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CASE REPORT

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Missing in action

Ademayowa Ademiluyi[a], Nicola Jackson[a], Shion Betty[a], Adedoyin Ademiluyi[b], James Appiah-Pippim[a] and Keisha Bonhomme[a]

[a]Graduate Medical Education, Piedmont Athens Regional Medical Center, Athens, GA, USA; [b]College of Medicine , American University of Antigua College of Medicine, Coolidge, Antigua and Barbuda

ABSTRACT

Ectopically located parathyroid adenoma is one of the major causes of persistent and recurrent hyperparathyroidism and hypercalcemia. Approximately 0.3–8% of parathyroid adenoma is found in an ectopic location. Ectopic parathyroid adenomas are uncommon causes of persistent hypercalcemia and can be present at uncommon locations, including the hypoglossal nerve, the posterior triangle of the neck, axilla, and pericardium [3]. A high index of suspicion is warranted when we see persistently elevated levels of parathyroid hormones (PTHs) and calcium levels post parathyroidectomy. Here, we present a patient who persistently had elevated calcium and PTH levels after parathyroidectomy.

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1. Introduction

Primary hyperparathyroidism is characterized by an elevated parathyroid hormone, which may result in hypercalcemia. The clinical presentation varies widely, but most affected patients are asymptomatic. The incidence of primary hyperparathyroidism varies from 0.4 to 21.6 cases per 100,000 person-years [1]. The incidence is higher in females and patients older than 50 years of age [1]. The parathyroid glands are located on the posterior surface of the thyroid gland. However, they can be located in ectopic locations due to aberrant migration during early development [2]. We present a patient with persistent hypercalcemia after parathyroidectomy who was found to have an ectopic adenoma.

2. Case description

An 82-year-old female was referred to our emergency department by her endocrinologist for further management of persistent hypercalcemia associated with fatigue and generalized weakness. The hypercalcemia was initially diagnosed 7 years prior and was complicated by nephrolithiasis and osteoporosis. A nuclear parathyroid sestamibi scan obtained 1 year before presentation showed no parathyroid adenoma.

In the emergency department, her blood pressure was 127/64 mmHg, heart rate was 88, and temperature of 98.1 F. She was breathing at 20 breaths per minute with an oxygen saturation of 99% on room air. The physical examination was unremarkable. Laboratory studies revealed sodium 137, potassium 3.6, calcium 11.5, creatinine 1.06, alkaline phosphatase 103, phosphorus 2.10, and a parathyroid hormone (PTH) 257.70.

Upon initial presentation, she was managed conservatively with IV fluids, IV zoledronic acid infusion as well as calcitonin 200 units twice daily. Four months later, she had a right superior and left inferior parathyroidectomy. Intraoperatively, the PTHi was 228 pg/ml, which later decreased to 169.40 pg/ml post-operatively Figure 1.

In the subsequent months following the surgery, she was noted to have persistent hypercalcemia with the highest calcium level of 11.3 mg/dl and PTH remaining significantly elevated. MRI of the neck with and without contrast showed a 9 mm enhancing nodule in the right superior mediastinum between the superior vena cava and the brachiocephalic artery as seen in Figure 1.

She was scheduled for partial sternotomy with resection of the mediastinal parathyroid adenoma. However, 2 months later she developed intracerebellar hemorrhages. At that time, goals of care were addressed, and the family decided to pursue hospice.

