Case Report

Thoracoscopic Removal of a Maternal Mediastinal Ectopic Parathyroid Adenoma Causing Neonatal Hypocalcemia: A Case Report

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Most parathyroid glands in hyperparathyroidism can be resected through a cervical approach. In approximately 2% of cases, the ectopic gland is in the mediastinum in a location that requires a thoracic approach. Advancement in video-assisted thoracoscopic surgical (VATS) techniques has decreased the need for sternotomy to successfully remove these ectopic glands. We describe a case involving a 29-year-old woman with hyperparathyroidism resulting from an ectopic mediastinal parathyroid adenoma that caused neonatal hypocalcemia, which was removed through VATS. (Ann Thorac Cardiovasc Surg 2008; 14: 325–328)

Key words: mediastinum, ectopic parathyroid adenoma, video-assisted thoracoscopic surgery, neonatal hypocalcemia

Introduction

The need to remove a parathyroid gland in the mediastinum is an infrequent occurrence.[1)] Although the removal of an ectopic parathyroid gland through a cervical incision may be successful in many patients, a median sternotomy is often required. This procedure has the potential for causing more morbidity and is associated with up to 12% incidence of complications in such patients.[2)] Advancement in video-assisted thoracoscopic surgical (VATS) techniques has decreased the need for sternotomy to successfully remove these ectopic glands.

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Case Report

A 29-year-old woman with no previous health problems gave a healthy birth. Post partum on the first month, her baby had convulsive attacks. This clinical situation was diagnosed as hypocalcemia and was treated. She did not accept the advice for an additional evaluation of her situation. In the past 2 months she had experienced muscle weakness, bilateral flank pain, and alopecia. Her symptoms became more severe and she was admitted to a hospital. After hospitalization, she had generalized abdominal pain and high amylase level. The serum calcium level was 13 mg/dl (normal = 8 to 10.4), potassium 4.15 mmol/L (normal = 3.50 to 5.50), alkaline phosphatase level 1,431 IU/L (normal = 30 to 126), parathyroid hormone (PTH) level 2,490 pg/ml (normal = 10 to 69), prolactin 1.25 ng/ml (normal = 1.20 to 29.93), and thyroid function tests were within normal ranges. Abdominal ultrasonography (USG) and abdomino-pelvic computed tomography (CT) revealed a diagnosis of acute pancreatitis and bilateral nephrolithiasis.

Thyroid USG and cervical magnetic resonance imaging (MRI) scan were normal. A technetium-99m

