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CASE REPORT

Brown tumor in a patient with ectopic mediastinal parathyroid adenoma: A case report

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KEYWORDS

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Abstract Brown tumors are uncommon focal giant-cell lesions that arise as a direct result of the effect of parathyroid hormone on bone tissue in some patients that have hyperparathyroidism. Primary hyperparathyroidism could be caused by ectopic mediastinal parathyroid adenomas. The occurrence of lesions is explainable on embryologic basis. We present a 55-year-old Saudi woman with a rare case of brown tumor of the maxilla due to ectopic mediastinal parathyroid adenoma.

1. Introduction

Brown tumor is one of the clinical manifestations of primary or secondary hyperparathyroidism. It is a component of a metabolic bone disease recognized as osteitis fibrosa, cystica generalisata or Recklinghausen's disease of bone (Neville et al., 2009; Pinar Sumer et al., 2004). It usually occurs in the long bones, pelvic girdle, clavicle, ribs, and the mandible. Brown tumor is rarely involved in the maxilla.

The reported prevalence of brown tumors is 0.1%, with a male to female ratio of 1:3 (Proimos et al., 2009). Symptoms can occur at any age; however, the disease is more common in persons older than 50 years

Primary hyperparathyroidism in 80% of the cases is due to a parathyroid adenoma; in over 15% of the cases it is due to a glandular hyperplasia, and extremely rare due to a parathyroid adenocarcinoma (Thompson et al., 1982). In most cases, adenomas are located in the neck, while in 1–3% of the cases they arise in an ectopic site and could be found anywhere between the angle of the jaw and the pericardium. As a result of variability in glandular tissue migration during embryologic life (Weller, 1933), in more than 80% of the cases the ectopic parathyroid adenoma is found inside the mediastinum.

Primary hyperparathyroidism initially manifesting as a brown tumor located in the anterior maxilla is rare. Our case report presents a 55-year-old woman with a brown anterior maxilla tumor arising in the ectopic mediastinal parathyroid adenoma.

2. Case report

A 55-year-old Saudi woman was admitted to the Oral and Maxillofacial Surgery unit at the King Abdulaziz University Hospital (Jeddah) with a complaint of anterior maxilla mass of one month duration. Her medical history was unremarkable.

Initial examination revealed a painful mass in the anterior maxilla (Fig. 1). An anesthesia consultation was requested,

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Figure 1 Preoperative image of the maxillary tumor.

and surgery (including the excision of the lesion and curettage of the bone under general anesthesia) was offered; however, surgery was not undertaken because routine laboratory investigations revealed hypercalcemia.

The endocrinologist was urgently consulted for investigation and treatment of hypercalcemia in this patient. She also reported a six-month history of asthenia, generalized muscle pain, polyuria and constipation. The patient appeared well and had an unremarkable physical examination.

Initial laboratory tests performed on admission showed the following: alkaline phosphates 143 IU/L (reference range, 50–136 IU/L), corrected serum calcium 3.2 mmol/L (reference range, 2.12–2.52 mmol/L), serum phosphate 0.55 mmol/L (reference range, 0.8–1.58 mmol/L), intact parathyroid hormone 120 pmol/L (1.6–6.9 pmol/L).

A provisional diagnosis was made for hypercalcemia due to hyperparathyroidism. Further investigations were performed. These included technetium thallium (99mTc-201Th) subtraction scintigraphy (Sestamibi scanning), which demonstrated a single, ectopic anterior mediastinal parathyroid adenoma (Fig. 2). Magnetic resonance imaging (MRI) of the thorax showed a mediastinal parathyroid adenoma (Fig. 3).

Treatment was initiated by hydration with normal saline and intravenous biophoshante until normalization of the patient's serum calcium level.

Initially the patient underwent extirpation of the mass and curettage of the bone under general anesthesia. Histological

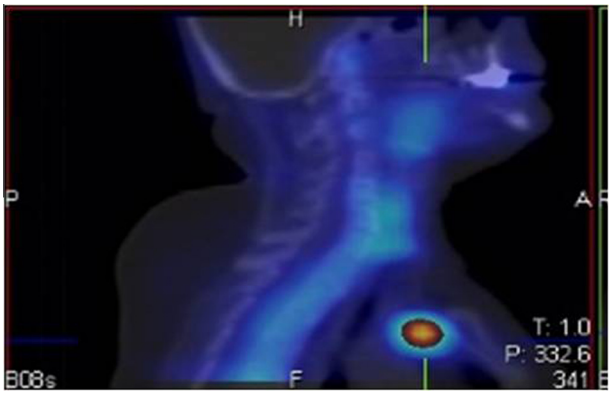

Figure 2 Sestamibi scan showing the ectopic mediastinal parathyroid adenoma.

sections showed multiple giant cells consistent with Brown tumor of primary hyperparathyroidism (Fig. 4).

The patient was readmitted after two weeks, and she underwent mediastinal parathyroidectomy by median sternotomy. The histopathological diagnosis of the lesion was parathyroid adenoma (Fig. 5). The post-operative course was uneventful and on the sixth post-operative day, the patient was discharged without complications. The results of postoperative laboratory tests were normal (Graphs 1 and 2).

3. Discussion

Hyperparathyroidism was first diagnosed by Sylvanus, while Recklinghausen was credited with the first description of the associated bone changes known as osteitis fibrosa cystic (Neville et al., 2009).

The brown tumor is mainly due to secondary hyperparathyroidism in patients with renal insufficiency, but it has also been known as a rare manifestation of calcium malabsorption and some forms of osteomalacia (Pinar Sumer et al., 2004). These days, brown tumors are an extremely infrequent manifestation of primary hyperparathyroidism because of routine screening of hypercalcemia and early diagnosis of hyperparathyroidism. In our patient, the histological diagnosis was indicative of a giant cell bone expanding lesion with a brown tumor being the most probable diagnosis. Blood tests established diagnosis of primary hyperparathyroidism. The localization of primary hyperparathyroidism revealed a big ectopic mediastinal parathyroid adenoma with overproduction of parathyroid hormone.

The treatment of hyperparathyroidism by resection of parathyroid adenoma is the initial step in the management of the brown tumor. Brown tumor regression and healing are predicted after the correction of hyperparathyroidism. However, many case reports of brown tumor in the litutre showed that the tumors grew parathyroidectomy or normalization of parathyroid hormone level. The treatment of choice in these cases should be brown tumor resection (Yamazaki et al., 2003).

The incidence of bone lesions in patients with hyperparathyroidism has reduced from 80% to 15%, this reduction has attributed to better hypercalcemia monitoring in asymptomatic patients and to the greater use of biochemical analyses (Praveen and Thriveni, 2012).

Brown tumors are one of the osseous manifestations of hyperparathyroidism. Brown tumors appear in around 10% of the cases in the advanced stages of the chronic kidney disease. It could be found in any part of the skeleton, most commonly seen on the ribs, clavicle and pelvis. 4.5% has been located in the mandibul (Pahlavan and Severin, 2006; Sia et al., 2012).

Parathyroid tumors are usually located on the posterior capsule of the thyroid but may be in other ectopic locations. Ectopic parathyroid adenomas report for 1–3% of all parathyroid tumors. Most of the tumors are located in the anterior mediastinum (Al-Mashat et al., 2009). An enlarged gland descends into the mediastinum because of its higher mass as well as a result of intrathoracic negative pressure and esophageal movement. Ectopic locations are certainly connected to the migratory pathways of embryologic parathyroid tissue to the adult positions (Weigel et al., 2005).

The most frequent location for ectopic parathyroid tumors is the mediastinum. Approximately 20% of parathyroid