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Short Report

About a rare cause of primary hyperparathyroidism

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ABSTRACT

Introduction: Primary hyperparathyroidism is observed in 35 to 44 subjects/ 100000 persons. The increased production of parathyroid hormones is secondary to primary glandular modifications consisting mainly in adenomas. The authors report a clear-cell hyperplasia causing primary hyperparathyroidism. **Observation:** We report the case of a 25-year-old man who was admitted to explore pathologic fractures of the left arm and a malignant hypercalcaemia. Complementary laboratory tests revealed primary hyperparathyroidism. A multiple endocrine neoplasia was excluded by radiologic examinations. Cervical ultra-sound examination revealed 2 parathyroid adenomas and per-operative exploration showed 3 « adenomas ». Microscopic examination of the 4 parathyroid glands specimen concluded to a clear cell hyperplasia. **Conclusion:** Clear cell hyperplasia is a benign cause of primary hyperparathyroidism. The diagnosis is based upon histologic findings and examination of the 4 glands.

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1. Introduction

Primary hyperparathyroidism is observed in approximately 35 to 44 persons/ 100000 subjects [1]. It occurs in the absence of a renal affection or an intestinal malabsorption. The over-production of parathyroid hormone is secondary to a parathyroid adenoma in 75 to 80 % cases, parathyroid hyperplasia in 12 to 18 % cases and parathyroid carcinoma in less than 5 % of the cases [2]. The authors describe a new case of clear cell hyperplasia diagnosed in a case of primary hyperparathyroidism.

1.Observation

A 25-year-old patient was explored for a pathologic fracture of the left arm and a malignant hypercalcaemia (4 mmol/l). He presented, many times ago, multiple fractures of the same arm and calcic renal lithiasis. Hormonal tests revealed a primary hyperparathyroidism with a parathyroid hormone level reaching 1339 mmol/l, a malignant hypercalcaemia reaching 4 mmol/l (N : 2,25- 2,62 mmol/l), a hypophosphoremia of 0,46 mmol/l (N : 0,8 - 1,45 mmol/l). An eventual multiple endocrinopathy was ruled out

thanks to many explorations. Cervical ultra-sound examination showed double adenomas, the first was median and the second was located in the left inferior parathyroid gland. Per-operative examination showed 3 adenomas. The first was isthmic, the second was located in the left inferior parathyroid gland and the third was situated in the posterior face of the left thyroid lobe. Grossly, we received 3 glands measuring 1cm, 2,5 cm and 3 cm. Microscopic examination revealed, in the three glands, a proliferation of monotonous clear cells organized in trabeculae and masses (Figure 1a). At a higher power, intra-cytoplasmic clear vacuoles were noted (Figure 1b). A normal parathyroid parenchyma wasn't observed in these glands. Post operatory, the calcaemia level decreased and the parathyroid hormone level remained elevated. A post operatory scintigraphy showed the persistence of an ectopic left latero-oesophageal parathyroid gland. The patient was reoperated. Microscopic analysis of the ectopic gland showed the same features as those of the remaining glands. The diagnosis of clear cell parathyroid hyperplasia was retained.

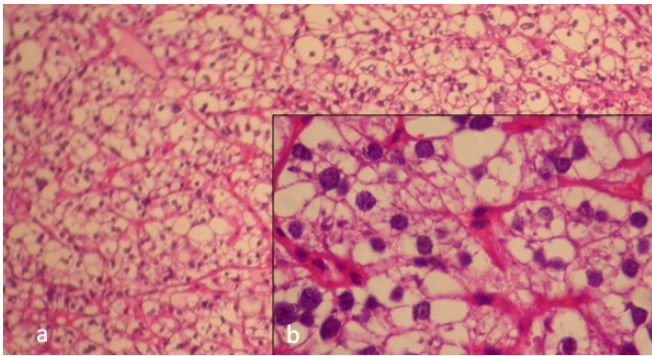
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Figure 1: a/ Proliferation of homogeneous water-clear cells arranged in sheets or nests b/ Intra-cytoplasmic clear vacuoles



3. Discussion

Primary hyperparathyroidism may be familial or sporadic. In case of familial hyperparathyroidism, microscopic examination reveals principal cell hyperplasia and is observed particularly in case of type I and III multiple endocrinopathies. In case of sporadic hyperparathyroidism, microscopic examination reveals adenoma or clear cell hyperplasia. This last entity is rare and is few documented. Initially, it was described by Albright in 1938. Its frequency seems to be less than 1% of all primary hyperplasia. It is mainly observed in men and never associated to a multiple endocrinopathy [1]. It is characterized by an important increase of the parathyroid parenchymal mass, the total weight of the four glands can be more than 100 g can be manifested grossly by a lesion mimicking an adenoma like the case presented. The particularity of this observation consists in the attempt of ectopic glands. In the opposition to principal cell hyperplasia, upper glands are visibly bigger than the inferior ones. these glands are soft in consistency with a chocolate like color. They are sometimes the site of haemorrhage or cysts [3]. Microscopic examination shows a monotonous proliferation of big clear cells with clear cytoplasm [1]. This lesion must be distinguished from clear cell adenoma or principal cell hyperplasia with a clear cell component ad a parathyroid metastases of a clear cell renal carcinoma. The distinction from an adenoma can be challenging in some cases. In fact, radiologic findings showed multiple adenomas in our case. Otherwise, the simultaneous attempt of three glands, the absence of atrophic or normal parathyroid parenchyma justify the diagnosis of parathyroid hyperplasia. [2]. Lawrence and colleagues proved in a retrospective study of 18 cases of hyperplasia, that nuclear pleomorphism in adenomas, the nodular architecture of hyperplasia represent criteria of differentiation that is statistically significant [4]. Principal cell hyperplasia is characterized by the presence of principle cells mixed with other types of cells. Some authors reported that principal cell hyperplasia is an initial stage of clear cell hyperplasia [5]. Clear cell carcinoma of the parathyroid gland is characterized by the invasion of the adjacent tissue. Microscopically, the capsular invasion, vascular emboluses, atypia and mitoses (> 5 mitoses/ 50 champs) are the major signs of malignancy [2]. A metastatic localization of a renal carcinoma could be suspected according to the clinical features and

confirmed thanks to immunohistochemical study. All these diagnoses must be suspected and ruled out in order to provide the adequate treatment. In fact, the treatment of the entity is surgical and consists in a sub-total parathyroidectomy.

4. Conclusion

Clear cell hyperplasia is a very rare cause of primary hyperparathyroidism. The diagnosis is based on the histological study of the four parathyroid glands.

Conflict of interest:

The authors declare that they haven't any conflict of interest.

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