

Two Cases of Intravagal Parathyroid Adenoma

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Background

- Primary hyperparathyroidism is the most common cause of hypercalcemia in the ambulatory population
- Approximately 75% of primary hyperparathyroidism can be attributed to a single parathyroid adenoma
- Parathyroid tissue can localize almost anywhere in the neck from skull base to mediastinum
- Approximately 16% of parathyroid adenomas are located in ectopic locations
- Multiple different forms of imaging have been developed to help localize parathyroid adenomas preoperatively to guide operative planning
- Intraneural parathyroid glands are extremely rare and particularly challenging to locate
- There are only 30 cases in the literature of intravagal parathyroid adenoma
- The objectives of this study are:
 - To describe two cases of intra-vagal parathyroid adenomas in the post-styloid parapharyngeal space effectively treated with surgery
 - Review the literature regarding this rare clinical entity

Methods

- A retrospective case series of two adult patients with an intravagal parathyroid adenoma treated surgically. Thorough chart review was performed.
- Existing literature was identified via PubMed search and was reviewed and summarized

Case Report

- Patient A is a 35 year-old male with previous history of thyroid cancer, total thyroidectomy, central neck dissection, and radioactive iodine who presented several years later with primary hyperparathyroidism
- Patient B is a 44 year-old female with history of total thyroidectomy for multinodular goiter who presented several years later with primary hyperparathyroidism

Patient	Symptoms at Presentation	Initial PTH	Initial Ca
A	Kidney stones, fatigue	106.5	10.3
B	Kidney stones, constipation, bone pain, brain fog, nocturia	187.2	10.8

Table 1 Characteristics of patient's primary hyperparathyroidism. PTH – parathyroid hormone in pg/mL, normal range 9-77. Ca – blood calcium in mg/dL, normal range 8.5-10.5.

	Patient A	Patient B
Preoperative imaging	Ultrasound, Sestamibi scan, and 4D CT scan	Ultrasound, Sestamibi scan, and 4D CT scan
Localization on imaging?	Not initially, on subsequent	Yes
Surgery	4-gland exploration followed by directed	Directed
Final PTH (pg/mL)	5	11

Table 2 Summary of initial patient work-up and treatment. Patient A underwent soft tissue neck ultrasound, Sestamibi scan which is a nuclear medicine technetium-99 scan used for parathyroid localization, and a four-dimensional CT scan which has an arterial and venous phase during which parathyroid adenomas should enhance. There was no identified adenoma during pre-operative imaging. Patient B underwent both ultrasound and 4D CT scan with left superior adenoma identified as a target at the skull base, Sestamibi scan showed no uptake

- Patient A underwent surgery where ultrasound-guided internal jugular vein (IJV) sampling revealed right>left parathyroid hormone
- Right side exploration showed normocellular right superior parathyroid, no inferior gland identified

Case Report

- Patient A Further jugular venous sampling intraoperatively showed a gradient within the right IJV with highest value in right level II

Patient	Initial IJV Sampling (PTH in pg/mL)		IJV Gradient (PTH in pg/mL)		
	Left	Right	Level II	Level III	Level IV
A	71	736	>2220	736	376
B	2154	119	3240	--	2154

Table 3 Summary of intra-operative internal jugular vein sampling. Patient A had higher levels on the right and level II of the neck. Patient B had higher levels on the left and in level II in the neck

- Repeat CT scan with more superior extent postoperatively showed suspected right parathyroid adenoma at skull base (Figure 1)

