

Prevalence of the Right Nonrecurrent Inferior Laryngeal Nerve in a Series of 100 Total Thyroidectomies

Prevalencia del Nervio Laríngeo Inferior Derecho no Recurrente en una Serie de 100 Tiroidectomías Totales

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SUMMARY: The right nonrecurrent inferior laryngeal nerve (NRILN) is a rare occurrence generally associated with an aberrant right subclavian artery. Its prevalence ranges from 0.3 to 1.8 %. It is found mainly in thyroid surgeries, the most frequently performed cervical surgeries. This neural anomaly is almost never diagnosed preoperatively. Dysphagia may be a warning symptom, sometimes being incorrectly related to esophageal compression due to a goiter or thyroid cancer. The postoperative diagnosis of an accompanying aberrant right subclavian artery should be done to confirm the clinical picture and inform the patient of any possible future medical/surgical procedures. The aim of this work is to determine the prevalence of the NRILN in patients undergoing total thyroidectomy in two reference centers for head and neck surgery in Paraguay. Prospective cross-sectional study in a series of 100 consecutive total thyroidectomies in the INCAN and the ORL Service in the Hospital Central of the IPS. 100 patients underwent a total thyroidectomy, 90 of whom were women. The average age was 47 years. 6 % also underwent a neck dissection for thyroid cancer. The preoperative diagnosis was multinodular goiter (MNG) in 84 cases and thyroid cancer in the remaining 16 (16 %). In one man aged 47 years (1 %) operated on for MNG and presenting slight to solid dysphagia, there was difficulty finding the right NRILN. It was located at the level of the lower edge of the inferior pharyngeal constrictor and its downward anomalous course was proven. Postoperatively, after confirmation of the diagnosis that the thyroid pathology was benign, a color echo-Doppler and a CT angiography corroborated the diagnosis of an associated aberrant right subclavian artery. Given the difficulty in locating the recurrent inferior laryngeal nerve in thyroid surgery, it is advisable to consider the possibility that it may be nonrecurrent and should be looked for at the level of its entrance to the larynx below the inferior constrictor. In such cases it is recommended that the patient be examined postoperatively to rule out an associated vascular anomaly. A tomography study of each patient with a thyroid pathology and dysphagia could contribute to the diagnosis of the vascular anomaly and maximize precautions in the nerve dissection, including use of a neurostimulator. The NRILN is a rare entity. In this case study, it represents 1 % of the patients operated on for thyroid pathology in the INCAN and IPS in a 1-year period (July 2016 - June 2017).

KEY WORDS: Right nonrecurrent inferior laryngeal nerve; Thyroidectomy; Neck.

INTRODUCTION

The inferior laryngeal nerve (ILN), a motor and sensory branch of the tenth cranial nerve or vagus nerve, provides the motor innervation for all the laryngeal intrinsic muscles with the exception of the cricothyroid muscle and the sensory innervation of the subglottic laryngeal mucosa. With respect to its origin, different in the right and left sides, the former at the base of the neck and the latter in the left superior

mediastinum, the course it presents from these origins to its target organ is an upward one, hence its name recurrent. Along its course, it emits cardiac, tracheal, pharyngeal branches (for the inferior constrictor); however, its terminal branches directed toward the previously described laryngeal musculature mark its most relevant functional detail (Testut & Latarjet, 1984; Rouvière & Delmas, 2005).

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The first description of the vagus nerve is attributed to Marinus in the year 100 AD (Plaza Rivas, 2008).

Galen (130-200 AD), the most important physician of the Greco-Roman period and considered the founder of medicine of the nervous system, contributed to knowledge of comparative anatomy by dissecting numerous animals. He believed that the brain was the center of the psychic pneuma, i.e., of the rational breath that informed and ordered the universe. According to him, this psychic pneuma was disseminated via the nerves, demonstrating, in addition, that the brain was the center of voluntary movement and sensitivity. In this context, he explained the losses of sensation and paralyzes as occurring with the section of certain nerves or the spinal cord at different levels. He thus distinguished between the sensory and motor nerves and suggested that the motor nerves came from the cerebellum and the sensory nerves entered it. Among the seven cranial nerves he described, the vagus nerve was included, in great detail, with its branches, including its superior laryngeal branches as well as the inferior, which he called “reversivi” when he noted that it returned towards the larynx to cause the vocal folds to open. These contributions are recognized in *Omnia Opera*, published in Venice in 1541, which depicts him performing a brilliant demonstration, cutting the recurrent laryngeal nerve in a pig, emphasizing its anatomical and functional importance, because when conducting the vivisection of this animal he noticed that the animal ceased to make noise when this nerve was pinched. We can state then that it was Galen who first described the vagus nerve as a cranial nerve (Campos & Henriques; Leoutsakos, 2004; Duque Parra *et al.*, 2014; Sarma *et al.*, 2015).

Abubakr Muhammad Ibn Zakaria Razi (Rhazes) (865-925 AD) was a student of neuroanatomy and described for the first time the recurrent nerve as a mixed sensory and motor nerve (Shoja & Tubbs, 2007).

The name of the vagus nerve was given by Domenico de Marchetti (1626-1688), of the University of Padua, Italy, and comes from the Latin “vagari”; due to its erratic and prolonged route, Willis, Soemmerring and Henle referred to it as the “nervus vagus”. The French and Spanish respectively preferred the name of pneumo-gastrique and pneumogástrico. Juan José Barcia Goyanes stated that this name was inadequate since, on the one hand, pneumo means air and not lung, as it means to convey, and on the other the name refers to only two of the numerous organs that the nerve innervates; therefore, he considered the name vagus appropriate (Plaza Rivas).

Stedman (1823) published the first description of a nonrecurrent laryngeal nerve (NRILN), originating from the

right vagus nerve, going directly into the larynx. This finding was made during a cadaver dissection (Ongaro *et al.*, 2016). This case was related to an right subclavian artery, also anomalous, because it stemmed from the aortic arch, to the left of the left subclavian artery, presenting in addition a retrotracheal and retroesophageal course. This arterial variation was called *arteria lusoria* by Arkin in 1936 (Henry *et al.*, 1988; Coady *et al.*, 2000).

