A Case of a Retroesophageal Parathyroid Adenoma with an Aberrant Right Subclavian Artery: A Potential Surgical Pitfall

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We report a case of retroesophageal parathyroid adenoma coexisting with an aberrant right subclavian artery (ARSA). A 63-year-old female presented with elevated serum calcium and intact parathyroid hormone levels. $^{99}$Tc-methoxyisobutylisonitrile (MIBI) scintigram and enhanced chest computed tomography (CT) revealed a mediastinal tumor measuring $25 \times 15 \times 7$ mm located posterior to the esophagus. Three-dimensional CT provided accurate anatomical location of the tumor and the ARSA. We safely resected the ectopic parathyroid adenoma using video-assisted thoracic surgery owing to the accurate location of the adenoma and its relationship to the surrounding anatomical structures including the ARSA.

Keywords: Ectopic parathyroid adenoma, aberrant right subclavian artery, three-dimensional computed tomography, video-assisted thoracic surgery

Introduction

Adenoma or hyperplasia of the parathyroid glands is the most common cause of primary hyperparathyroidism (PHPT). Surgical resection of hyper-functioning parathyroid glands is the treatment of choice in patients with PHPT. Approximately 25% adenomas are ectopic, and intra-operative identification of ectopic adenomas located deep in the mediastinum is challenging. We report here an extremely rare case of a retroesophageal ectopic parathyroid adenoma coexisting with an aberrant right subclavian artery (ARSA) that was successfully treated using video-assisted thoracic surgery (VATS).

Case Report

A 63-year-old woman underwent a medical check-up two years ago and was diagnosed with osteoporosis and hypercalcemia. Although she was under alendronate sodium hydrate and alfacalcidol therapy, elevated serum calcium (total calcium, 11.5 mg/dL; normal, 8.5 mg/dL–10.5 mg/dL) and intact parathyroid hormone (259 pg/mL; normal range, 11 pg/mL–68 pg/mL) levels persisted. Bone density of the lumbar spine and hip was more than 2.5 standard deviations below peak bone mass (t-score <-2.5). $^{99}$Tc-methoxyisobutylisonitrile (MIBI) scintigram revealed an area of uptake in the upper posterior mediastinum (Fig. 1A). Enhanced chest computed tomography (CT) revealed a well-circumscribed round tumor measuring $25 \times 15 \times 7$ mm that was located posterior to the upper esophagus (Fig. 1B). Simultaneously, pre-operative three-dimensional (3D) CT revealed an ARSA arising as a terminal branch of the aortic arch and

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Retroesophageal Parathyroid Adenoma with ARSA

traversing between the upper esophagus and cervical vertebral column (Fig. 2). No stenotic lesion or aneurysm was observed in the ARSA. Neck ultrasonography and magnetic resonance imaging (MRI) revealed no abnormalities. Abdominal MRI and other hormonal blood examinations did not indicate any evidence of other endocrine neoplasm. The patient was diagnosed with PHPT due to a retroesophageal parathyroid adenoma for which she underwent complete resection using VATS (Fig. 3). The patient was placed in the right lateral decubitus position and intubated with a double-lumen endotracheal tube for single lung ventilation. A 2 cm access port was made at the fourth intercostal space on the anterior axillary line. Three additional stab wounds approximately 2 cm each were also made. The thoracoscope was 10 mm in diameter and had a flexible lens (Olympus, Tokyo, Japan). Except for hypocalcemia detected on the first post-operative day, the post-operative course was uneventful, and the patient was discharged on the third post-operative day. Pathological examination of the tumor revealed a parathyroid adenoma. The patient’s serum calcium levels decreased to 9.2 mg/dL in the fifth post-operative week, and normal serum calcium levels have been maintained two months after surgery.

Discussion

Eighty-five percent of PHPT cases are caused by parathyroid adenomas, and approximately 25% of these adenomas are ectopic.\textsuperscript{1}) Most ectopic parathyroid adenomas of the lower parathyroid gland or supernumerary gland are located in the thymus or generally in the cervical tongue of the thymus in the anterior mediastinum. Ectopic parathyroid glands in the retroesophageal space are rare (3.2%) and are considered to be embryologically derived from the left superior parathyroid gland that descends along the tracheoesophageal groove.\textsuperscript{2,3)} In addition, this patient had an ARSA, which is the most common vascular ring anomaly with a prevalence of 0.5%–2%. We considered that this retroesophageal parathyroid adenoma with an ARSA was an even rarer case. Eighty percent of ARSA cases, the artery traverses between the esophagus and the cervical vertebral column, which is similar to our case. However, in 15% of cases it traverses between the esophagus and the trachea, whereas in 5% it passes anterior to the trachea and esophagus.\textsuperscript{4)}

CT scan images are occasionally equivocal because of the size or location of the parathyroid adenomas. Parathyroid adenomas measuring greater than 15 mm in diameter are usually detected on conventional chest
was useful in planning our surgical approach. 13,14) We dis-

accurate anatomical location of the abnormal artery, which

erative 3D-CT. We previously used 3D-CT to provide an

ical knowledge of the esophagus and ARSA using preop-

The accurate localization of the adenoma and the anatom-

In this case, we could complete the resection owing to

We identified the ectopic adenoma using both 99m Tc-MIBI scintigraphy and con-

vventional chest CT.

VATS has gained more popularity than thoracotomy or

sternotomy because it is less invasive, results in decreased
general morbidity, and requires a shorter hospital stay. Thoracoscopic can provide clearer and more magnified

image than conventional surgery through a thoracotomy.

For ectopic parathyroid adenomas, the most important

intra-operative consideration is its accurate detection.

Although some authors have indicated that radioiso-

to-ge-guided surgery or intravenous methylene blue

administration is effective for identifying tumors, we

selected a thoracoscopic approach without these methods

in this case. The gamma probe is not helpful for localiza-

tion because it is difficult to differentiate nuclear counts

in the parathyroid adenoma from the background activity

of the great vessels and the heart. 7,8) We considered that

application of the gamma probe for the tumor on the

ARSA becomes difficult to detect it or even technically

impossible. Adachi reported successful thoracoscopic resection of mediastinal parathyroid adenomas by using

intraoperative methylene blue administration, but intra-

operative percutaneous oxygen saturation could not be monitored for several minutes after using it. 9) Some studies

reported symptoms of severe mental symptoms leading to
cencephalopathy because of increased central serotonin

transmission and nitric oxide inhibition by the methy-

lene blue.10-12) Careful monitoring is needed to use it.

In this case, we could complete the resection owing to the

accurate localization of the adenoma and the anatomical

knowledge of the esophagus and ARSA using preoperative

3D-CT. We previously used 3D-CT to provide an

accurate anatomical location of the abnormal artery, which

was useful in planning our surgical approach. 13,14) We dis-

sected the mediastinal adenoma without causing injury to the recurrent laryngeal nerve, thoracic duct and most importantly the ARSA. Considering these advantages, we advocate VATS as a safe and less invasive procedure for resecting deep mediastinal parathyroid tumors.

Conclusion

We encountered a rare case of retroesophageal para-

thyroid adenoma coexisting with an ARSA. Pre-operative

3D-CT was extremely useful in determining the anatomical

location of the tumor as well as the ARSA, injury of

which could lead to catastrophic event. We were able to

successfully resect the retroesophageal parathyroid ade-

noma using VATS owing to the accurate localization of

the adenoma using pre-operative chest CT images.

Disclosure Statement

The authors declare that they have no conflicts of interest.

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