

Intravagal Ectopic Parathyroid Presenting as Vocal Cord Paralysis: Case Report and Review of the Literature

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Variability in the location of the parathyroid glands is well recognized. In humans, there are usually four parathyroid glands located in the vicinity of the thyroid gland. These are divided into two superior and two inferior glands.¹ An ectopic or supernumerary parathyroid gland can occur anywhere along the line of descent from the angle of the mandible to the upper border of the pericardium.² Parathyroid tissue has been identified in a variety of locations, including the in the lateral neck, in ectopic thymic tissue, near the hyoid bone, or near the submandibular salivary gland.^{3,4}

Intravagal parathyroid adenomas are rare, and only a small number of cases are reported in the English literature. All reported cases so far presented clinically as hyperparathyroidism. In this report, we present a unique case of ectopic intravagal parathyroid tissue that presented as vocal cord paralysis and normal serum calcium and parathyroid hormone. We also reviewed the available English literature with respect to the incidence and embryogenesis of ectopic parathyroid tissue in the vagus nerve.

Case Report

A 66-year-old male presented with a history of hoarseness for approximately 7 weeks. Flexible endoscopy showed left vocal cord paralysis with no evidence of a laryngeal lesion. A helical computed tomographic (CT) scan showed a soft

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tissue mass at the level of the thyroid and cricoid cartilage suspicious for malignancy. A subsequent CT scan with contrast revealed a mass between the left carotid artery and left jugular vein (Figure 1). Serum calcium, phosphorus, and calcitonin levels were normal. Surgery was undertaken, which showed a lesion involving the left vagus nerve. The lesion was removed along with a portion of the nerve.

The lesion was a partially solid, partially cystic, tan lesion, 3.5 cm in length and 1.5 cm in diameter, with attached portions of nerve on both sides (Figure 2). Microscopically, the lesion was well circumscribed and composed of benign-looking oncocytic and chief parathyroid cells. Cystic degenerative change and evidence of rupture were also evident. The nerve fibres were seen encircling the parathyroid tissue. Nerve fibres and parathyroid tissue were confirmed by immunohistochemical stains (parathyroid hormone and S-100, respectively) (Figure 3). Overall, the features were consistent with an intravagal parathyroid adenoma with cystic degenerative changes.

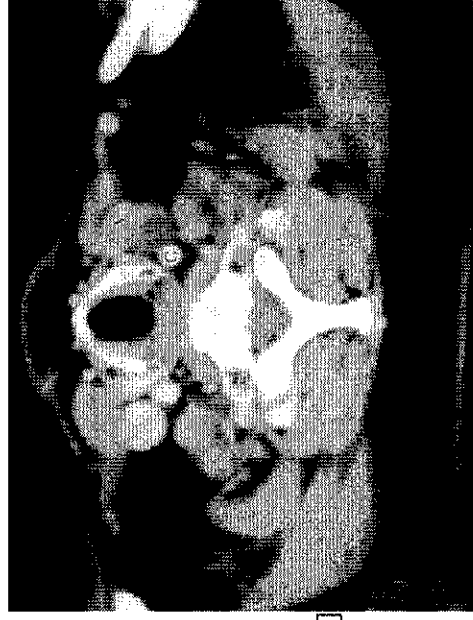


Figure 1. Computed tomographic scan of the parathyroid adenoma. A = parathyroid adenoma inside the vagus nerve; C = carotid artery; V = jugular vein.

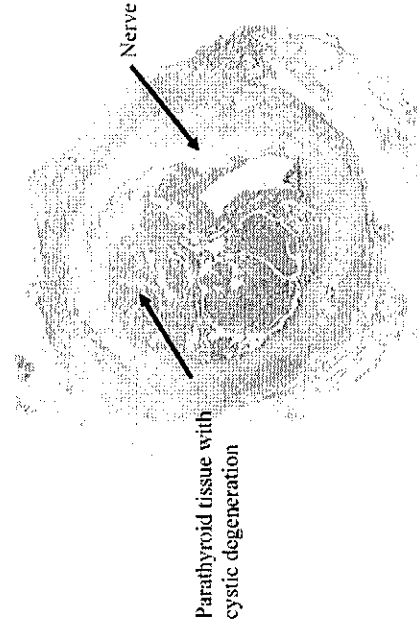


Figure 2. Whole mount of tissue of intravagal parathyroid adenoma (cross section).