The surgical importance of the nonrecurrent inferior laryngeal nerve began to be considered from the publication by Pemberton & Beaver (1932), after its finding in a cervical surgery. They reported the association with an anomalous origin of the right subclavian artery (Kato *et al.*, 2016).

It was not until 1935 that reports began of nonrecurrent left inferior laryngeal nerves, generally associated with situs inversus (Coady *et al.*).

The presence of the NRILN is directly related to embryonic development. As the pharyngeal arches develop throughout the fourth week, these are irrigated by arteries from the aortic sac, the aortic arches. Six pairs of aortic arches usually develop, but not all at the same time. By the time the sixth pair forms, the first two have disappeared. This sixth left pair will give rise in its proximal portion to the left pulmonary artery distal to the ductus arteriosus and on the right side in its proximal portion to the right pulmonary artery while distally it degenerates. The fifth pair is rudimentary and also degenerates, while the fourth pair on the left gives rise to the aortic arch and on the right side to the right subclavian artery.

The vagus nerve forms at the end of the fifth week, and the branch that will give rise to the ILN becomes evident at the end of the sixth week. This last one is associated with the sixth pharyngeal arch and passes directly to the larynx. As the embryo develops, the neck extends and the larynx moves cranially, whereas the heart descends and aortic arch and the associated vessels remain in the thorax and, with them, the ILN.

This branch, which reaches the larynx directly in the first stage, forms the recurrent loop characteristic of the adult around the distal part of the sixth aortic arch, i.e., the ductus arteriosus on the left and of the right fourth arch (right subclavian artery) by the degeneration of the portion corresponding to the fifth and sixth arches. Therefore, the course of the ILN is determined by the pattern of development of the arteries with it is related, and the variations in this pattern will determine the variations in the anatomical disposition of this nerve (Campos & Henriques; Moore & Persaud, 2004; Ongaro *et al.*).

In summary, if the recurrent laryngeal nerve (RLN) develops without alterations, it will originate from the vagus nerve and, on the right it will surround the right subclavian artery as the left surrounds the aortic arch. Before reaching its point of penetration in the larynx, in most cases the RLN ascends into the neck in the tracheoesophageal groove. During this ascending step, the RLN will cross the inferior thyroid artery (ITA).

The NRILN is attributed to abnormal embryological development of the subclavian artery, generally the absence of the brachiocephalic trunk or base and anomalous course of the right subclavian artery, this being able to pass behind the esophagus (80 %), between the esophagus and the trachea (15 %) or past the trachea (5 %). In these cases, the right posterior communicating artery (PCA) stems directly from the aortic arch. If the fourth aortic arch is absent, the right RLN reaches the larynx directly, resulting in a right NRILN (Cai *et al.*, 2013).

This is not possible on the left, since the sixth arch remains as the ductus Botalli until birth. This is why the NRILN is more frequent on the right side, as a left NRILN is feasible only in the case of dextrocardia or situs inversus viscerum (Lee *et al.*, 2011; Ongaro *et al.*). However, cases of NRILN on both sides have been described with no association with these vascular anomalies (Coady *et al.*; O'Neill *et al.*, 2011).

Identification of the NRILN is important to prevent its intraoperative injury, not only in surgeries on the thyroid and parathyroid glands, but also in other cervical surgeries such as vascular, esophageal, tracheal and neck dissections to mention the most common (Coady *et al.*; Cai *et al.*; Sarma *et al.*; Ongaro *et al.*). It is necessary to consider that its origin and course can present variations, being more frequent on the right side with an approximate prevalence of 0.3 to 1 % (Miranda *et al.*, 2006; O'Neill *et al.*, 2011; Yetisir *et al.*, 2014).

Dissection of the inferior laryngeal nerve is advisable in thyroid surgeries so as to avoid its injury and the resulting morbidity this causes on the innervation of the larynx and the patient's quality of life (Ngo Nyeki, *et al.*, 2015). Its relation to the inferior thyroid artery is useful as a landmark, even with its anatomical variations (Campos & Henriques).

Studies on preoperative images of thyroid surgery, for example computed tomographies, conducted due to the presence of an endotracheal projection of a goiter, or for some other cause, can sometimes reveal vascular anomalies, which due to its high prevalence of association can mean the existence of a NRILN (O'Neill, *et al.*).

The existence of the NRILN is a cause of its iatrogenic injury during cervical surgeries, and therefore it must be taken into consideration (Henry *et al.*, 2017).

The prevalence of NRILN is presented in a series of 100 consecutive total thyroidectomies performed at the National Cancer Institute (INCAN in Spanish) and the Social Security Institute (IPS in Spanish) of Paraguay between June 2016 and May 2017. Postoperative images were used to search for an anomalous subclavian artery. A color echo-Doppler raised the suspicion of its existence, which was fully confirmed with a contrast CT angiography.

The general goal of this work is to determine the prevalence of the NRILN in patients who underwent a total thyroidectomy in two reference centers for head and neck surgery in Paraguay.

The specific objectives of the present study are the following:

- To determine the prevalence of nonrecurrent inferior laryngeal nerve;
- To evaluate the associated symptomatology that can lead to suspicion of its presence;
- To study possible associated vascular anomalies in case of a positive surgical finding.

MATERIAL AND METHOD

Prospective cross-sectional study in a series of 100 consecutive total thyroidectomies in the INCAN and the ORL Service in the Hospital Central of the IPS.

Inclusion and exclusion criteria. Inclusion criteria were patients who underwent a total thyroidectomy due to a multinodular goiter (MNG) or thyroid cancer, with or without a neck dissection between the months of July 2016 and June 2017.

The exclusion criteria were the following: Patients who had undergone totalization of a previous partial thyroidectomy; Patients who had undergone a thyroid lobectomy plus isthmusectomy; Patients who had had previous cervical surgeries; Patients with a history of severe cervical trauma.

