

Retroesophageal Right Subclavian Artery with Nonrecurrent Laryngeal Nerve: Unique Variation of Surgical Importance

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ABSTRACT

A nonrecurrent laryngeal nerve (NRLN) is a rare anatomical variant and is not detected on routine preoperative imaging. NRLN is related with absence of the brachiocephalic trunk and aberrant retroesophageal course of the right subclavian artery (arteria lusoria) which is evident on contrast-enhanced computerized tomographic (CT) scan of the chest. We report three cases of NRLN accidentally documented during total thyroidectomies for papillary carcinoma of the thyroid. All the three NRLNs seen on the right side were of the type IB variety. All the three NRLN were seen with its direct branch from the vagus at the level of the isthmus entering the cricothyroid muscle 2.5 to 3 cm away from the vagus without a recurrent course. Reviewing of contrast-enhanced CT scan of the chest revealed retroesophageal right subclavian artery (arteria lusoria) a variation of type I variety was evident in all the three cases. NRLN is a rare anatomical variant and routine preoperative imaging studies are not indicated. The risk of injury to the nerve is nullified by routine dissection and identifying its course along the paratracheal region. NRLN is related with absence of the brachiocephalic trunk and aberrant (mainly retroesophageal) course of the right subclavian artery which is evident on contrast-enhanced CT scan of the chest. Expensive imaging modalities are not warranted to screen a variation which is less than 1%.

Keywords: Right subclavian artery, Nonrecurrent laryngeal nerve, Thyroidectomy, Arteria lusoria, Papillary carcinoma.

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INTRODUCTION

Thyroidectomy procedure done for various pathologies are a common surgical procedure worldwide.¹⁻³ Some common complications of the surgery include bleeding, hypoparathyroidism and recurrent laryngeal nerve (RLN) injury.^{1,4,5} The RLN innervating the muscles of phonation may be damaged unilaterally causing hoarseness and rarely bilaterally causing dyspnea and often life-threatening glottal obstruction.⁶⁻⁸

These nerve damages are more in revision surgeries and advanced thyroid malignancies of the thyroid and vascular

thyroid of Graves disease.^{9,10} All said and done is to identify the nerve during all surgical procedure on thyroid and parathyroid glands to save it.^{11,12} The RLN branches of the vagus then loops under the subclavian artery on the right while under the ligamentum arteriosum on the left innervating all the intrinsic laryngeal muscles except the cricothyroid muscle.¹³

The nerve can be damaged in the neck due to its relative long course in the neck where it maybe damaged in surgeries of the cervical and upper thoracic regions.¹³ Basic information of frequent variations of the nerve along with the nonrecurrent variant reduces the risk of intraoperative injury.¹⁴ A nonrecurrent laryngeal nerve (NRLN) is a rare anatomical variation and routinely checking for it is unwarranted.¹⁴ NRLN is related with absence of the brachiocephalic trunk and aberrant (mainly retroesophageal) course of the right subclavian artery which is evident on contrast-enhanced computerized tomographic (CT) scan of the chest.¹⁴ A smaller hint at the variation can be seen in high resolution sonography of the neck if the retroesophageal vascular variation is present.¹⁵

MATERIALS AND METHODS

We report three cases of NRLN accidentally documented in our institute in patients who underwent total thyroidectomy for suspected malignancies of the thyroid. Two females and one male who were having swelling in the thyroid region since 4 to 6 months were investigated with fine needle aspiration biopsy which reported as papillary carcinoma of the thyroid. The level 2, level 3 lymph nodes were palpated in the two female patients. Sonography of the neck was done and the volume of the thyroid and nodal status was confirmed. The thyroid profile was found to be within normal limits (Table 1).

The thyroglobulin and antithyroglobulin status were found to be elevated. After confirming the vocal cord mobility on laryngoscopically, they were operated under general anesthesia. Total thyroidectomy and functional neck dissection were done in the two female cases and no nodes were palpable in male and so no nodal dissection was done.

