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Resection of a large ectopic parathyroid adenoma: A case report

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ABSTRACT

INTRODUCTION: Parathyroid adenomas are the most common cause of primary hyperparathyroidism. However, cases of parathyroid adenomas greater than 4 cm with osteitis fibrosa cystica are extremely rare. Herein, we report a case of resection of a large ectopic mediastinal parathyroid adenoma.
CASE PRESENTATIONS: A 46-year-old female with chief complaints of bone pain and gait disturbance was referred to our hospital. Physical examination revealed many mobile teeth in her oral cavity, distortion of the vertebral body, and bowlegs. Laboratory tests showed hypercalcemia, hypophosphatemia, and elevated serum levels of intact parathyroid hormone. Chest CT revealed a 42-mm well–defined, enhancing mass in front of the left-sided tracheal bifurcation. Her findings were diagnosed as primary hyperparathyroidism due to an ectopic mediastinal parathyroid tumor. We performed a median sternotomy and resected the tumor. The tumor was a solid, yellowish-brown mass measuring 42 × 42 mm. Pathologically, the tumor consisted mainly of chief cells with some oxyphil cells; there were no necrotic areas or nuclear atypia, and few mitotic figures. We diagnosed the tumor as an ectopic mediastinal parathyroid adenoma. Eight months after the resection, her serum calcium, phosphorus, and intact PTH levels were normal.
DISCUSSION AND CONCLUSIONS: Parathyroid adenomas and parathyroid carcinomas have disparate natural histories, but they can be difficult to differentiate on the basis of preoperative clinical characteristics. We believe that long-term follow-up of these cases is required because there have been few reports on the postoperative natural history of large parathyroid adenomas.

1. Introduction

Differentiation of benign and malignant parathyroid tumors is sometimes difficult. In fact, because parathyroid carcinomas have a very low incidence rate, no staging system has been established by the American Joint Committee on Cancer (AJCC). Generally, diagnoses of parathyroid adenoma and parathyroid carcinoma are made on the basis of both the clinical findings and the histological criteria as proposed by Schantz and Castleman [1]. Herein, we report our experience with a case of a large ectopic mediastinal parathyroid adenoma with osteitis fibrosa cystica, which was diagnosed as a parathyroid adenoma based on the pathological findings, despite parathyroid carcinoma being initially suspected due to preoperative clinical findings.

2. Presentation of case

A 46-year-old female with complaints of bone pain, gait disturbance, and a feeling of weakness was referred to the Division of Thoracic and Cardiovascular Surgery of our hospital for evaluation of an abnormal shadow on a chest computed tomography (CT). Her past medical history was significant only for a thoracic wall deformity which had developed 4 years earlier. Physical examination revealed many mobile teeth in her oral cavity; distortion of the vertebral body; and bowlegs, which are associated with the features of osteitis fibrosa cystica; but no presence of palpable mass on her neck.

Laboratory tests showed slight anemia (10.4 g/dL; normal range, 11.5–14.0 g/dL); elevated alkaline phosphatase level (7617 IU/L; normal range, 120–325 IU/L); hypercalcemia (15.8 mg/dL; normal range, 8.7–11.0 mg/dL); hypophosphatemia (2.2 mg/dL; normal range, 2.6–4.4 ng/dL); elevated serum levels of intact parathyroid hormone (PTH) (2560 pg/mL; normal range, 10–65 pg/mL); and 1,25-dihydroxyvitamin D (82.9 pg/mL; normal range, 20–60 pg/mL). On pulmonary function testing, vital capacity (VC) and percent VC were 2160 mL and 79.0%, respectively. A chest X-ray revealed a remarkable thoracic wall deformity (Fig. 1A,

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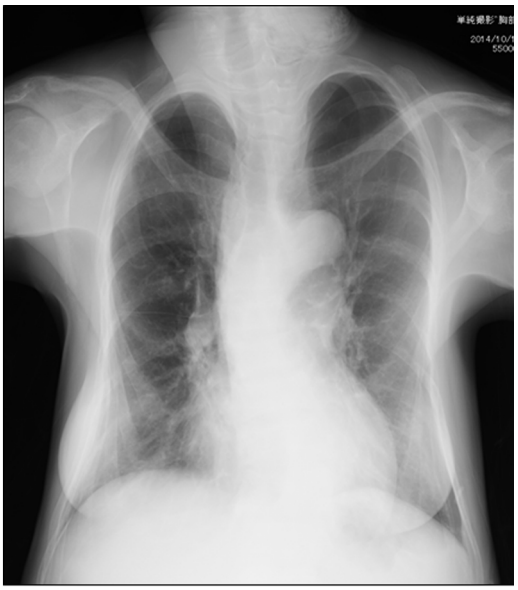
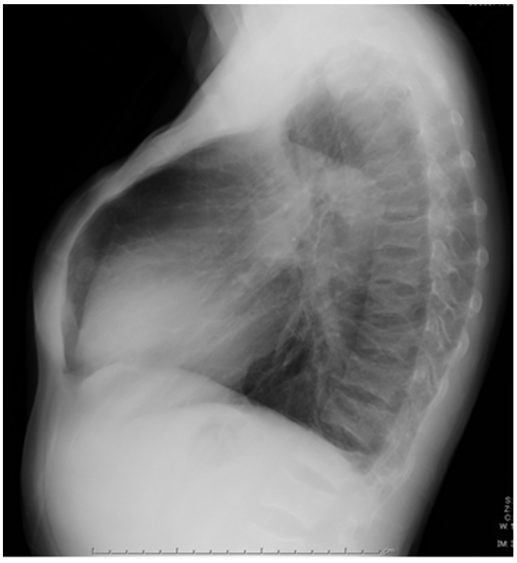