RESULTS

100 patients underwent a total thyroidectomy, of whom 90 (90 %) were women. The average age was of 47 years, ranging from 19 to 77 years. Six patients (6 %) also underwent a neck dissection for thyroid cancer.

The preoperative diagnosis was MNG in 84 cases and thyroid cancer in the remaining 16 (16 %).

In every case, a meticulous bilateral search was made for the inferior laryngeal nerves in their cervical course, noting relation with the inferior thyroid artery and Zuckerkandl's

tubercle. This search was systematized for the left side in the tracheoesophageal groove and for the right at the level of the triangle formed by the trachea, common carotid artery and inferior thyroid artery, also known as Simon's triangle (Simon, 1943).

In one man aged 47 years operated on for MNG and presenting slight to solid dysphagia, there was difficulty finding the right NRILN. When it could not be found in Simon's triangle and with the dissection of the inferior thyroid artery, a search was made for the nerve at its entrance to the larynx below the inferior constrictor of the pharynx. This maneuver was effective and from this point its dissection demonstrated a downward course behind the carotid artery, which could be dissected up to its origin in the vagus nerve (Fig. 1). The right lobe presented a grade 2 Zuckerkandl's tubercle below the nerve, so its visualization was not difficult. The middle thyroid vein, also present on the right side, at no point was a cause for confusion with the nerve. The contralateral nerve was found on its usual course following tracheoesophageal groove. This lobe presented a grade 1 Zuckerkandl's tubercle.

There were no postoperative complications, except for a temporary dysphonia, which cleared up after 3 weeks.

With this finding, a color Echo-Doppler of the neck vessels was requested, which revealed the absence of the brachiocephalic trunk and the base of both carotids very close to one another, as well as the base of the right subclavian artery directly from the aortic arch to the left of the left subclavian artery (Fig. 3A). A contrast CT showed that this anomalous vessel presented a retroesophageal course at its base (Figs. 3B-E).

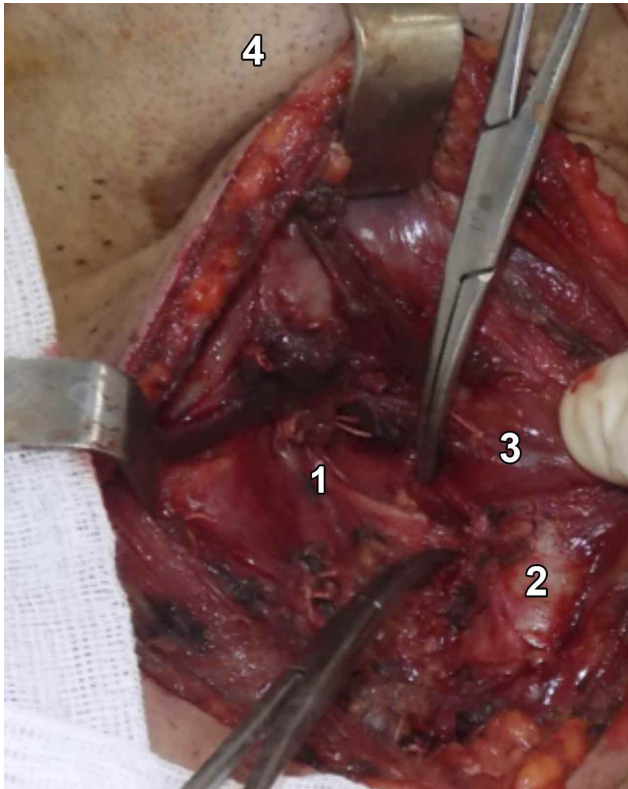


Fig. 1. Surgical dissection of the right side neck. 1. Right nonrecurrent inferior laryngeal nerve (NRILN); 2. Trachea; 3. Inferior pharyngeal constrictor; 4. Chin.

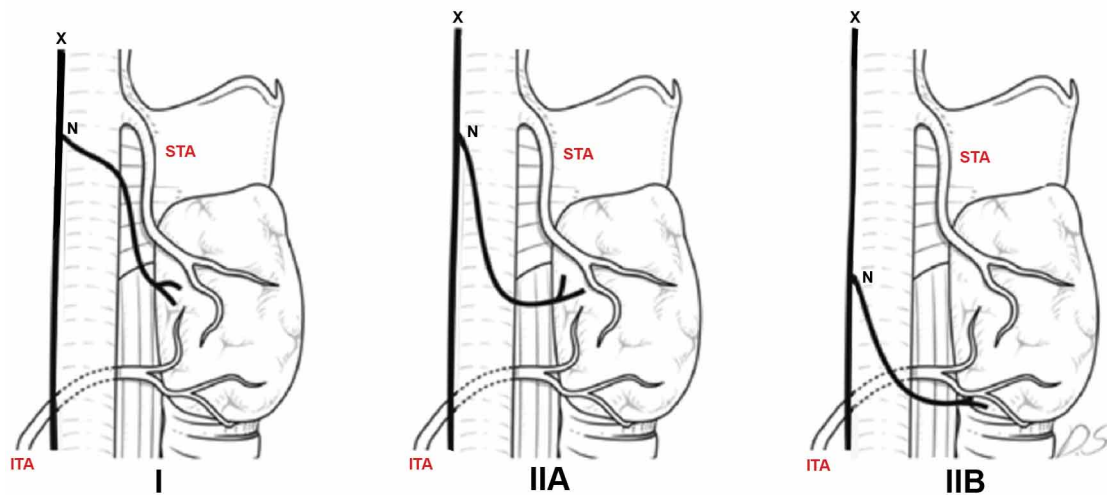


Fig. 2. Avisse Classification. X. Vagus nerve; N. Nonrecurrent inferior laryngeal nerve; ITA. Inferior thyroid artery; STA. Superior thyroid artery.