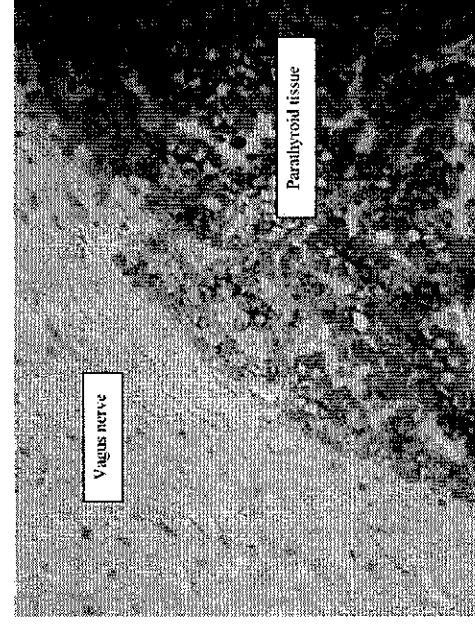


Figure 3. Immunohistochemical staining of the parathyroid tissue. The parathyroid hormone-positive parathyroid tissue is seen at the lower left, and the immunonegative wavy nerve fibres are seen at the top right.

Discussion

The tendency of parathyroid tissue to locate in obscure sites is well recognized. Intravagal parathyroid adenomas are rare. As shown in Table 1, only 10 cases are reported in the English literature. The largest series was that of four cases reported by Chan and colleagues in 2003.⁵ The most common site is around the carotid bifurcation (see Table 1). In our case, an intravagal parathyroid adenoma was present below the carotid bifurcation, at the level of the thyroid gland, and between the common carotid artery and jugular vein.

Persistent or recurrent hyperparathyroidism is the usual clinical presentation in most, if not all, of the cases. All cases showed normalization of the calcium level after removal of the ectopic tissue. The surgical procedure can be carefully done, if anticipated, to dissect the mass of ectopic tissue with minimal or no sacrifice of the nerve fibres.³ To the best of our knowledge, our case represents the first report of ectopic parathyroid tissue with normal serum calcium and parathyroid hormone.

Embryologically, the superior parathyroid glands arise from the fourth pharyngeal pouch and descend with the lateral lobes of the thyroid gland. Most of these glands are located at the cricothyroid junction and the dorsum of the upper pole of the thyroid. Occasionally, these are found in the retropharyngeal or retroesophageal space. The inferior glands arise from the third pharyngeal pouch and descend with the thymus toward the mediastinum. Therefore, these glands are commonly located in the upper parts of the thyroid and the thymic tongue. They are occasionally located in the upper or lateral neck or the mediastinum.

Studies done on stillborn infants and postmortem analysis of parathyroid tissue in infants less than 1 year old showed that the third branchial pouch, which gives rise to the

Table 1. Reported Cases of Intravagal Ectopic Parathyroid Tissue

No.	Location	Size (cm)	Calcium Level	Parathyroid Level	Reference
1	Carotid bifurcation	0.5 × 0.8	High	NA	7
2	Between carotid and internal jugular	1.7 × 2.5	High	High	8
3	C2: adjacent to internal carotid artery and jugular vein	1.2 × 0.8	High	High	9
4	Carotid bifurcation	1.2 × 1.0	High	High	3
5	Carotid bifurcation	1.4 × 1.0	High	High	3
6	Above carotid bifurcation	1.2 × 0.8	High	High	10
7	Above carotid bifurcation	1.3 × 0.8	High	High	5
8	Above carotid bifurcation	0.6 × 0.5	High	High	5
9	Above carotid bifurcation	1.2 × 0.8	High	High	5
10	Below carotid bifurcation	2.0 × 0.8	High	High	5
11	Between carotid and internal jugular	3.5 × 1.5	Normal	Normal	Current report

inferior parathyroid glands, was found to be in direct relation to the ectodermal structures, particularly the vagus nerve.^{2,6} It is hypothesized that during embryogenesis, parathyroid tissue may split off and become embedded in or adjacent to the vagus, leading to ectopic or supernumerary parathyroid tissue.

Interestingly, the pioneer work done by Lack and colleagues showed an unexpectedly high incidence of ectopic parathyroid tissue in 6% of all postmortem sectioned vagus nerves of 32 children up to 1 year of age.² Taken together, these findings emphasize the fact that intravagal adenomas, although rare, do occur. They should be strongly considered in the differential diagnosis, especially when four normal glands are identified in patients with persistent or recurrent hyperparathyroidism. Our report shows also that ectopic parathyroid tissue is not necessarily hormone producing and should be included in the differential diagnosis of mass lesions in the vagus nerve, even without clinical manifestations of hyperparathyroidism. Successful localization of the site of ectopic parathyroid tissue can now be achieved by technetium scanning.

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