



Right Non-recurrent Inferior Laryngeal Nerve Discovered During Carotid Endarterectomy: A Case Report and Literature Review

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Abstract: The recurrent inferior laryngeal nerve (RILN) originates from the vagus trunk and, recurring into the mediastinum, courses then into the larynx. Sometimes this nerve can assume an unusual pathway, coursing directly into the larynx. This anomaly is the so-called non-recurrent inferior laryngeal nerve (NRILN) and represents a rare entity, with an incidence between 0.3% and 1.6%. It is commonly caused by an embryologic anomaly of the aortic branches and, for this reason, it occurs most on the right and is closely associated with an aberrant subclavian artery (also named *arteria lusoria*). The peculiar anatomy of this nerve must be well-known by surgeons, in order to prevent accidental injuries and avoid post-operative complications, such as vocal cords paralysis. There are other anomalies of cervical nerves (i.e. sympathetic-inferior laryngeal nerve anastomotic branches, SILAB) that simulate the NRILN and it is of paramount importance for the surgeon to distinguish them. In this article, we present the case of a patient undergoing a carotid endarterectomy (CEA), in which a right NRILN was found intraoperatively. We found that this NRILN was associated with no right aberrant subclavian artery, but with an anomalous origin of the brachio-cephalic trunk. Key points in differential diagnosis, embryological origin and surgical implications are discussed.

Keywords: Non-recurrent Laryngeal Nerve, Carotid Endarterectomy, Aberrant Subclavian Artery, Cranial Nerves Injury, Neurological Complications

1. Introduction

The RILN is a branch of the vagus nerve, that supplies the intrinsic muscles of the larynx, with the exception of the cricothyroid muscles. This nerve is not symmetrical, as the left nerve branches off the vagus at the aortic arch and wraps under it, while the right one originates at the right subclavian artery and loops under it. Then they both ascend in the mediastinum and run alongside the trachea, until they reach the larynx.

Due to an embryologic anomaly of the aortic arch, sometimes the RILN runs an unusual pathway, coursing directly into the larynx, and is closely associated with an aberrant subclavian artery (also named *arteria lusoria*). This

anomaly, known as the non-recurrence of the RILN, is quite rare, having an incidence about 0.3-1.6% [1] and in most of cases occurs on the right side. Left NRILN is rarer [2-3-4], with an incidence of 0.04%, since it is related to *situs viscerum inversus*. The presence of a NRILN is a dangerous pitfall during every type of neck surgery, despite it is easier to be discovered during thyroid and parathyroid surgery. The peculiar anatomy of this nerve must be well-known by surgeons, in order to prevent accidental injuries and avoid post-operative complications, such as vocal cords paralysis.

We present a case of a right NRILN discovered during carotid endarterectomy (CEA). The surgical importance of recognizing this anomaly and differentiating it from other similar conditions, such as sympathetic-inferior laryngeal nerve anastomotic branches (SILAB), are discussed.

2. Case Presentation

A man of 74 presented to us with transient ischemic attacks (TIAs) and hypoesthesia of the mouth and left hand proved to have a hemodynamically significant (80%) right ICA stenosis by Color Doppler ultrasonography and CT angiography with a brachio-cephalic trunk originating close to left CCA (Figure 1).

The patient was therefore admitted for CEA. Before surgery, he underwent a neurological and an otorhinolaryngologic examination, revealing no neurological damage but a reduced left arytenoid motility of unknown origin. Pre-operative blood tests, chest X-ray and electrocardiogram were within normal limits.

Under general anesthesia, the surgical team proceeded to a right-sided CEA, using the standard surgical technique and monitoring the patient's cerebral activity with somato-sensory evoked potentials. After isolating the carotid artery bifurcation and identification of the vagus and hypoglossal nerves in their normal location, we found a nervous structure crossing the carotid bifurcation arising from the vagus nerve and passing to the larynx (FIGURE 2), which we believe to be the NRILN.

A CEA was performed with primary closure of the artery. The somato-sensory evoked potentials were stable, hence, no shunt was used. The patient's post-operative course was uneventful and he was discharged from the hospital three days later.

After six months, the patient did not develop complications and enjoyed good health.

3. Discussion

In 1823, Stedman [5] described first the case of a right NRILN during a cadaver dissection, identifying the path of this nerve as emerging from the vagus and coursing directly into the larynx. He also found an aberrant right subclavian artery, arising to the left part of the aortic arch and passing posterior to the esophagus. Other Authors [6-7] described then similar cases of a NRILN with an atypical right subclavian artery in cadavers, but this anatomic anomaly did not draw the attention of surgeons until 1932, when Pemberton and Beaver [8] discovered it during cervical surgery. Since then, it has always been clear the effective importance and function of this nerve, and thus its surgical implications.