A

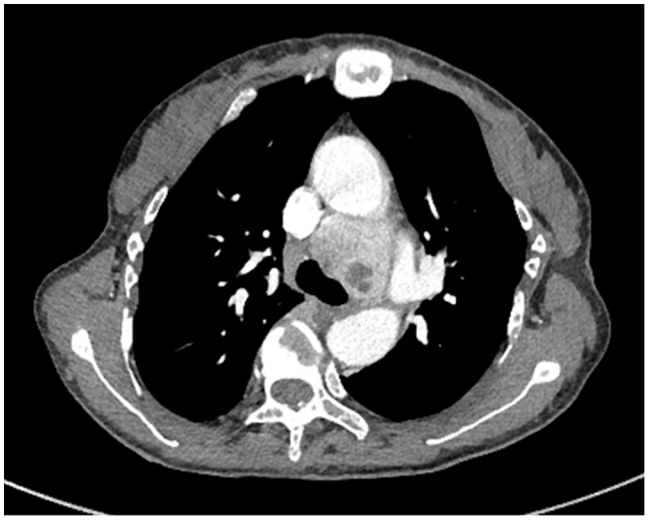


B

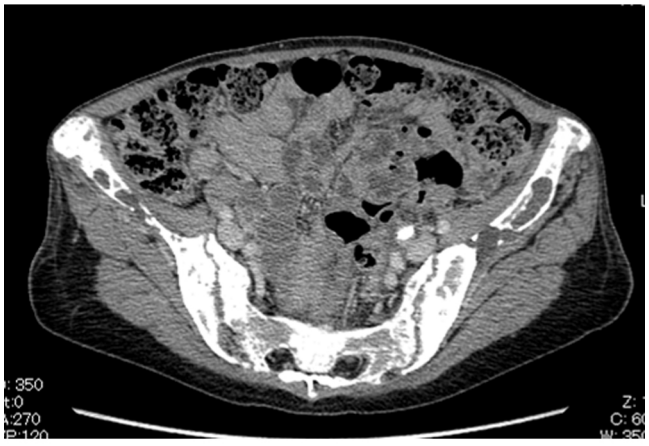
Fig. 1. Chest X-ray revealing a remarkable thoracic wall deformity on the frontal (A) and lateral view (B).

B), but no tumor was identified. A CT scan revealed a 42-mm, well-defined, enhancing mass, including a partial low-density area in front of the left-sided tracheal bifurcation (Fig. 2A), and extremely low bone mineral density (Fig. 2B). 99m Tc-methoxy-isobutyl-isonitrile (MIBI) scintigraphy showed an accumulation in the mediastinum in both the early and the late phases. We then diagnosed her findings as primary hyperparathyroidism due to an ectopic mediastinal parathyroid tumor.

On clinical examination, because a malignant tumor could not be ruled out, we scheduled the patient for a resection of the mediastinal ectopic parathyroid tumor, including the surrounding lymph nodes. We performed a median sternotomy and a pericardial incision. After dissection between the superior vena cava and the ascending aorta, we made an incision in the pericardium behind these vessels and exposed the ectopic parathyroid tumor. We were able to bluntly resect the tumor from the proximal right pulmonary



A



B

Fig. 2. CT scan revealing a 42 mm, well-defined, enhancing mass, including a partial low-density area in front of the left-sided tracheal bifurcation.
(A) The iliac bone mineral density is extremely low.

artery and left main bronchus. Macroscopically, the tumor was a solid, yellowish-brown mass measuring 42 × 42 mm, with partial foci of hemorrhage. The entire specimen seemed to be enclosed by a thin, fibrous, capsule-like structure and by adipose tissue. The tumor was well-circumscribed, but there was no capsule and few small nodules in the surrounding adipose tissue (Fig. 3A).

On pathological examination, the tumor consisted mainly of chief cells and some oxyphil cells (Fig. 3B). There were no necrotic areas or nuclear atypia, and few mitotic figures. Ki67 immunostaining was generally positive in about 1% of the tumor. There was no blood vessel invasion on Elastica Van Gieson staining, and no lymphatic involvement on D2-40 staining. A small rim of normal tissue was existed. From the above histopathological results, the tumor was diagnosed as an ectopic mediastinal parathyroid adenoma.

Postoperatively, we had great difficulty treating her persistent hypocalcemia and hypophosphatemia (so-called "hungry bone syndrome") [2]. After rehabilitation, which mainly comprised gait training, she was discharged from the hospital on postoperative day 65, ambulating with an independent gait. Eight months after the resection, her serum calcium, phosphorus, and intact PTH levels were normal (9.2 mg/dL, 3.5 mg/dL, and 47 pg/mL, respectively) (Fig. 4).