Contrast-enhanced CT was done in all the three cases and the nodal status and the great vessel anatomy were

Table 1: Description of the pathology and the type of variation in the three patients

Sr no.	Age	Sex	History	Pathology	Surgery	Type of NRLN	Other RLN	CT chest	Postoperative voice
1	73	F	8 m	Papillary carcinoma thyroid	Total thyroidectomy + (L) FND	IB	(N)	Arteria lusoria (+) type I	(N)
2	19	F	4 m	Papillary carcinoma thyroid	Total thyroidectomy + (L) FND	IB	(N)	Arteria lusoria (+) type I	(N)
3	28	M	5 m	Papillary carcinoma thyroid	Total thyroidectomy	IB	(N)	Arteria lusoria (+) type I	(N)+

F: Female; M: Male

documented in all the three cases. The metastasis of the neck nodes were confirmed on aspiration biopsy. Total thyroidectomy was done with left-sided functional neck dissection in two cases and, while identifying the RLN, we found the NRLN on the right side. All the variation was of type IB variety (Figs 1A and B). No abnormality was seen in the recurrent nerve on the left sides. The postoperative period was uneventful and large dose scan revealed very minimal thyroid tissue and so ablation was done for the same.

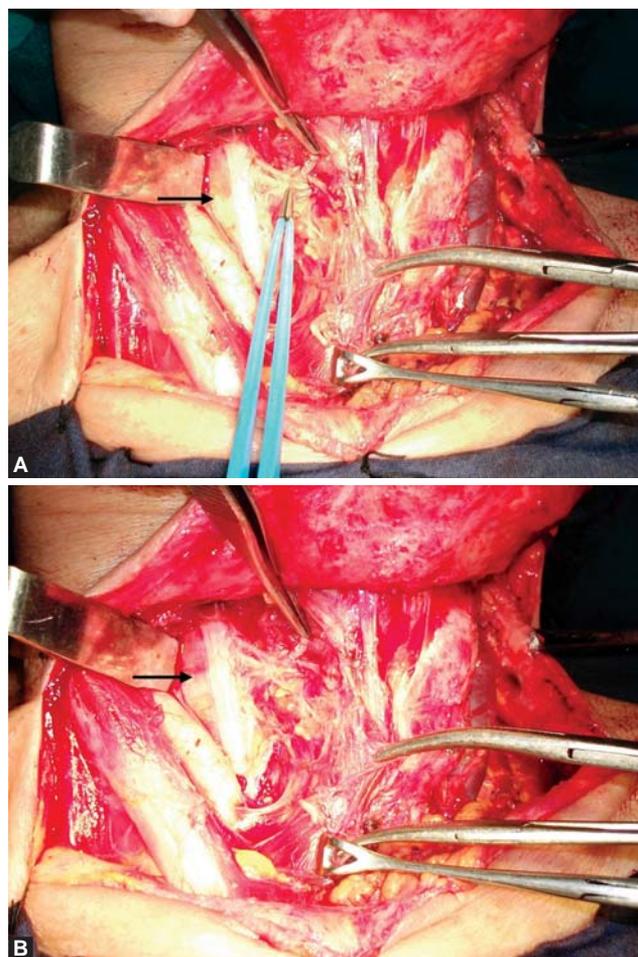
All the three NRLN were seen with its direct branch from the vagus at the level of the level of the isthmus entering the cricothyroid muscle 2.5 to 3 cm away from the vagus without a recurrent course. The contrast chest CT were reviewed and in retroesophageal right subclavian artery (arteria lusoria) a variation of type I variety was evident in all the three cases (Figs 2A and B). The pathology was confirmed as papillary carcinoma of the thyroid in all the three cases.