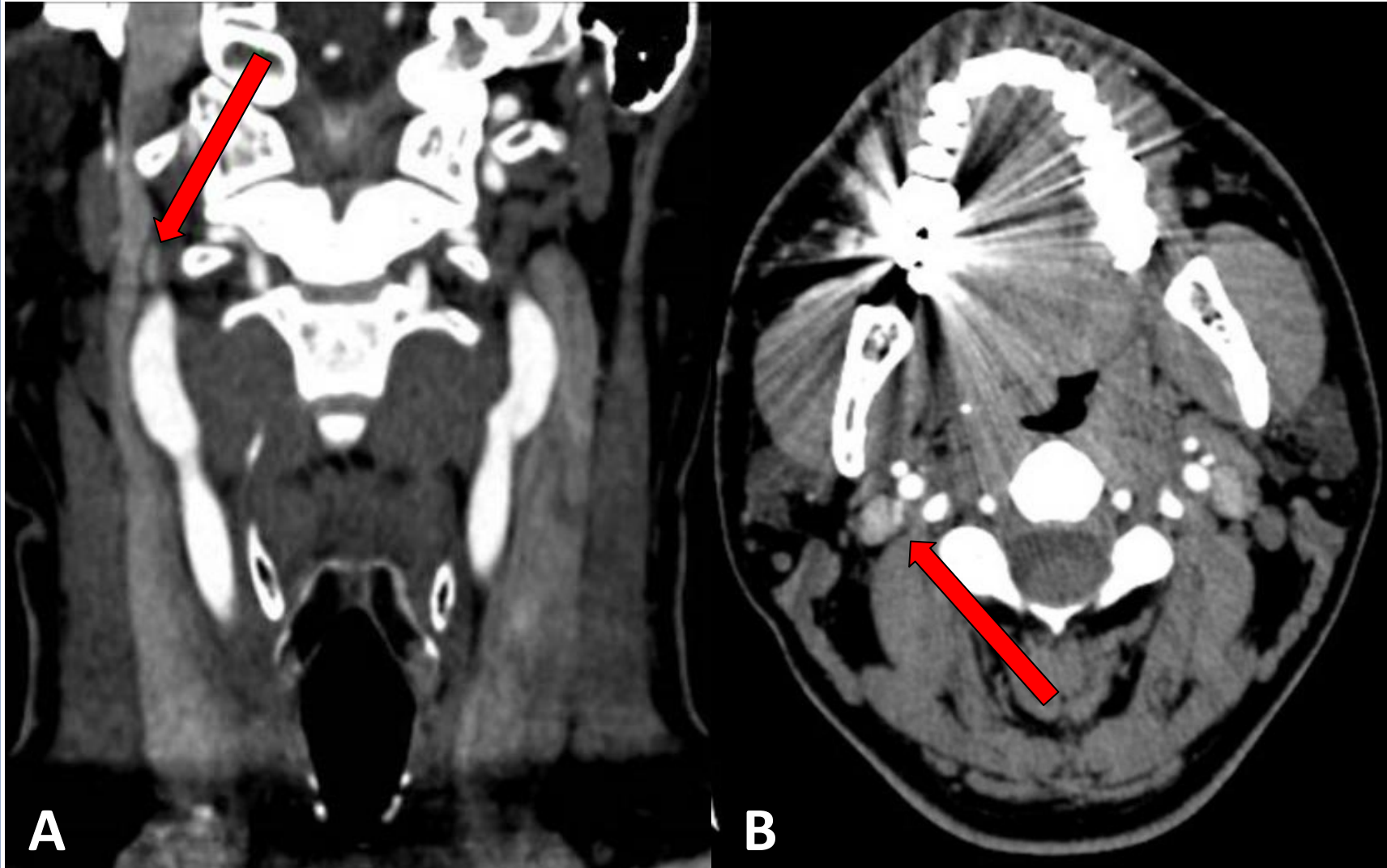


Figure 1. Patient A second 4D CT scan findings. A. Coronal plane imaging with red arrow indicating potential parathyroid adenoma at right skull base. B. Axial plane imaging with red arrow indicating potential parathyroid adenoma at right skull base.