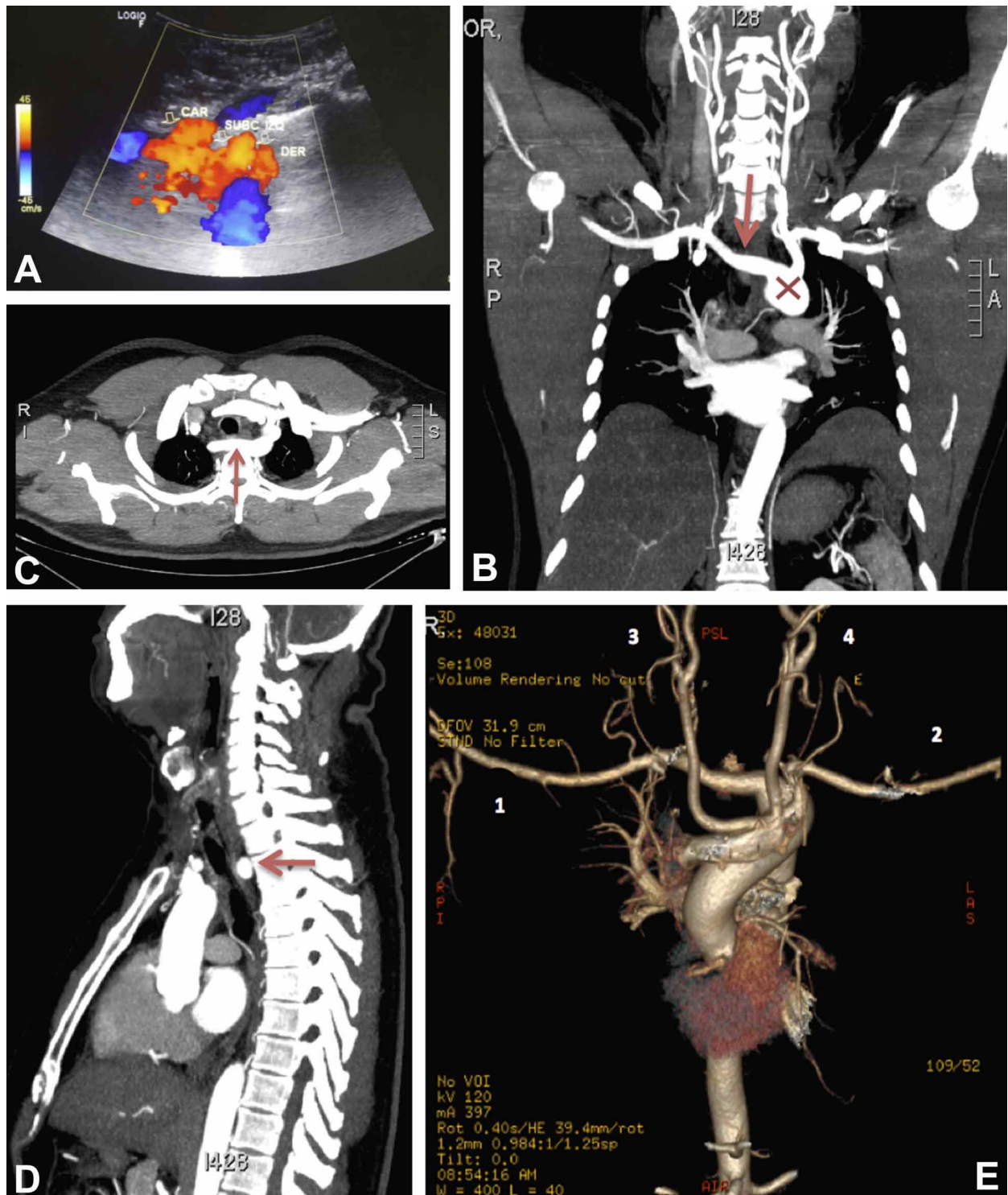


Fig. 3. A. Echo-Doppler color. It showed the anomalous birth of the right carotid artery at the level of the aortic arch to the left of the left subclavian artery. B. CT (coronal section). The right subclavian artery (arrow) is born in the aortic arch (X) behind and to the left of the left subclavian artery. Cross the mediastinum transversely and go to the right base of the neck. C. CT (axial section). The right subclavian (arrow) is born from the aortic arch and passes behind the esophagus, compressing it. D. CT (sagittal section). The subclavian artery in cross section (arrow) behind the esophagus. E. Angio-CT. The right subclavian is born to the left and behind the left subclavian, has a later trajectory. Both common carotids are born very close to the aortic arch (1. right subclavian artery, 2. left subclavian artery, 3. right primitive carotid artery, 4. left carotid artery)

DISCUSSION

The ILN stems from the vagus nerve, which descends vertically in the cervical neurovascular bundle, between and posterior to the primitive carotid artery (PCA) and the internal jugular vein (IJV). The vagus can also display some anatomical variations, and these may or may not coincide with the origin and anomalous course of the ILN. Among the related variations, we draw attention to the location that it presents within the neurovascular bundle in the neck, occupying an anterior or medial position instead of the usual central or posterior one (Gurleyik, 2015; Kato *et al.*).

The ILN is a predominantly somatic motor nerve that originates in the inferior portion of the ambiguous nucleus of the spinal nerve, reaches the vagus nerve or tenth cranial nerve and binds to it in its intracranial portion before exiting via the jugular foramen. The information from the ILN continues its course attached to the vagus nerve and separates only when it has descended to the lower portion of the neck on the right side and to the upper portion of the thorax on the left side. Once separated from the vagus nerve, the right ILN forms a loop at right subclavian artery level, whereas the left one separates from the vagus nerve in the mediastinum, with the first lateral ascending to the trachea and the second via the tracheoesophageal groove. On their course, they are related to the inferior thyroid artery (ITA), an important landmark for the identification of the nerve. The final part of its route within the larynx, along with the internal branch of the superior laryngeal nerve, forms the laryngeal loop (Galen's loop). Its functional integrity permits phonation, keeps the airway permeable during swallowing, breathing and the sensory innervation of the laryngeal mucosa found below the vocal folds (Testut & Latarjet).

