

A rare case of an intravagal parathyroid adenoma in the post-styloid parapharyngeal space

Deepa Danan MD, MBA¹, Alexandra Nichols MD², David C. Shonka, Jr. MD¹

¹University of Virginia Health System, Dept. of Otolaryngology/Head and Neck Surgery

²University of Virginia Health System, Dept. of Pathology

ABSTRACT

Objectives: Describe a case of an intravagal parathyroid adenoma including the presentation, diagnosis, radiologic and histologic findings.

Study Design: Case presentation.

Methods: A 35 year old male with a history of papillary thyroid carcinoma status post total thyroidectomy and central neck dissection presented with symptomatic primary hyperparathyroidism. Thyroid ultrasound (US), 4D computed tomography (CT) of the neck, and single photon emission CT (SPECT) did not reveal an adenoma.

Results: The patient was consented for parathyroid exploration. Intraoperative internal jugular venous (IJV) sampling was performed bilaterally under US guidance. Intraoperative parathyroid hormone (ioPTH) was 736 on the right and 71 on the left. Right central neck exploration through the prior thyroidectomy incision was performed, however an adenoma was not identified. ioPTH sampling was performed along the right IJV, which revealed a peak superiorly at the level of the carotid bifurcation. As this area could not be safely explored through the prior thyroidectomy incision, the case was terminated. A repeat 4D CT revealed an enhancing lesion in the right parapharyngeal space at the skull base, in between and posterior to the IJV and internal carotid artery. Subsequent surgical exploration of this area revealed a firm mass within the right vagus nerve. The mass splayed the nerve and was completely encompassed by nerve fibers. Microdissection of the mass out of the nerve allowed preservation of the nerve fibers. Pathological analysis of the mass revealed hypercellular parathyroid encased in normal appearing nerve tissue. There was an appropriate drop in ioPTH and vocal fold function was normal postoperatively.

Conclusions: Intraneural parathyroid adenomas are exceedingly rare entities. The radiologic and histologic findings of an intravagal parathyroid adenoma in the post-styloid parapharyngeal space are described.

CASE

A 35yo male with a history of papillary thyroid carcinoma status post total thyroidectomy, central neck dissection, and postoperative radioactive iodine ablation presented with primary hyperparathyroidism that had been followed for several years. He developed kidney stones and was sent for surgical evaluation. Preoperative imaging including thyroid ultrasound (US), 4-D computed tomography (CT) of the neck, and single-photon emission CT (SPECT, Figure 1), was non-localizing.

