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Case report

A giant parathyroid carcinoma cause deformation of thorax: Case report

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ABSTRACT

We present a rare case of a large parathyroid carcinoma (PC) characterized by severe hyperparathyroidism, hypercalcemia, and osteoporosis. Long-standing calcium loss resulted in thoracic and facial deformities, initially misdiagnosed as oral malignancy.

1. Introduction

The 2022 WHO classification distinguishes the parathyroid neoplasms in parathyroid adenoma (benign), APT (parathyroid tumour of uncertain malignant potential), and parathyroid carcinoma (PC). Parathyroid carcinoma (PC) is an extremely rare malignancy, accounting for less than 1 % of all parathyroid tumors and approximately 0.005 % of all malignancies. Most PCs are functional tumors and often present with common hyperparathyroidism symptoms [1]. This case report highlights a patient with parathyroid carcinoma presenting with classic hyperparathyroidism symptoms. We present a case of a 55-year-old Chinese man who suffered from hyperparathyroidism due to a giant size of PC.

2. Case/case series presentation

A 55-year-old Chinese man with a history of oral carcinoma diagnosed 6 months ago, resulting in eating difficulties, was referred to a maxillofacial surgeon(Figs. 3 and 4). He had also sustained two traumatic injuries more than 20 years prior (significance to be determined later). In the past 7 years, he developed a progressive chest collapse and deformity(Figs. 1 and 2). More recently, within the last 10 months, he experienced maxillofacial dysmorphism, characterized by protrusion of the right maxilla and mandible, leading to difficulty achieving proper occlusion and further worsening his eating difficulties.

Based on the patient's medical history and clinical manifestations, the maxillofacial surgeon initially suspected a maxillofacial malignant tumor and planned surgery. Preoperative examination revealed severe hypercalcemia (Ca^{2+} 3.75mmol/L, Colorimetric Assay), hyperparathyroidism (PTH 2104pg/ml), MRI examination revealed multiple bone metastases, suggestive of parathyroid malignancy(Fig. 5). The intraoral mass exhibited osteolytic changes on pathology, consistent with hyperparathyroidism. Due to severe hypercalcemia, the patient was transferred from thyroid surgery after a rise in blood calcium levels. Subsequent nuclear medicine

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Fig. 1. Thorax deformation.

examination revealed a hypermetabolic state in the left lower parathyroid gland(Fig. 6). In addition, the patient was found to have urinary tract stones, which is another typical manifestation of hyperparathyroidism(Fig. 7).

During surgery, the left lower parathyroid gland was found to be significantly enlarged. The mass was completely removed after careful identification and preservation of the left recurrent laryngeal nerve. A concomitant left thyroid lobectomy was also performed.

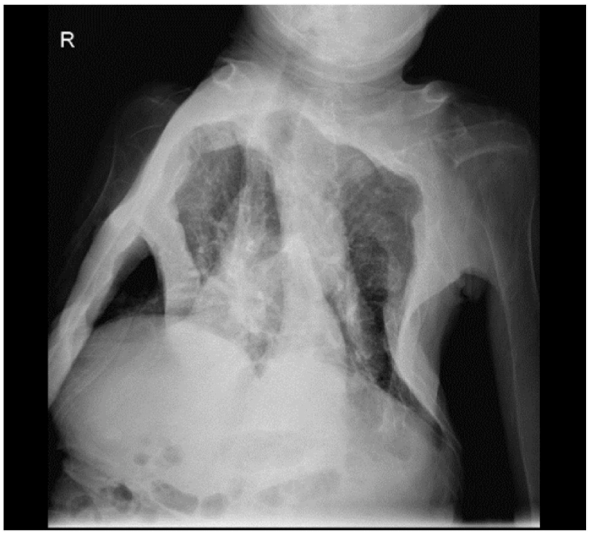


Fig. 2. X-Ray for thorax deformation.