Nowadays there are several reports in literature of NRILNs, but most of all in thyroid and parathyroid surgery. A NRILN has been described just in a few cases during carotid surgery [2-9]. The overall incidence of this condition is about 0.3-1.6% [1], with most occurring on the right side. Left NRILN is rarer [2-3-4], with an incidence of 0.04%, since it is related to *situs viscerum inversus*. The NRILN assumes different pathways, that had been historically classified on the basis of their relationship with the thyroid gland and thyroid arteries (FIGURE 3). The most used is Avisse's [10] classification, that divides the course of NRILN in two types: in type 1

NRILN originates in correspondence to the upper thyroid pole and follows the superior thyroid artery, while in type 2 NRILN makes a small curve downward, running parallel to the inferior thyroid artery and then ascending into the larynx. Toniato et al. [1] divided then type 2 in two different subtypes: in type IIA the NRILN runs over the inferior thyroid artery, while in type IIB, it passes between the branches of the artery. Type IIA seems to represent the major part of cases reported in literature.

The nonrecurrence of the inferior laryngeal nerve has its primary basis in embryological development, as the NRILN is commonly supposed to be caused by an anomaly of the aortic arches. When the heart descends in the embryo, the inferior laryngeal nerves assume their recurrent course, being wrapped around the sixth arches bilaterally. Then, on the right the sixth arch disappears and so the inferior laryngeal nerve ascends till the fourth aortic arch, that will become the right subclavian artery. If also the fourth aortic arch is absent, the inferior laryngeal nerve is free to reach the larynx directly, resulting in an NRILN. On the left, this is not possible, as the sixth arch remains until birth as the ductus Botallii [11]. This is the reason why the NRILN is more frequent on the right side, as a left NRILN is feasible only in case of dextrocardia. This also explains the tight association between this anatomic anomaly and the absence of brachio-cephalic trunk and an aberrant right subclavian artery. In these patients in fact, the right subclavian artery originates in the left part of the aortic arch and, in its path toward the right arm, enters the mediastinum passing to the back of esophagus (80%). It can also run between trachea and esophagus (15%) and even ahead of trachea (5%) [12].

Most of times this condition is asymptomatic, but in 10% of cases it manifests itself with dysphagia by esophageal extrinsic compression. The gold standard technique in identifying this anomaly is CTA, but sometimes it may be suspected on the basis of the so-called bayonet sign, an indentation on the posterior esophageal wall during the barium swallow. A diagnosis can be made also during an esophagogastroduodenoscopy, by a pulsation in the posterior wall of esophagus. It is likely to discover this anomaly during carotid surgery, as CTA often enters pre-operative exams and can easily point it out as an incidental finding. In thyroid and parathyroid surgery indeed, it is not an ordinary practice ordering CTA, unless in case of thyroid or parathyroid cancer or intrathoracic goiters. It is a common opinion however, that it is not cost-effective undergoing CTA to predict an aberrant subclavian artery, posing the suspect of a NRILN.

Concerning our patient, we can conclude that he corresponds to a type I NRILN and it is to notice that this case of NRILN is not associated with the so-called *arteria lusoria*, but with another vascular anomaly involving the aortic branches. He presented an aberrant brachio-cephalic trunk, that originated in the left part of the aortic arch, with a common origin with the left CCA. This anomaly is described here for the first time related to a NRILN. Traditionally, literature supports the fact that the NRILN is always associated with an aberrant subclavian artery, but our patient

did not present an *arteria lusoria*.

Kato et al. [13] reported the case of a NRILN associated with an anomalous vertebral artery, that originated from the subclavian artery in the same position of the CCA and ascended medial to it. Moreover, Kobayashi et al. [14] claim that these patients tend to have multiple vascular anomalies and both a major risk of ischemic vascular disease. They even guess that an accidental finding of an *arteria lusoria* should lead to subsequent investigations so as to prevent unexpected life-threatening events.