DISCUSSION

Hunauld in 1735 first described the right aberrant subclavian artery, later in 1794 Bayford demonstrated the first case of retroesophageal right subclavian artery with clinical symptoms of dysphagia.^{16,17} Bayford coined the term 'dysphagia lusoria' to describe the symptomatic vascular anomaly due to the variant vessel.¹⁶ Steadman in 1823 reported the first case of NRLN and its clinical importance during surgery was reported in early 1932 by Pemberton while Berlin reported the left-sided variation in 1935.¹⁷⁻¹⁹ RLN is the nerve of the sixth branchial arch and associated with sixth arch arteries with the ventral branches form the pulmonary arteries.¹⁴ The right subclavian artery and the aortic arch on the left is formed by the fourth arch arteries while the dorsal branches of the fifth and sixth arteries disappear.¹⁴ So, the RLN on the right arches the right subclavian arising from the fourth arch while itself being a nerve of the sixth arch which explains the variation.²⁰

The classical conceptions of the Rathke's scheme describes the double anomaly to be due to unusual evolution of the aortic arches.²¹ Inferior laryngeal nerve is the nerve

of the sixth visceral arches which takes a recurrent course under the distal part of the sixth left and right aortic arches.²² Normally, the fifth left and right aortic arches as well as the distal part of the right sixth aortic arch, would regress and the nerve would follow its recurrent course toward the cricothyroid membrane differently on the right side.²² On the left side, it would pass under the distal part of the sixth arch-ductus arteriosus and under the fourth arch which will form the aortic arch.²² The right subclavian artery is formed from the distal part of the right dorsal aorta and from the seventh segmental artery.²³ Arising from the left part of the aortic arch, it would usually reach the right upper limb by a



Figs 1A and B: Type IB nonrecurrent laryngeal seen on the right side

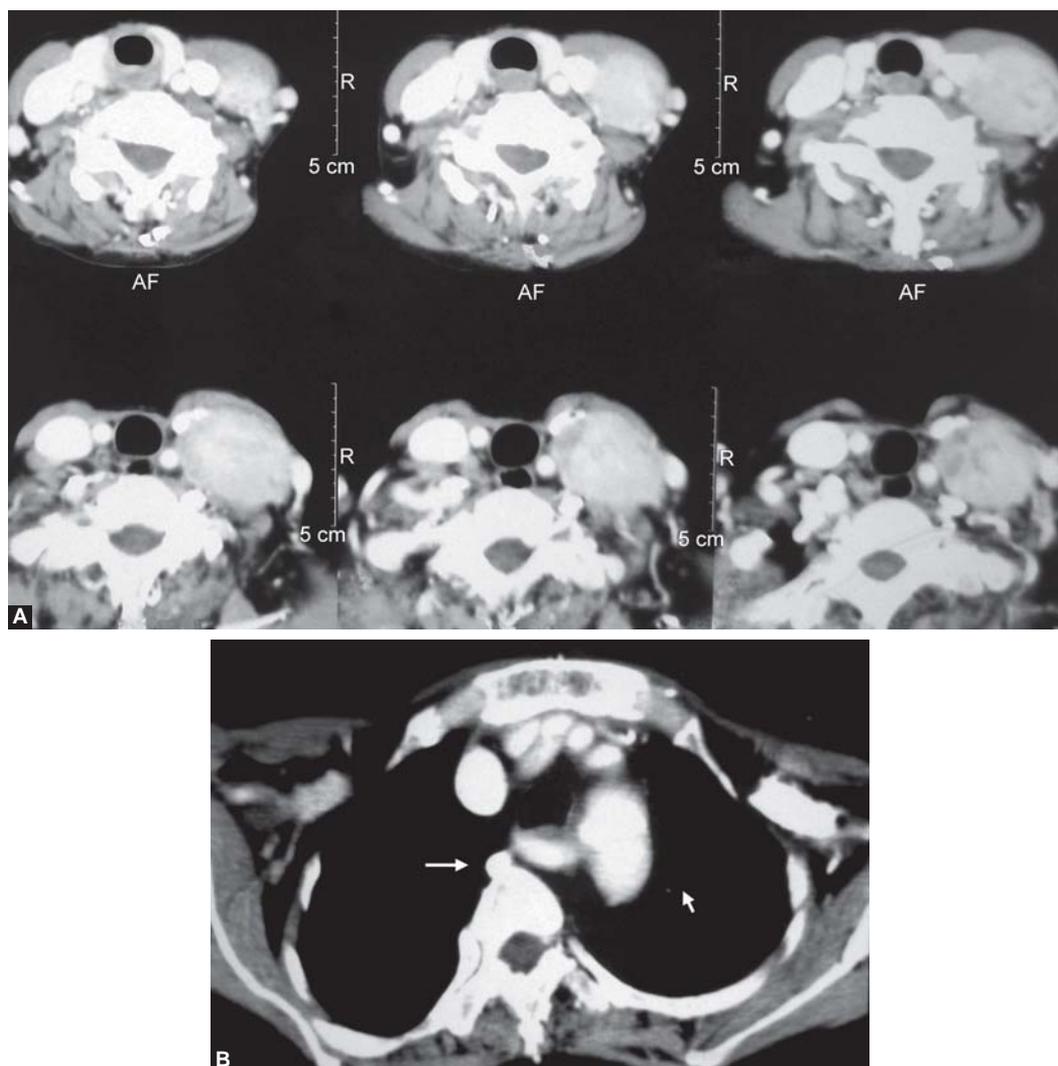
retrosophageal course, but could sometimes be inter-tracheoesophageal, exceptionally pretracheal.²³