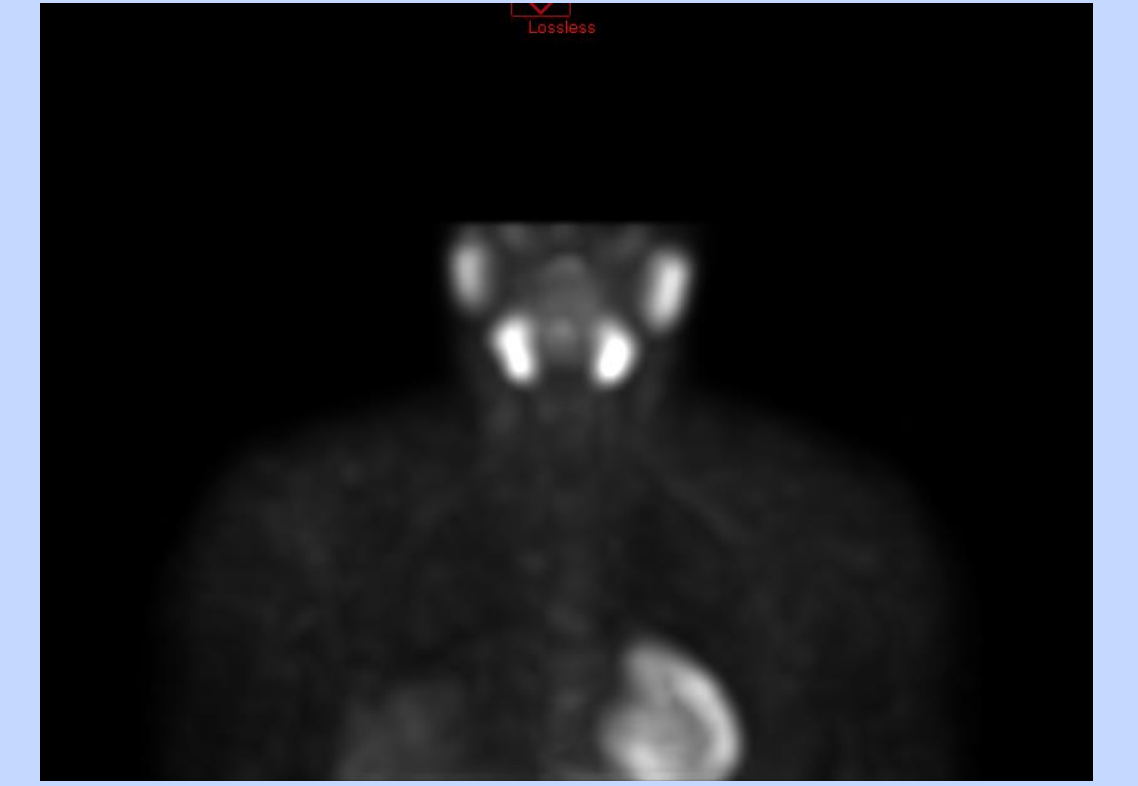


Figure 1. Non-localizing SPECT/CT scan

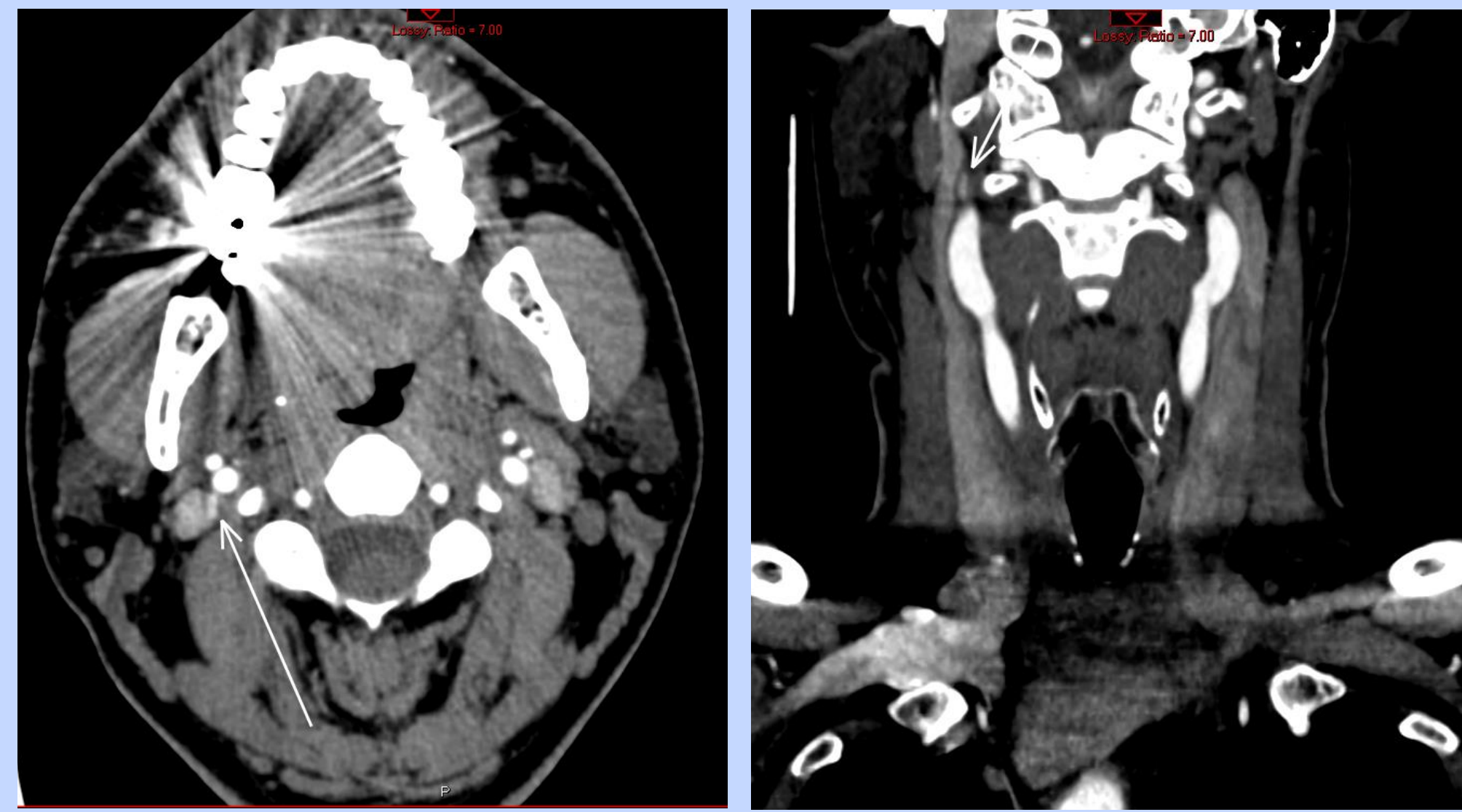


Figure 2a (left) and 2b (right). Axial (2a) and coronal (2b) images revealing an enhancing lesion in the right parapharyngeal space (arrow).

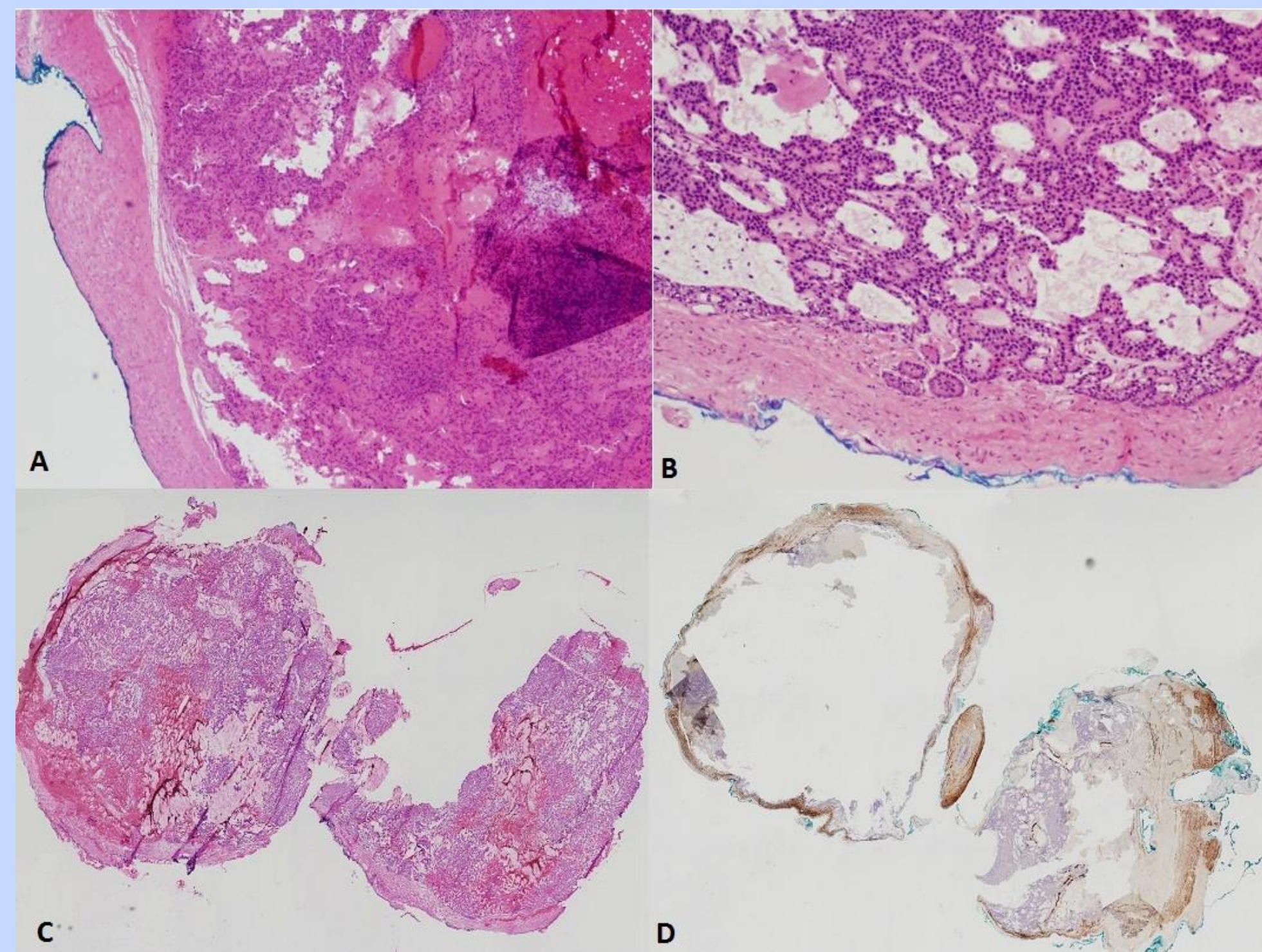


Figure 3. Hematoxylin and eosin stained slides demonstrating a hypercellular parathyroid gland encased by peripheral nerve at 40X (A) and 200X (B), consisting of chief cells arranged in cords and sheets. Hematoxylin and eosin stained frozen section slide at 1X (C) with accompanying immunohistochemical stain for S100 (D), demonstrating positive cytoplasmic staining in the nerve.