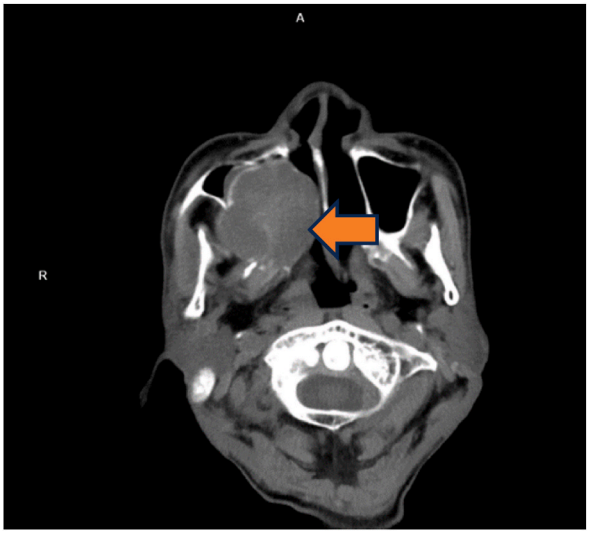


Fig. 3. CT for maxillary tumor.

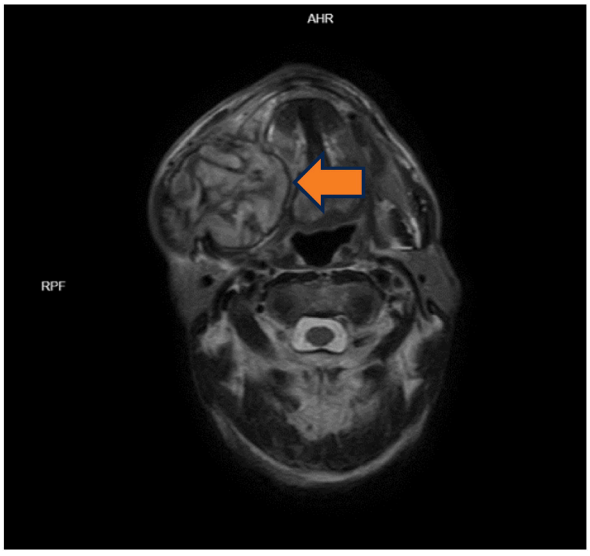


Fig. 4. MRI for maxillary tumor.

Pathological examination of the resected mass, measuring 50 × 41 × 20 mm and weighing 60 g, revealed a parathyroid adenocarcinoma (Figs. 8 and 9). Intraoperatively, a blood sample for serum calcium level was obtained, which returned elevated at 4.95 mg/dL (495 ng/mL). A repeat measurement 2 h later showed a significant decrease to 0.75 mg/dL (75 ng/mL). Parathyroid hormone levels normalized by postoperative day 2, and serum calcium levels returned to normal by day 3. By postoperative day 4, the mass in the right mandibular region began to shrink, and the patient could even attempt to chew food. We initiated treatment with oral calcium and alfacalcidol tablets.

At the six-month follow-up, the patient reported feeling well and comfortable, with normal serum calcium levels.

3. Discussion

This case report presented a 55-year-old man with hypercalcemia, a classic sign of parathyroid disease. While initially suspected to have parathyroid carcinoma due to clinical presentation and imaging, surgical resection revealed a parathyroid adenoma. This case underscores the importance of definitive diagnosis through histopathological examination, as clinical presentation and imaging may not always be conclusive for differentiating between parathyroid carcinoma and adenoma. The patient's postoperative course was successful, with normalization of calcium levels and improvement in symptoms.

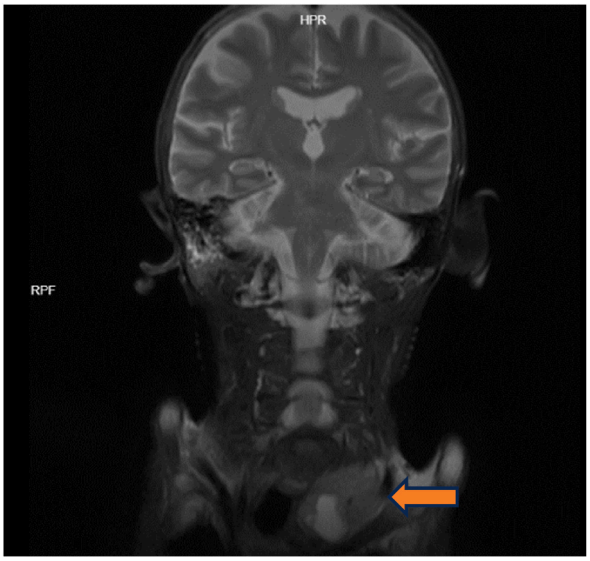


Fig. 5. MRI for parathyroid (LL) .