More recently, the dogma of the association between NRILN and an aberrant right subclavian artery has been questioned. An NRILN without vascular anomaly as a genuine entity has been described in a few cases [3-15-16], despite there is still no appropriate anatomical and embryological explanation. Some other authors [17] consider it the so-called “false NRILN”, being in fact an anastomotic trunk between sympathetic cervical nerves and a thin recurrent ILN. It is well-known that there are some nerves connecting the sympathetic cervical ganglions with laryngeal nerves. In general sympathetic-inferior laryngeal nerve anastomotic branches (SILAB) are very thin or plexiform, but sometimes they can reach such a dimension, misinterpreted as NRILN. These anastomotic branches frequently arise from the middle cervical sympathetic ganglion, less frequently from the inferior and superior ganglion, from the stellate ganglion or directly from the sympathetic trunk [18-19]. SILAB represents a rare anatomic variant, having an incidence of 1.5% on right in surgical reports [20], however more frequent than NRILN. As a matter of a fact, in four of the five cases reported of a NRILN without vascular anomalies, the presumed NRILN was accompanied by an ipsilateral RILN, so it is possible that SILAB has been mistaken with a more rare case of NRILN (TABLE 1). In support of this, also in the majority of cases in which the NRILN was on the left side, the patient did not present dextrocardia. The real matter in distinguish NRILN from SILAB however is whether they have a different function, namely if SILAB is as well involved in vocal cords mobility. Although the sympathetic nerves influence the glandular secretion and the vascularisation of vocal cords, it

has not still been confirmed if they have a direct effect on their motility.

Another structure that can also be confused with the NRILN is the so-called Galen nerve. This nerve is composed by the anastomosis between a RILN and an external branch of the superior laryngeal nerve. They can be distinguished because the Galen nerve courses more vertical when compared to NRILN [21].

Although NRILN has mostly been recognized during thyroid and parathyroid surgery, this topic is similarly an important issue during carotid surgery, in order to avoid accidental damages to vocal cords motility. In particular, type I NRILN (as our patient's one) may represent the principal type involved during CEA, because of its pathway nearer to the CCA bifurcation. Furthermore, since at this time we cannot definitely differentiate SILAB from NRILN and the accurate role of the sympathetic anastomotic branches has not been clarified yet, it is as much important recognizing this condition too.

As a suggestion for further research, it would be interesting to investigate whether the incidence of RILN injuries reported during carotid surgery is most frequent on the right side. In this case, the damage of an unknown NRILN as the principal cause of vocal cords paralysis and hoarseness may be confirmed, according to our hypothesis.

4. Conclusions

It is of paramount importance for surgeons to master every anatomical variant of the neck, both vascular and neurological, in every type of surgery. Vascular anomalies can be seen in pre-operative CTA and predict nervous ones. NRILN and SILAB are different anomalies that probably could lead both to impairment of laryngeal cord motility. As a strong recommendation, it is prudent to preserve any structure coursing transversely between carotid sheaths and thyroid. If we encounter a suspicious nerve, this should be dissected so as to find out if it originates from the vagus or the sympathetic ganglion. Careful handling of tissues and a atraumatic surgical technique with sharp dissection close to the arterial wall would lessen any risk of complications.

Table 1. Key points in differentiating NRILN from SILAB and Galen nerve.

	NRILN	SILAB	GALEN NERVE
<i>Origin</i>	vagus	sympathetic ganglions	anastomotic branch between inferior and superior laryngeal nerves
<i>Nerve diameter</i>	thicker	thinner, sometimes plexiform	thinner
<i>Nerve trajectory</i>	usually horizontal	more variable	usually more vertical
<i>Branching to other tissues</i>	rare	more frequent	none
<i>Vascular anomalies</i>	yes	not associated	not associated
<i>Damage in case of injury</i>	vocal cords paralysis, with respiratory distress	(Horner syndrome?)	none important known

Abbreviations: CEA carotid endarterectomy; NRILN non-recurrent inferior laryngeal nerve; RILN recurrent inferior laryngeal nerve; SILAB sympathetic-inferior laryngeal nerve anastomotic branches; CCA common carotid artery; ICA internal carotid artery; ECA external carotid artery; CTA computed tomography angiography.