- Patient A underwent surgery and right intravagal parathyroid adenoma was resected with appropriate PTH drop and preserved RLN function
- Patient B had preoperative 4D CT scan with candidate adenoma (Figure 3), intraoperative IJ sampling with L>R PTH
- Intraoperatively, adenoma identified within the vagal nerve sheath (Figure 3) and removed with preserved nerve function and appropriate PTH drop, this was confirmed on pathology (Figure 4)



Figure 2. Patient B 4D CT scan. A. Left candidate parathyroid adenoma at the skull base between carotid artery and internal jugular vein on axial view (red arrow). B. Left candidate parathyroid adenoma at the skull base on coronal view (red arrow)

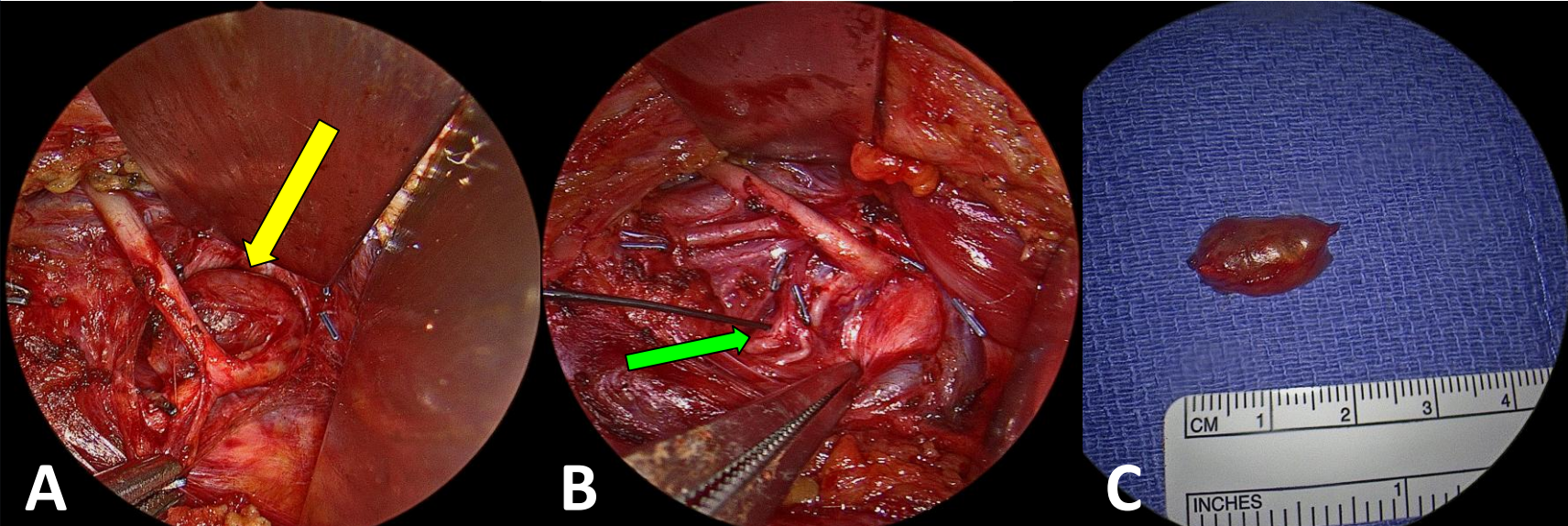


Figure 3. Intra-operative photos from Patient B's surgery. A. Yellow arrow indicating intravagal parathyroid adenoma. B. Green arrow indicating vagus nerve being stimulated, parathyroid adenoma has been removed, nerve stimulated well. C. Excised parathyroid adenoma approximately 2x1cm