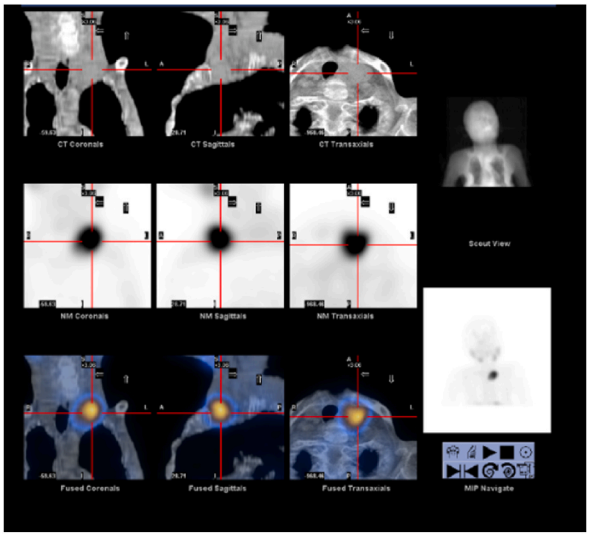


Fig. 6. 99Tmc-MIBI for parathyroid (LL).

Parathyroid carcinoma (PC) is a rare disease, accounting for less than 1 % of primary hyperparathyroidism cases. Preoperative diagnosis is challenging due to its rarity and lack of specific clinical features. Studies like the pTRANI Study report higher rates of bone involvement in PC compared to benign parathyroid disease, which aligns with the severe osteoporosis observed in this case [2]. Ultrasonography and 99mTc-MIBI are commonly used for initial detection, while CT and MRI scans aid in localization and evaluation of potential spread [3]. This case presented a unique challenge. The patient initially presented with clinical and imaging findings suggestive of oral cancer during dental surgery. However, the presence of severe osteoporosis with thoracic deformity prompted further investigation. Laboratory tests revealed hyperparathyroidism (elevated PTH) due to severely abnormal thyroid function. Subsequent 99mTc-MIBI and MRI scans confirmed a large parathyroid mass. These findings aligned with previously reported cases of parathyroid carcinoma, which was ultimately confirmed by pathology.

Fortunately, the patient underwent a successful parathyroidectomy, achieving complete resection with no capsular rupture, which is crucial to minimize the risk of local recurrence in PC [4]. While complete and oncologic surgical resection with clear margins is ideal for PC or suspicious PC to improve long-term outcomes [5,6], the patient's family declined the surgeon's recommendation for further surgery to remove the thyroid gland. The potential implications of this decision for the patient's long-term prognosis warrant close follow-up [7].

After adequate surgical resection, further therapies are not standardised: no evidence of helpful adjuvant radiotherapy or

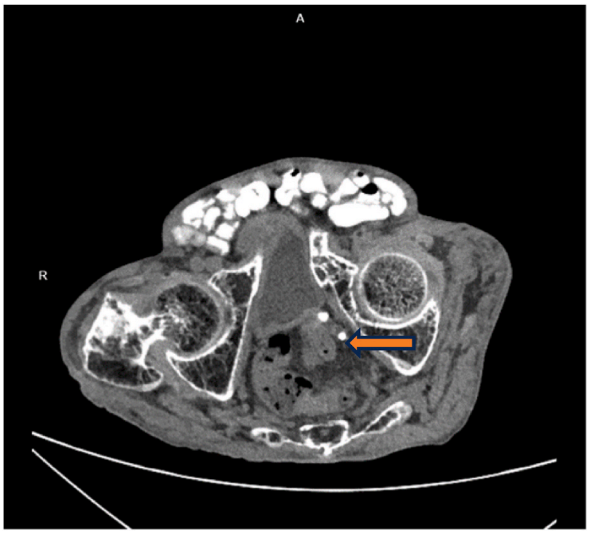


Fig. 7. CT for ureter stone.