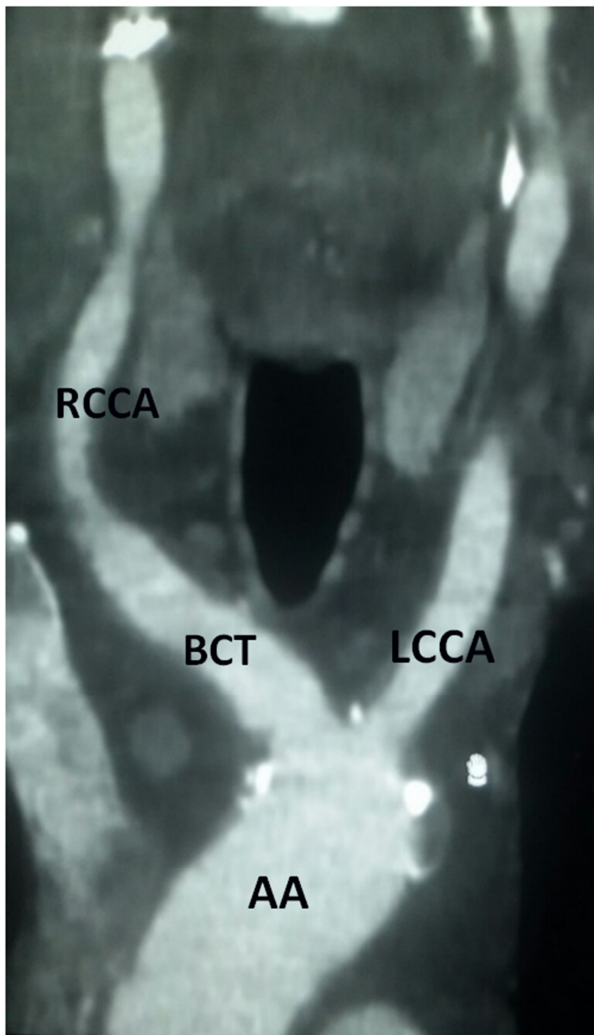


Figure 1. Patient pre-operative CT angiography, that depicts no right aberrant subclavian artery but an anomalous origin of the brachio-cephalic trunk.

AA aortic arch – BCT brachio-cephalic trunk – LCCA left common carotid artery – RCCA right common carotid artery.

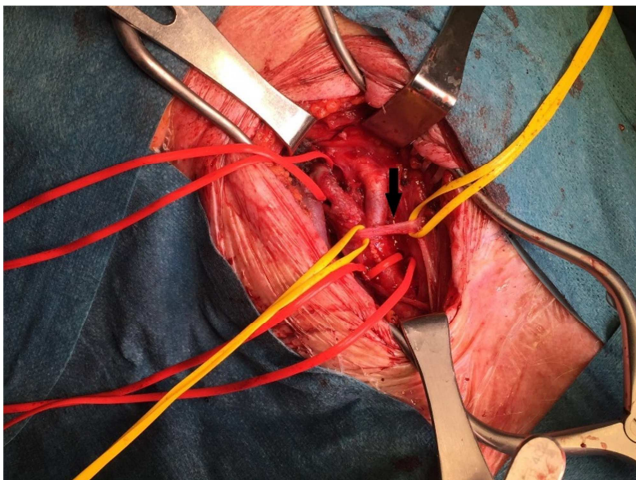


Figure 2. Intraoperative photograph of a NRILN (type I), during right CEA. An arrow indicates the anomalous nerve.

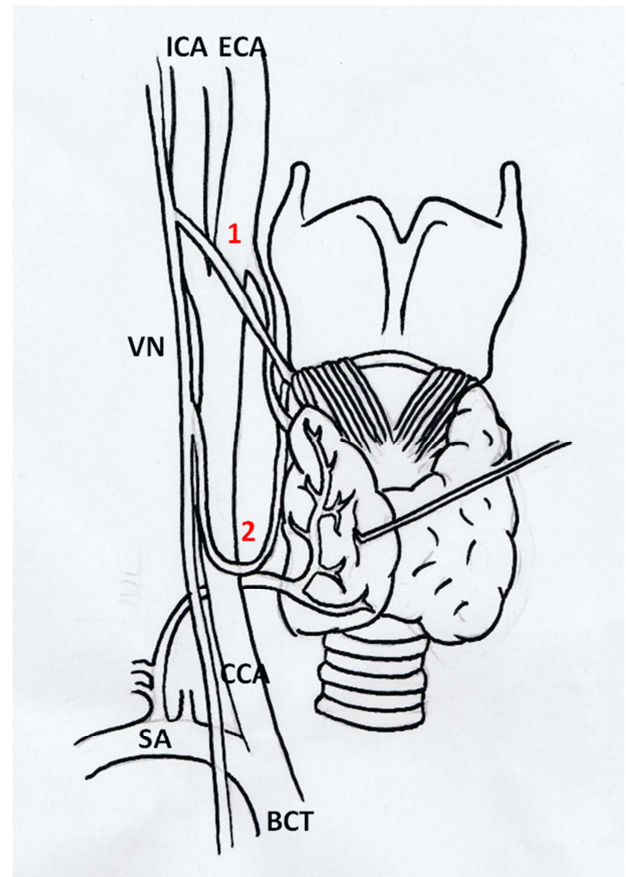


Figure 3. Classification of NRILN (draw by Deborah Ongaro).

BCT brachio-cephalic trunk - SA subclavian artery - CCA common carotid artery - ICA internal carotid artery - ECA external carotid artery – VN vagus nerve.

1 NRILN type 1 – 2 NRILN type 2.

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