The NRILN is a rare anatomical variation and is the result of a vascular anomaly during early embryonic development of the aortic arches and is identified generally by the absence of the brachiocephalic artery and the formation of an aberrant right subclavian artery (arteria lusoria). It can be associated with other anomalies; Peña *et al.* (2013) describe a case with NRILN, arteria lusoria and thoracic duct on the right side. Morais *et al.* (2015) describe a common trunk for both carotids. It is almost always observed on the right side, since on the left side it is usually accompanied by severe vascular anomalies that cause fetal death in most cases (Cai *et al.*).

Recently, the dogma of the association between the NRILN and an aberrant right subclavian artery has been questioned. The NRILN has been described as an entity not

associated with a vascular anomaly despite there being no suitable anatomical and embryological explanation to date. Some other authors consider it a "false NRILN", being in fact an anastomotic trunk between the cervical sympathetic nerves and a the NRILN (Raffaelli *et al.*, 2000; Calzolari *et al.*, 2008; Tateda *et al.*, 2008). The case presented is a well-developed trunk, with no branching and aimed directly towards the lower edge of the inferior constrictor.

The prevalence of the right NRILN is 0.3 % to 1.8 % of the general population, whereas on the left side it is 0.04 % (Lee *et al.*; Ongaro *et al.*), being even less than 0.004 % according to other authors (Mahmodlou *et al.*, 2013). However, although it is considered that this situation only appears in cases of situs inversus (Guerreiro *et al.*, 2014), in some cases this does not occur (Coady *et al.*; Ongaro *et al.*). In this work the right NRILN appears with a prevalence of 1 %, and no case is described on the left side.

Dissection of the inferior laryngeal nerve is advisable in thyroid surgeries so as to avoid its injury and the resulting morbidity this causes on the innervation of the larynx and the patient's quality of life (Ngo Nyeki, *et al.*). Its relation to the inferior thyroid artery is useful as a landmark, even with its anatomical variations (Campos & Henriques). Anitha *et al.* (2014), in a work on cadavers, found that in 59 % the ILN passes behind the ITA, in 30 % of the cases in front of and in 11 % between the branches of the artery. In addition, they described the relation of the nerve with Berry's ligament, in which 64 % passed superficially, 11 % deeply and 25 % through the ligament. The approximate length of the left ILN is 12 centimeters, whereas that of the right nerve is 5 to 6 centimeters (Duque & Londoño, 2009).

There are several anomalies the ILN can present and in addition to the NRILN we can mention extralaryngeal bifurcation in the anterior (motor) and posterior (sensory) branches before entering the organ, and coexistence between the ILN and NRILN on the same side (Sanders *et al.*, 1983; Yalçin, 2006; Obaid *et al.*, 2014; Cetin *et al.*, 2016; Kato *et al.*). Gurleyik (2013) reports that the ILN presents fine branching to adjacent structures, observable only with a microscope and of little surgical value. They describe the prevalence of the extralaryngeal bifurcation of the nerve as between 18 and 43 % and trifurcation 2 %.

In all the patients who underwent a thyroidectomy, we systematically performed a bilateral search for the ILN in its usual course on the left side at tracheoesophageal groove level and on the right side in the laterotracheal space in relation to the triangle formed by the trachea, common carotid artery and inferior thyroid artery, also known as Simon's triangle (Rojas *et al.*).

Noting the lack of systematization in the description of the course and relations of the ILN, mainly on the right side, by anatomists of the time, Simon (1943) presented the aforementioned triangle which bears his name in an effort to prevent injuries to it, stating that, “only those who have seen sufficiently severe injuries of this nerve as to require a tracheostomy can appreciate its importance.” In this publication he also gives an interesting account of Lahey, after 3000 surgeries, saying that the nerve was of sufficient diameter to be observed during surgery and suggested that palpation of the nerve against the trachea could be useful in its identification.

Sarma *et al.* refer to the ILN as being located anterior to the tracheoesophageal groove (41.6 %), posterior to the inferior thyroid artery (35.8 %), lateral to Berry’s ligament (88.1 %), below the inferior edge of the inferior constrictor muscle (90.4 %), entering the larynx prior to its terminal division (54.6 %). Other authors also present their contributions on the matter.

Given the inability to find the nerve in its usual location and after assessing the anatomical landmarks used to this effect, the retrograde dissection is useful, i.e., from the entrance of the nerve to the larynx below the fascicles of the cricopharyngeal portion of the inferior constrictor, always taking the possibility of its extralaryngeal division into consideration (Ngo Nyeki *et al.*). An alternative is the delicate digital palpation of the nerve (Hardy’s maneuver) in the tracheoesophageal junction (Duque & Londoño). In that case, when the ILN was not found in its usual position and when it was not perceived by palpation, this was rightly looked for at the point of its entrance into the larynx, being found and dissected from this point.

An important anatomical landmark is the presence of Zuckerkandl’s tubercle, which sometimes covers the upper third of the ILN. This subsequent prolongation of the lobe, which has its embryological origin in the merging of the central primordium originating from the floor of the pharynx and the lateral primordia of the fifth pharyngeal pouch, can make dissecting the nerve difficult (Gil-Carcedo Sañudo *et al.*, 2012). On rare occasions it can be associated with a NRILN (Gurleyik, *et al.*, 2012). Gurleyik *et al.* (2017) report the case of a NRILN and extralaryngeal bifurcation of the ILN in close relation with Zuckerkandl’s tubercle, all on the right side. In the case that we present in this work, the tubercle was present but below the nerve, and therefore its dissection was not difficult. Indeed, the absence of the nerve below the tubercle was the step prior to its search below the inferior constrictor of the pharynx.

Its presentation is usually accompanied by vascular anatomical variations, particularly the right subclavian artery, which in these situations usually stems directly from the aortic arch and presents a retroesophageal course, with this vessel receiving the name *arteria lusoria*, being accompanied by the absence of the brachiocephalic arterial trunk. Often these anomalies are demonstrated by postoperative tomographies (CTs) (Mahmodlou *et al.*).