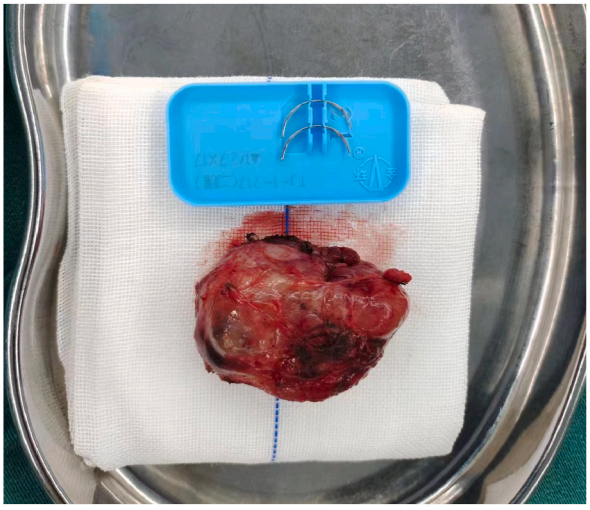
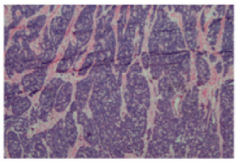


Fig. 8. Gross specimen.

巨　检：
灰白灰红色组织一块，体积5×4.1×2cm，切开切面灰红，质中。

镜像图像：



病理诊断：
（左侧甲状旁腺）甲状旁腺肿瘤，经常规多取材，可见包膜侵犯，脉管内可见癌栓，考虑甲状旁腺腺癌。
注：请结合临床。

Fig. 9. Pathology report.

chemotherapy is available [8,9]. The patient's postoperative course was uneventful, and the symptoms of hypercalcemia resolved after surgery. Since uncontrolled hypercalcemia is the primary driver of morbidity and mortality in PC, the mainstay of pharmacotherapy focuses on controlling calcium levels in patients with PC or APT and persistent/recurrent hypercalcemia [10,11]. Regular follow-up will include monitoring serum calcium levels, parathyroid hormone levels, and imaging studies to detect potential recurrence. The patient will also require lifelong surveillance for hypocalcemia, a potential complication of parathyroidectomy [12].

4. Conclusion

This case of giant parathyroid carcinoma presents a rare and challenging clinical scenario. The patient's severe thoracic deformity and maxillofacial changes underscore the importance of considering parathyroid carcinoma in the differential diagnosis of patients with unexplained skeletal abnormalities. Despite the challenges in preoperative diagnosis, early detection and aggressive surgical management are crucial for improving patient outcomes.

Our report highlights the need for a high index of suspicion for parathyroid dysfunction in patients with multiple fractures, renal calculi, or atypical bone pain, even in the absence of classic hypercalcemia symptoms. This case underscores the importance of a multidisciplinary approach involving endocrinologists, orthopedic surgeons, and urologists for the timely diagnosis and management of parathyroid carcinoma.

Ethics statement

We hereby confirm that we have read and complied with the policy on ethical conduct.

The patient (or his proxies/legal guardians) provided written informed consent for the publication of all clinical data and other data included in the main manuscript.

Gross description

A solitary, well-circumscribed, gray-white to gray-red soft tissue mass measuring 5 x 4.1 $\times$ 2 cm was submitted for pathological examination. On cut section, the tumor has a homogeneous, gray-red, firm texture.

Microscopic diagnosis

Microscopic examination of multiple sections reveals a neoplastic proliferation of parathyroid cells arranged in sheets and nests. The tumor cells exhibit moderate to marked nuclear pleomorphism, increased mitotic activity, and prominent nucleoli. The tumor demonstrates loss of the distinct cellular boundaries and invades the surrounding adipose tissue and blood vessels. Notably, there is evidence of capsular invasion and intratubular carcinoma, characterized by neoplastic cells within the lumina of small blood vessels. Based on these findings, a diagnosis of parathyroid adenocarcinoma is rendered.

CRediT authorship contribution statement

Lu Guo: Writing – original draft, Project administration, Investigation. **Zhixin Shen:** Resources. **Luyao Zhang:** Writing – review & editing, Writing – original draft, Project administration.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.heliyon.2024.e38519.

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