the tissue polymorphism of the cervico-mediastinal floors, this would explain the sensitivity and above all the relatively specificity of the ultrasound, CT and sometimes even MRI in the identification of ectopic parathyroid tissue [4,6]. Ishabashi and al, promote the realization of MIBI-Tc 99m scintigraphy before any other imaging including CT or MRI. These authors report a sensitivity of 70% and a specificity of 88% for scintigraphy, results superior to CT and MRI [2]. Other studies refer to MIBI scintigraphy as the best diagnostic technique with a sensitivity approaching 90% [7]. The most common etiology of primary hyperparathyroidism remains the single parathyroid adenoma (80-85% of cases), followed by parathyroid hyperplasia (10-15% of cases), then double thyroid adenoma (2%). -5% of cases) and parathyroid carcinoma (1% of cases) [8,9]. Mediastinal localization represents 10% of ectopic parathyroid adenomas [4]. In the ectopic parathyroid

adenoma series of Roy *et al.*, The intrathyroid locations accounted for 18% after the thymic and retrooesophageal localizations but before the mediastinal location [8]. Brown tumors are non-neoplastic masses of reactional tissue; these lesions correspond to the final stage of bone remodeling secondary to hyperparathyroidism. Brown tumors are most commonly linked to primary hyperparathyroidism, but have been reported occasionally in patients with secondary or tertiary hyperparathyroidism [10]. Classically affected sites are the face bones, ribs, pelvis, femur and other long bones. Radiologically, they are well defined lytic lesions, without sclerosis, eccentric or cortical, sometimes thinning, blowing or rupture of the cortex [11]. Histologically; it is an area of osteoclast hyperresorption containing hypervascularized inflammatory connective tissue, giant cells, hemosiderin deposits, and areas of osteoid tissue that will replace normal bone [10]. In the case of our patient, the biopsy of the lower extremity of the left tibia had revealed grade I giant cells. The progress of brown tumors after parathyroidectomy is variable; in general, the increased risk of fracture disappears in the year following surgery, suggesting a rapid restoration of the biomechanical properties of the bone [11]. Whole body bone scintigraphy is of interest in postoperative follow-up in the medium and long term.

CONCLUSION

Parathyroid adenoma is the most common cause of primary hyperparathyroidism, which is often revealed by high levels of calcemia and parathormonemia, and also osteolytic fractures that can be multiplied. However, ectopic localization of the parathyroid adenoma remains exceptional. The treatment is mainly surgical, which attitude is oriented by preoperative location of the ectopia especially in mediastinal level by MIBI-Tc 99m scintigraphy.

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