According to a meta-analysis performed by Henry *et al.* (2017), the association between the right NRILN and the presence of anomalies of the subclavian artery is reported in 86.7 % of the cases.

CTs are not part of the usual preoperative studies in thyroid surgeries; however, the preoperative diagnosis of these vascular anatomical variations causes one to assume the coexistence of a NRILN and thus to steer the search directly to the level of the entrance to the larynx (Wang *et al.*, 2014). Kato *et al.* describe three cases of *arteria lusoria* diagnosed preoperatively by CT and that coexisted with the NRILN as an intraoperative finding, two of which emerged at cricoid cartilage level and another at the inferior pole of the thyroid gland. This last case presented a vertebral artery anomaly that stemmed from the right subclavian, medial to the base of the PCA and with a course also medial to the base. In the three cases, the vagus nerve occupied a medial position in the carotid sheath. The search for the nerve in these cases can be done with intraoperative pneumomonitoring (Pardal-Refoyo, 2012; Cai *et al.*; Gao *et al.*, 2014; Juárez-Ferrer *et al.*, 2016).

Intraoperative neuromonitoring makes it possible to evaluate not only the ILN and its branches and its variations, including the NRILN, but also to differentiate it from anastomotic sympathetic branches (Chiang *et al.*, 2010; Donatini *et al.*, 2013).

In this patient an echo-Doppler was done on the neck vessels, revealing the absence of the brachiocephalic arterial trunk, and even being able to individualize the anomalous base of the right subclavian artery in the aortic arch to the left of the left subclavian. To confirm this finding, a contrast CT was done that did indeed confirm it, showing in addition the retroesophageal course of the anomalous artery.

The patient was referred for slight to solid dysphagia, which were related to compression caused by a grade 2 goiter. This symptom (*dysphagia lusoria*) is only present in 5 to 10 % of those with *arteria lusoria*. Other accompanying symptoms can be irritative cough and ischemia of the upper right limb (Guerreiro *et al.*; Henry *et al.*, 2017).

The plain chest x-ray is part of the preoperative studies of patients undergoing thyroid surgery. The finding of situs inversus warns of the possibility of finding a nonrecurrent left inferior laryngeal nerve. However, 20 % of the cases show an oblique linear shadow which can be observed in the anteroposterior incidence as well as a rounded object going up and to the right of the superior part of the aortic arch, which may correspond to an aberrant right subclavian artery. A barium swallow can also reveal the bayonet sign (i.e., an extrinsic compression on the left edge and posterior face of the esophagus at third dorsal vertebral level below the aortic arch), but this sign can be lost easily if the upper thoracic esophagus is not carefully examined and if lateral or oblique views are not obtained. An ultrasound scan can show the presence of the brachiocephalic trunk and the base of its two branches; performed by highly trained personnel, it offers important data for the suspicion of a NRILN (Morais *et al.*; Gurleyik & Gurleyik, 2016; Korschake *et al.*, 2016). Iacobone *et al.* (2015) report a 98 % effectiveness with the method for this purpose.

Other studies that can contribute but which are not used routinely in thyroid pathology are endoscopy, which can show extrinsic extrinsic pulsating compression on the posterior wall of the esophagus, although this sign is only found in 40 % of cases, as well as transendoscopic ultrasound and arteriography (Lee *et al.*; Ongaro *et al.*).

Avisse *et al.* (1998) defined two types of NRILN. Type 1 stems directly from the vagus and runs together with the superior thyroid vessels. Type 2A has a cross-sectional course over the trunk of the ITA, with this variety being the most frequently described. Type 2B runs with a parallel cross-sectional course, under or between the branches of the ITA (Toniato *et al.*, 2004; Guerreiro *et al.*; Lee *et al.*). The case related in this casuistry corresponds to Type 1 (Fig. 2).

Injury to the Type 1 NRILN generally occurs during the dissection and section of the superior thyroid pedicle and in Type 2 with handling of the ITA (Lee, *et al.*). We consider that the technique used by us of not binding the superior pedicle before correctly individualizing the ILN contributed to avoiding injury to the nerve.

The prevalence of permanent injury to the ILN in thyroid surgery is 1.8 %, whereas in cases of NRILN this percentage increases to 12 % (Toniato *et al.*; Gao *et al.*). Nevertheless, as previously mentioned, the use of intraoperative neuromonitoring reduces this likelihood to a minimum (Gurleyik & Gurleyik; Korschake *et al.*)

The NRILN may be confused with a “false NRILN”, formed by anastomotic branches of the sympathetic nerve, these generally being plexiform and fine. However, sometimes they have a considerable diameter. These branches can stem from the middle cervical sympathetic ganglion with more frequency, although also from the superior or inferior ganglia, and even directly from the sympathetic chain. Although this situation is rare (1.5 %), it is more frequent than the NRILN. These branches influence the secretion of the laryngeal mucous glands, the vascularization of the vocal cords, but their direct action on the motility of the vocal cords has not been shown. Sometimes Galen’s loop originates outside the larynx, with its course being more internal and vertical than the NRILN. Other cross-sectional structures that may cause confusion are the middle thyroid veins (Cai *et al.*; Yetisir *et al.*, 2013; Ongaro *et al.*). The case related presented a middle thyroid vein but at no point was it confused with a neural structure. Forde & Williams (2015) recommend not binding any cross-sectional structure between the carotid sheath and the larynx, except for the middle thyroid veins, until the ILN has been individualized. We routinely look for this vessel and we bind and section it to be able to dislocate the thyroid lobe and look for the ILN in its usual course.

CONCLUSIONS

The NRILN is a rare entity. In this case study, it represents 1 % of the patients operated on for thyroid pathology in the INCAN and IPS in a 1-year period.