RESULTS

The patient was taken to the operating room for parathyroid exploration. Pre-incision parathyroid hormone level was 70 pg/mL. Intraoperative internal jugular venous (IJV) sampling was performed bilaterally under US guidance. Intraoperative parathyroid hormone (ioPTH) was 736 on the right and 71 on the left. Right central neck exploration through the prior thyroidectomy incision was performed, however an adenoma was not identified. A normocellular right superior parathyroid gland was identified. Extensive dissection was unable to identify the right inferior gland in all the usual locations. Further venous sampling on the right IJV showed a PTH gradient with the highest levels in the level II neck (376 level IV, 736 level III, >2220 level II). The operation was terminated as right level II neck could not be adequately explored through the prior thyroidectomy incision.

A 4-D CT was repeated, up to and including the skull base, which was not imaged during the initial CT scan (Figure 2a and 2b). An enhancing lesion was noted in the right parapharyngeal space at the skull base, in between and posterior to the IJV and internal carotid artery. Surgical exploration of this area revealed a firm mass within the right vagus nerve. The mass splayed the nerve and was completely encompassed by nerve fibers. Microdissection of the mass out of the nerve allowed preservation of the nerve fibers. Pathological analysis of the mass revealed hypercellular parathyroid encased in normal appearing nerve tissue (Figure 3). There was an appropriate drop in ioPTH and vocal fold function was normal postoperatively.

DISCUSSION

The inferior parathyroid glands are derived from the third pharyngeal pouch along with the thymus. The thymus and inferior parathyroid glands eventually separate as they migrate together toward the mediastinum. Usually, the inferior parathyroid glands land near the inferior poles of the thyroid, and the thymus continues to migrate toward the mediastinum. However, the location of the inferior parathyroid glands have significantly more variability than the superior parathyroid glands given their migration with the thymus, which descends from the angle of the mandible to the pericardium. As a result, ectopic inferior parathyroid glands can lie anywhere along this course, including the carotid sheath.^{1,2}

Intravagal parathyroid adenomas are exceedingly rare. Review of the literature only yields 10 documented cases.³ The embryology of the vagus nerve has been purported to play a role in this phenomenon: a portion of the vagus nerve is derived from the fourth pharyngeal arch, which is bordered by the third and fourth pharyngeal pouches.^{4,5} The vagus nerve should be considered a possible location for an ectopic or supernumerary parathyroid adenoma when none is found in the usual locations. Localization studies can be utilized, however imaging may need to include more superior cuts than usual. In general, meticulous dissection of the gland from the surrounding nerve fibers will preserve nerve function.⁶

REFERENCES

- Langman J, Sadler TW. Langman's Medical Embryology. 8th ed. Philadelphia, Pa: Lippincott Williams & Wilkins; 2000. 504.
- Wang C. The anatomic basis of parathyroid surgery. *Ann Surg.* 1976 Mar. 183(3):271-5.
- Daruwalla J, Sachithanandan N, Andrews D, Miller JA. Ectopic intravagal parathyroid adenoma. *Head Neck.* 2015 Dec;37(12):E200-4.
- Benson MT, Dalen K, Mancuso AA, Kerr HH, Cacciarelli AA, Mafee MF. Congenital anomalies of the branchial apparatus: embryology and pathologic anatomy. *Radiographics* 1992;12:943-960.
- Lack EE, Delay S, Linnoila RI. Ectopic parathyroid tissue within the vagus nerve. Incidence and possible clinical significance. *Arch Pathol Lab Med* 1988;112:304-306.
- Pawlik TM, Richards M, Giordano TJ, Burney R, Thompson N. Identification and management of intravagal parathyroid adenoma. *World J Surg.* 2001 Apr;25(4):419-23.

CONTACT

Deepa Danan
University of Virginia Health system
Email: dd2na@virginia.edu
Phone: 434-924-5700