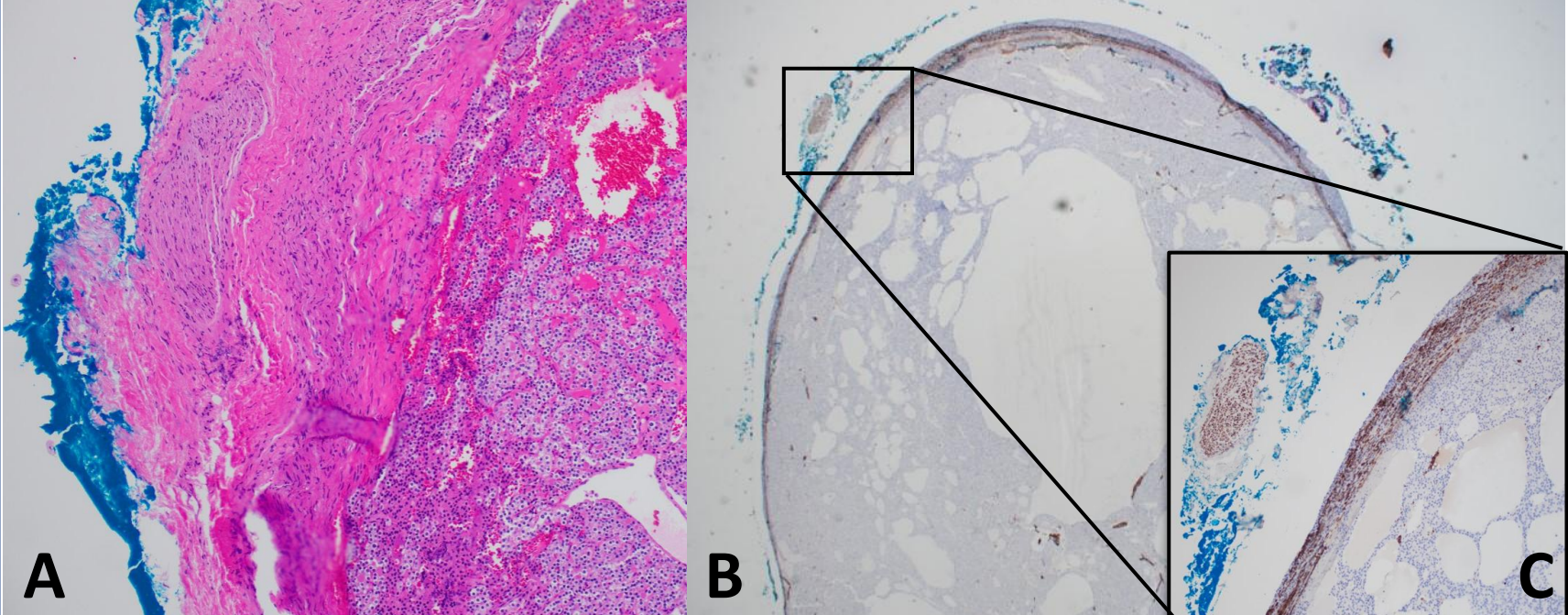


Figure 4. Pathology from Patient B's surgery. A. H&E staining of parathyroid adenoma demonstrating hypercellular parathyroid tissue. B. S100 staining in brown demonstrates nerve tissue completely surrounding the parathyroid adenoma. C. 10x magnification of S100 staining showing neural tissue stained brown on periphery of hypercellular parathyroid tissue

Discussion

In both cases, localization imaging was helpful in identifying ectopic parathyroid adenomas

- For patient A, the initial 4D CT scan did not extend superiorly enough to identify the adenoma at the skull base
- 4D CT scan may be the best imaging modality for identifying ectopic adenomas, while ultrasound has day-of surgery applications. Sestamibi scans can only identify larger adenomas and up to 30% do not have uptake (Gulati 2023)
- For both patients, Sestamibi scan was unable to identify a target parathyroid adenoma which could be secondary to size or for patient B could be due to the cystic nature of the adenoma

Intra-operative internal jugular vein sampling allowed for confirmation of imaging findings and further dissection guidance

- Weber et al 2006 found that for patients undergoing repeat exploration for parathyroidectomy, internal jugular vein sampling had better success rates than noninvasive imaging
- For patient A, IJV sampling is what prompted repeat 4D CT scan with increased superior extent

Intraoperative nerve monitoring and stimulation can be used during surgery to verify nerve position and continued nerve function during dissection

- In both cases described, the patients vagal nerve function was preserved with careful dissection and use of nerve stimulation
- Nerve function was verified both intra-operatively with the use of the nerve stimulator and post-operatively with flexible nasolaryngoscopy

Conclusion

- Intravagal parathyroid adenomas are an extremely rare cause of primary hyperparathyroidism
- Use of multi-modality imaging as well as intraoperative PTH internal jugular vein sampling can help localize these glands
- It is critical for imaging to capture the skull base to the mediastinum
- For intravagal parathyroid adenomas, careful dissection can allow preservation of vagal nerve function

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