The slight dysphagia presented by the patient with a grade 2 goiter is noteworthy, and may warn of the presence of a cancer or some extrinsic compression of the cervical esophagus of another etiology. This symptom could be related to the presence of an aberrant right subclavian artery which was not evaluated. This leads us to the conclusion that for a patient with a thyroid pathology and dysphagia, it is necessary to maximize imaging diagnostics that can raise suspicion of vascular anomalies associated with a nonrecurrent inferior laryngeal nerve. This also enables all the precautions to be taken to reduce the risks of nerve injury.

In the event of an intraoperative finding of this neural anomaly, we consider that imaging can offer a valuable contribution postoperatively in the search for the associated vascular variations to inform the patient of possible future clinical-surgical situations.

MEDINA RUÍZ, B. A.; OSORIO FLEITAS, M.; PERSANO, M. B.; DAMI, H. R.; VEGA, R. B. & OTTONE, N. E. Prevalencia del nervio laríngeo inferior derecho no recurrente en una serie de 100 tiroidectomías totales. *Int. J. Morphol.*, 36(1):149-158, 2018.

RESUMEN: El nervio laríngeo inferior no recurrente (NLINR) del lado derecho es una entidad rara asociada generalmente a una arteria lusoria. Su prevalencia oscila entre el 0,3 al 1,8 %. Su hallazgo se da sobre todo en cirugías tiroideas, al ser éstas las cirugías cervicales más frecuentemente realizadas. El diagnóstico preoperatorio de esta anomalía nerviosa es realizado en contadas ocasiones. La disfagia puede ser un síntoma de alerta ante esta situación, siendo a veces erróneamente relacionada con la compresión esofágica por un bocio o cáncer tiroideo. El diagnóstico postoperatorio de arteria lusoria acompañante debería realizarse para certificar el cuadro e informar al paciente ante eventuales procedimientos médico-quirúrgicos futuros. El objetivo de este trabajo consistió en determinar la prevalencia del nervio laríngeo inferior no recurrente en los pacientes operados de tiroidectomía total en dos centros de referencia en cirugía de cabeza y cuello del Paraguay. Estudio prospectivo de corte transversal en una serie de 100 tiroidectomías totales consecutivas, operados en el INCAN y el Servicio de ORL del Hospital Central del IPS. Fueron sometidos a tiroidectomía total 100 pacientes, de los cuales 90 fueron del sexo femenino. La edad promedio fue de 47 años. En 6% se realizó además vaciamiento cervical por cáncer de tiroides. El diagnóstico preoperatorio fue bocio multinodular (BMN) en 84 casos y cáncer de tiroides en los restantes 16 (16%). En un varón de 47 años, (1%) operado por bocio multinodular, y que refería disfagia leve a sólidos, se tuvo dificultad para encontrar al nervio laríngeo inferior derecho. Siendo el mismo localizado a nivel del margen inferior del constrictor inferior de la faringe y comprobándose su trayecto anómalo descendente. En el postoperatorio, una vez confirmado el diagnóstico de benignidad de la patología tiroidea, se realizó un ecodoppler color y una angiotomografía que corroboraron el diagnóstico de arteria lusoria asociada. Ante la dificultad en localizar al nervio laríngeo inferior recurrente en una cirugía tiroidea, es conveniente considerar la posibilidad de que el mismo sea no recurrente e ir a buscarlo a nivel de su ingreso a la laringe por debajo del constrictor inferior. En estos casos conviene estudiar al paciente en el postoperatorio para descartar una anomalía vascular asociada. El estudio tomográfico en todo paciente portador de patología tiroidea y disfagia podría contribuir al diagnóstico de la anomalía vascular y extremar los recaudos en la disección del nervio, incluso utilizando el neuroestimulador. El NLINR es una entidad rara. En esta casuística representa el 1 % de los pacientes operados por patología tiroidea en el INCAN e IPS en el periodo de 1 año (julio 2016/ junio 2017).

PALABRAS CLAVE: nervio laríngeo inferior no recurrente derecho; Tiroidectomía; Cuello.

REFERENCES

Anitha, T.; Dombé, D. & Dharmendra, P. Clinically relevant variations of recurrent laryngeal nerve. *J. Dent. Med. Sci.*, 13(9):59-62, 2014.