Balaji et al found arteria lusoria in 0.5 to 1.8% of the population and described it as a common variation.²⁴ The dysphagia is classical, but not severe, yielding to nutritional management mainly due not only to the arterial compression but also to the existence of an associated tracheomalacia, so that they tend to a spontaneous amelioration.²³ An arteria lusoria is often discovered incidentally during a systematic scanning, or during assessment or treatment for another malformation, such as persistent ductus arteriosus or aortic coarctation.²³ It may also be associated with another visceral malformation.²³ Variant right subclavian may traverse behind the esophagus in 85% cases (type I), between the esophagus and trachea in 15% (type II) and in front of the trachea in 5% of the cases (type III).¹⁴⁻¹⁶

Three types of NRLN have been described in literature, Type IA: the nerve has a straight course at the level of superior thyroid pedicle which may be damaged during upper pole ligation; while type IB: (most common) the nerve

runs transversely at the level of isthmus.²⁵ In type II, the nerve has a downward course and loops upward before reaching tracheoesophageal groove which can be damaged while ligating the inferior thyroid vessels.²⁵ True NRLN are associated with arteria lusoria on the right, and the nerve should be traced to its attachment to the vagus as a branch of the sympathetic trunk as part of the sympathetic-recurrent laryngeal anastomotic branches mimics the same.²⁶

NRLN have a variable incidence ranging from 0 to 3.9%.^{27,28} Henry et al reported an incidence of 0.54% (17 cases in 3,098) on the right side and 0.07% on the left (two cases in 2,846) when compared to the worldwide incidence of 0.32%.²⁹ The common consensus on the anomaly is the presence of the vascular defect called arteria lusoria where the fourth right aortic arch is abnormally absorbed and the vessel fails to drag the right RLN caudally when the cardia descends and neck elongates during embryonic development.^{22,30} The incidence of arteria lusoria globally is 0.5 to 2% and is defined as the right subclavian artery arising as a branch of normal aortic arch and passing



Figs 2A and B: Contrast enhanced CT chest shows retrosophageal variant of aberrant subclavian artery (arteria lusoria)

upward to the right behind the esophagus.³¹ The anomaly is usually asymptomatic but sometimes presents as dysphagia called as dysphagia lusoria due to arterial tortuosity or premature arteriosclerosis and a rarely an aneurysm is seen.³¹ On the left side, NRLN is seen in cases of dextrocardia.³¹