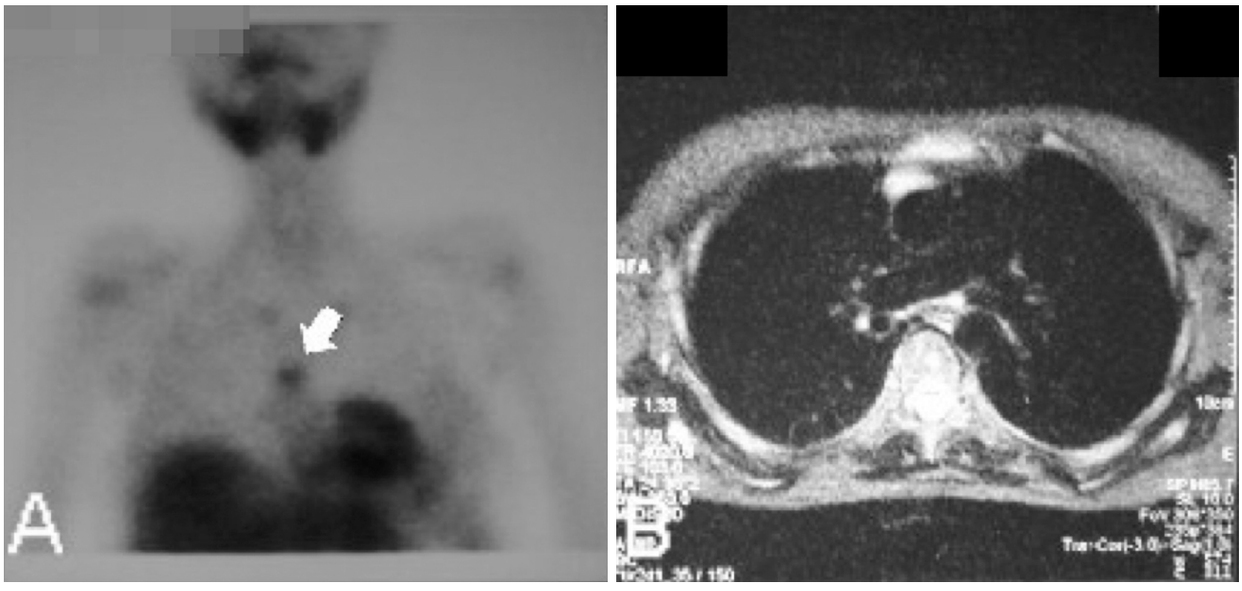

Fig. 1.
A: Preoperative technetium-99m pertechnetate methoxy-isobutyl-isonitrite (^{99m}Tc-MIBI) nuclear scan revealing a focus of increased uptake in the anterior mediastinum (arrow).
B: Chest magnetic resonance imaging (MRI). It shows a lesion with high signal intensity on a short tau inversion recovery (STIR) sequence, typical of parathyroid adenoma.

pertechnetate methoxy-isobutyl-isonitrite (^{99m}Tc-MIBI) and single photon emission CT (SPECT) nuclear scanning revealed a focus of increased uptake in anterior mediastinum (Fig. 1A). A spiral CT and MRI of the chest showed a 2.5 to 1.5 cm lesion in the anterior mediastinum (Fig. 1B). We focused on the diagnosis as an ectopic parathyroid gland, according to the clinical data of the patient and planned surgery.

The patient was placed in the left lateral decubitus position and intubated with a double-lumen endotracheal tube for single lung ventilation. A thoracoscopic exploration from the right hemithorax using four thoracal openings was performed, and a 2.5 × 3 cm mass in the anterior mediastinum was visualized (Fig. 2A). The lesion was resected with surrounding mediastinal fatty tissue (Fig. 2B). Before extubation, a 28F thoracostomy tube was placed through the inferior thoracal opening. The thoracostomy tube was removed on the first postoperative day, and there were no surgical complications. Postoperative serum calcium levels dramatically decreased to the levels of 6 mg/dL, and she was supported with oral calcium and vitamin D. The histopathological diagnosis confirmed the lesion as parathyroid adenoma.

Discussion

Eighty-five percent of all primary hyperparathyroidism cases are caused by parathyroid adenomas, whereas parathyroid hyperplasia and parathyroid carcinoma account for 15% and less than 1%, respectively. Mediastinal ectopic parathyroid glands in primary hyperparathyroidism have been as high as 20%.[3] Hyperparathyroidism patients have symptoms related to hypercalcemia, including hypercalciuria, polyuria, thirst, fatigue, and vomiting. Visceral calcification and bone resorption can be seen. Hyperparathyroidism during a pregnancy period may cause neonatal hypocalcemic tetany secondary to a suppression of PTH production by high fetal calcium levels.[4]

Preoperative imaging studies for the localization of abnormal glands include USG, CT, ^{99m}Tc pertechnetate-thallium 201 scintigraphy, MRI, selective venous sampling, selective arteriography, and more recently ^{99m}Tc-MIBI scintigraphy**.** Parathyroid glands larger than 1.5 cm are typically seen in CT scans, but smaller glands may be difficult to identify.[1] Compared with CT, the sensitivity of MRI was higher for detecting mediastinal lesions, but not as good as ^{99m}Tc-MIBI alone.[5] It appears that the ^{99m}Tc-MIBI scintigraphy may