3. Discussion

Ectopically located parathyroid adenoma is one of the major causes of persistent and recurrent hyperparathyroidism and hypercalcemia. Approximately 0.3–8% of parathyroid adenoma are found in an ectopic location. This can delay the diagnosis of primary hyperparathyroidism. The most frequent location of an ectopic parathyroid gland is the mediastinum, along the path of the

CONTACT Ademayowa Ademiluyi ademayowa.ademiluyi@piedmont.org Graduate Medical Education, Piedmont Athens Regional Medical Center, Athens, GA, USA

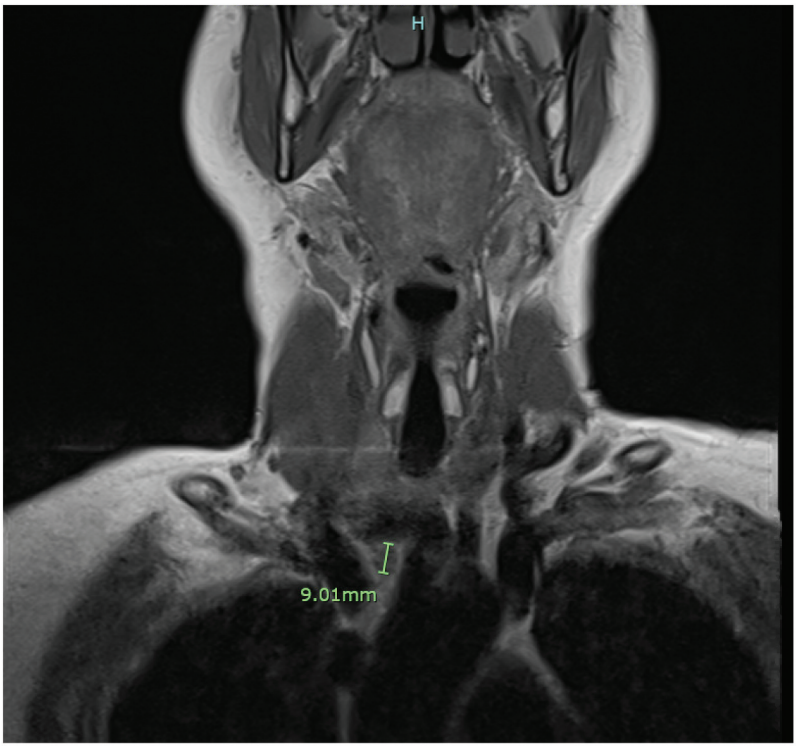

Figure 1. Showing enhancing nodule in the right superior mediastinum between the superior vena cava and brachiocephalic artery.

vagus nerve and recurrent laryngeal nerve and within the thyroid parenchyma. Uncommon locations include the hypoglossal nerve, the posterior triangle of the neck, axilla, and pericardium [3].

The parathyroid glands are divided into two pairs: the upper and lower. The lower pair originate from the dorsal part of the third pharyngeal pouch with the thymus, which can explain their ectopic location in the mediastinum, along the path of the vagus or recurrent laryngeal nerves [3]. The upper pair is often located in the transesophageal groove and retroesophageal region [4]. When the glands present in a normal anatomical location, they may be enlarged and be displaced to an ectopic location because of the lack of capsular fixation [4]. Less frequently, the location of an ectopic parathyroid gland includes the axilla and pericardium [3].

Clinical presentation is similar to other causes of hypercalcemia. However, most patients are asymptomatic. Importantly, an ectopic location does not increase the chance of malignant transformation of the gland [5]. An ectopic adenoma can be removed by a median sternotomy or thoracotomy and also by video-assisted thoracic surgery techniques [6].

There have been case reports that a combination of different imaging modalities can increase the sensitivity of screening to detect ectopic parathyroid adenoma. There was a case report that combined C-methionine PET-CT and tesla simultaneous PET-MRI, which detected the ectopic adenoma behind the left sternoclavicular joint where ultrasonography and sestamibi scan did not show the active mass [7]. The ultimate goal of surgery is cure. There is no role to routinely check PTH levels in patients with normal calcium levels but failure to return to normal after 6 months or longer can point towards early surgical failure, which should prompt evaluation for secondary causes of hyperparathyroidism [8].

4. Conclusion

Consider an ectopic parathyroid adenoma in a patient who has persistent or recurrent hypercalcemia following parathyroidectomy.

Disclosure statement

No potential conflict of interest was reported by the author(s).

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