Commonly, NRLN is diagnosed first and arteria lusoria is rarely suspected in cases of dysphagia.³² Preoperative diagnosis of a NRLN is difficult using CT scan or magnetic resonance imaging (MRI) but visualizing the coexisting arterial anomaly arteria lusoria gives a lead to possible diagnosis.³³ Screening for dysphagia using barium esophagogram may denote a possible arteria lusoria indirectly, but should be confirmed by multiplanar MRI angiography where the anatomy of this vascular variation, especially its origin in comparison with other vessels and its course in relation to the esophagus and trachea is defined.³⁴

The anatomical variations of RLN have been associated with iatrogenic nerve injuries especially NRLN but no added reports have confirmed that the preoperative diagnosis of arteria lusoria have reduced the risks during surgery.³⁵ Incidences of RLN injuries during thyroid surgeries worldwide is around 1 to 2% and the total thyroidectomies have replaced other conservative procedures for thyroid malignancies.³⁶ The nerve injuries are minimal with expertize and less with capsular dissection for benign diseases compared to lateral dissection for malignant diseases.³⁶

The nerve injury manifests as irregular hoarseness and can be better avoided by identifying and carefully tracing the path of the recurrent nerve.³⁶ Factors affecting the injuries are surgeon's expertize, histopathologic diagnosis, previous thyroid surgeries, technique adapted and anatomical variations.³⁷ The nerve can be injured in various ways like partial transection, traction, rough handling, contusion, crush, burn, clamping, misplaced ligature and compromised blood supply.^{36,37} The vocal folds do not approximate each other in unilateral injuries and traction injuries to the axons lasts for 6 months while transaction of the nerve due to cutting ligature or cauterization lasts more than 6 months.^{36,37}

Bilateral RLN injuries leads to severe airway compromise warranting an emergency tracheostomy.³⁶ The upper two tracheal rings is the area where the RLN is frequently damaged as the nerve is close to the thyroid lobe in the area of the Berry's ligament.³⁸ Dissection of the RLN and preserving it all along the course is advised even no significant reduction in damage rates has been reported by doing it.³⁶ Fahri et al reported ultrasonography as a very reliable tool in preoperative assessment to identify vascular

anomaly associated with NRLN to make optimal surgical decisions against nerve damage in patients undergoing thyroidectomy or parathyroidectomy.³⁹

Hazem et al reported favorable results with complete dissection of the nerve and rationalized it as 'total dissection of the recurrent nerve over its entire cervical course precludes an incorrect alignment.'³⁶ They reported 7.6% RLN injury with nerve not dissected and significantly lesser with complete dissection where verifying its anatomic integrity and extralaryngeal ramifications is possible.³⁶ The RLN injury rates worldwide on complete dissection and identification is 0 to 2.1%, while in revision cases its higher up to 2 to 12% and marginally higher if the nerve is not dissected at all.³⁶

The use of nerve monitoring devices during surgery detecting vocal cord movement when the RLN is stimulated has not decreased the RLN injury rates as it is already less with meticulous dissection.⁴⁰ RLN injury are higher during thyroidectomies for thyroid carcinoma, hyperthyroid (toxic) goiter and recurrent goiter cases as nerve identification and hemostasis are difficult here.⁴⁰ Postoperative adhesions, anatomical displacements and neovascularization of the gland makes the nerve more vulnerable in revision surgeries.⁴⁰ Echternach et al reported more intubation related laryngeal injuries than due to nerve injuries of larynx during thyroid surgeries.³⁵ Nerve identification during surgery reduces the risk of its damage and intraparenchymal dissection and subtotal excision has no role in present day thyroid surgeries.³⁵

CONCLUSION

NRLN is a rare anatomical variant and routine preoperative imaging studies are not indicated. The risk of injury to the nerve is nullified by routine dissection and identifying its course along the paratracheal region. NRLN is related with absence of the brachiocephalic trunk and aberrant (mainly retroesophageal) course of the right subclavian artery which is evident on contrast-enhanced CT scan of the chest. Expensive imaging modalities are not warranted to screen a variation which is less than 1%.

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