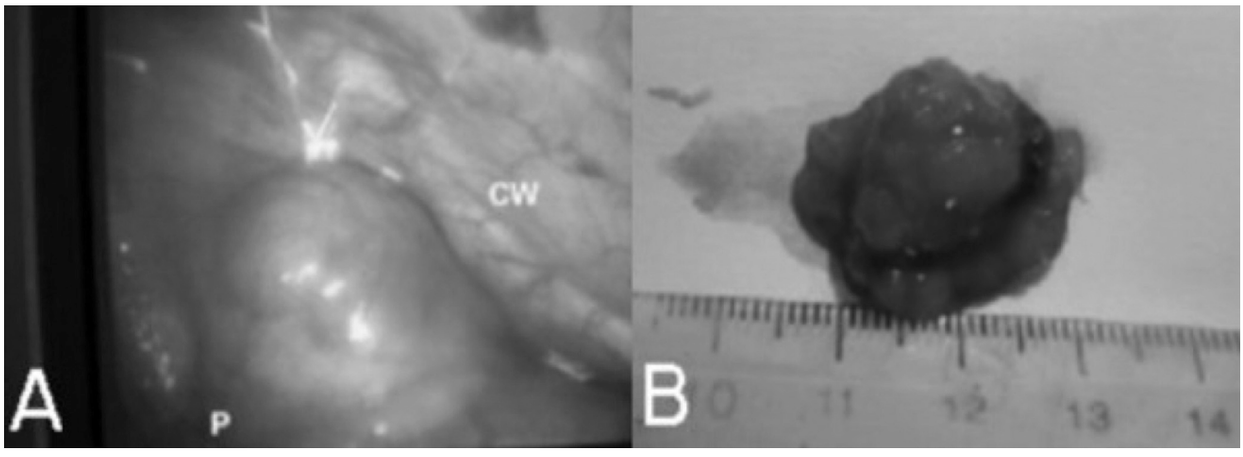

Fig. 2.
A: Intraoperative view during right thoracoscopy. The adenoma is denoted by the asterisk. CW, chest wall; P, pericardium.
B: Excised parathyroid adenoma (a 2.5 × 3 cm round, well-shaped mass).

offer the highest identification rate. ^{99m}Tc-sestamibi SPECT scanning with the gamma probe can also help intraoperative localization of the lesion.[6] The lesion localization was well depicted by both CT and MRI examinations; moreover, MRI demonstrated the hyperintensity of the lesion on a short tau inversion recovery (STIR) sequence with the possibility of parathyroid adenoma. After mediastinal localization by ^{99m}Tc-MIBI SPECT, either a chest CT or an MRI should be used to anatomically confirm the location of the mass. Alternatively, if a combined SPECT/CT device is available, it can be used as an initial imaging procedure so that it can provide the functional and anatomical details simultaneously.

Parathyroidectomy is the only curative treatment for primary hyperparathyroidism. If the parathyroid gland is in the superior mediastinum, a cervical approach with or without sternotomy could be considered. However, in most cases VATS would be preferred.[7,8] Also, radioguided parathyroidectomy via VATS, combined with intraoperative PTH assay, can also be performed for some patients.[8] Advancement in VATS techniques has decreased the need for sternotomy to successfully remove these ectopic glands. If the thoracoscopic removal of ectopic hyperfunctioning parathyroid tissue is to be considered a superior alternative to either median sternotomy, it must offer theoretical advantages. In comparison to median sternotomy, thoracoscopy offers all the benefits of surgical resection plus the potential of a marked decrease in general morbidity and hospital stay.[7,9] In our case, the thoracostomy tube was removed on the first postoperative day, and no surgical complication was observed.

Hyperparathyroidism in pregnancy causing neonatal hypocalcemia is a rare clinical condition. This condition may be related to ectopic parathyroid adenoma. We present our case of a successful thoracoscopic removal of the ectopic hyperfunctioning parathyroid adenoma located in the mediastinum, and suggest that this approach is a viable alternative in patients with this affliction.

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