- Avisse, C.; Marcus, C.; Delattre, J. F.; Marcus, C.; Cailliez-Tomasi, J. P.; Palot, J. P.; Ladam-Marcus, V.; Menanteau, B. & Flament, J. B. Right nonrecurrent inferior laryngeal nerve and arteria lusoria: the diagnostic and therapeutic implications of an anatomic anomaly. Review of 17 cases. *Surg. Radiol. Anat.*, 20(3):227-32, 1998.
- Cai, Q.; Guan, Z.; Huang, X.; Yuan, J.; Pan, Y.; Zheng, Y.; Liang, M. & Fan, S. The usefulness of preoperative computed tomography and intraoperative neuromonitoring identification of the nonrecurrent inferior laryngeal nerve. *Eur. Arch. Otorhinolaryngol.*, 270(7):2135-40, 2013.
- Calzolari, F.; Misso, C.; Monacelli, M.; Lucchini, R.; Sanguinetti, A.; D'Ajello, M.; Vannucci, J.; Galasso, V.; Bartolo, M.; Ragusa, M. & Avenia, N. Non-recurrent inferior laryngeal nerves and sympathetic-inferior laryngeal anastomotic branches: 6 years' personal experience. *Chir. Ital.*, 60(2):221-5, 2008.
- Campos, B. A. & Henriques, P. R. Relationship between the recurrent laryngeal nerve and the inferior thyroid artery: a study in corpses. *Rev. Hosp. Clin. Fac. Med. São Paulo*, 55(6):195-200, 2000.
- Cetin, F.; Gürleyik, E. & Dogan, S. Morphology and functional anatomy of the recurrent laryngeal nerve with extralaryngeal terminal bifurcation. *Ant. Res. Int.*, 2016:9503170, 2016.
- Chiang, F. Y.; Lu, I. C.; Chen, H. C.; Chen, H. Y.; Tsai, C. J.; Hsiao, P. J.; Lee, K. W. & Wu, C. W. Anatomical variations of recurrent laryngeal nerve during thyroid surgery: how to identify and handle the variations with intraoperative neuromonitoring. *Kaohsiung J. Med. Sci.*, 26(11):575-83, 2010.
- Coady, M. A.; Adler, F.; Davila, J. J. & Gahtan, V. Nonrecurrent laryngeal nerve during carotid artery surgery: case report and literature review. *J. Vasc. Surg.*, 32(1):192-6, 2000.
- Donatini, G.; Carnaille, B. & Dionigi, G. Increased detection of non-recurrent inferior laryngeal nerve (NRLN) during thyroid surgery using systematic intraoperative neuromonitoring (IONM). *World J Surg.*, 37(1):91-3, 2013.
- Duque Parra, J. E.; Barco Ríos, J. & Duque Quintero, V. Historic view of the structure and function of the nerve. Pregalenic and Galenic view. *Int. J. Morphol.*, 32(3):987-90, 2014.
- Duque, F. C. S. & Londoño, B. A. F. Nervio laríngeo no recurrente en cirugía de tiroides: reporte de dos casos. *Acta Otorrinolaringol. Cir. Cabeza Cuello*, 37(2):107-11, 2009.
- Forde, R. & Williams, E. W. The non-recurrent laryngeal nerve -- A rare phenomenon which requires vigilance. *West Indian Med. J.*, 64(3):303-4, 2015.
- Gao, E. L.; Zou, X.; Zhou, Y. H.; Xie, D. H.; Lan, J. & Guan, H. G. Increased prediction of right nonrecurrent laryngeal nerve in thyroid surgery using preoperative computed tomography with intraoperative neuromonitoring identification. *World J. Surg. Oncol.*, 12:262, 2014.
- Gil-Carcedo Sañudo, E.; Menéndez Argüelles, M. E.; Vallejo Valdezate, L. A.; Herrero Calvo, D. & Gil-Carcedo García, L. M. Zuckerkandl's tubercle. Location, shape and dimensions. *Acta Otorrinolaringol. Esp.*, 63(6):443-9, 2012.
- Guerreiro, S.; Lamas, M.; Candeias, H.; Eusébio, R. & Rocha, V. The non-recurrent laryngeal nerve: An anatomical "trap". *Rev. Port. Endocrinol. Diabetes Metab.*, 9(1):84-7, 2014.
- Gurleyik, E. & Gurleyik, G. Nonrecurrent laryngeal nerve in the era of intraoperative nerve monitoring. *Adv. Med.*, 2016:1606029, 2016.
- Gurleyik, E. Extralaryngeal terminal division of the inferior laryngeal nerve: anatomical classification by a surgical point of view. *J. Thyroid Res.*, 2013:731250, 2013.
- Gurleyik, E. Non-recurrent nerve from the vagus antero-medially located in the carotid sheath. *Ulus Cerrahi Derg.*, 31(3):182-4, 2015.
- Gurleyik, E.; Dogan, S. & Cetin, F. Coexistence of right nonrecurrent nerve and bifurcated recurrent laryngeal nerve pointed by Zuckerkandl's tubercle. *Cureus*, 9(3):e1078, 2017.
- Gurleyik, E.; Dogan, S.; Gunal, O. & Pehlivan, M. The rare coincidence: Nonrecurrent laryngeal nerve pointed by a Zuckerkandl's tubercle. *Case Rep. Med.*, 2012:143049, 2012.

- Henry, B. M.; Sanna, S.; Graves, M. J.; Vikse, J.; Sanna, B.; Tomaszewska, I. M.; Tubbs, R. S.; Walocha, J. A. & Tomaszewski, K. A. The non-recurrent laryngeal nerve: A meta-analysis and clinical considerations. *Peer J.*, 5:e3012, 2017.
- Henry, J. F.; Audiffret, J.; Denizot, A. & Plan, M. The nonrecurrent inferior laryngeal nerve: review of 33 cases, including two on the left side. *Surgery*, 104(6):977-83, 1988.
- Iacobone, M.; Citton, M.; Pagura, G.; Viel, G. & Nitti, D. Increased and safer detection of nonrecurrent inferior laryngeal nerve after preoperative ultrasonography. *Laryngoscope*, 125(7):1743-7, 2015.
- Juárez-Ferrer, J. C.; López-Chavira, A.; Rivera-Martínez, C. G.; Pacheco-Ramírez, M. A. & Mateos-Aguilar, O. O. Utilidad del neuromonitoreo intraoperatorio del nervio laríngeo recurrente durante la cirugía tiroidea. *An. Otorrinolaringol.*, 61(4):263-70, 2016.
- Kato, K.; Toriumi, Y.; Kamio, M.; Nogi, H.; Shioya, H. & Takeyama, H. Nonrecurrent inferior laryngeal nerves and anatomical findings during thyroid surgery: report of three cases. *Surg. Case Rep.*, 2(1):44, 2016.
- Konschake, M.; Zwierrzina, M. E.; Pechriggl, E. J.; Moriggl, B.; Brenner, E.; Hörmann, R. & Prommegger, R. The nonrecurrent laryngeal nerve: A clinical anatomic mapping with regard to intraoperative neuromonitoring. *Surgery*, 160(1):161-8, 2016.
- Lee, Y. S.; Son, E. J.; Chang, H. S.; Chung, W. Y.; Nam, K. H. & Park, C. S. Computed tomography is useful for preoperative identification of nonrecurrent laryngeal nerve in thyroid cancer patients. *Otolaryngol. Head Neck Surg.*, 145(2):204-7, 2011.
- Leoutsakos, V. A short history of the thyroid gland. *Hormones (Athens)*, 3(4):268-71, 2004.
- Mahmodlou, R.; Aghasi, M. R. & Sepehrvand, N. Identifying the non-recurrent laryngeal nerve: preventing a major risk of morbidity during thyroidectomy. *Int. J. Prev. Med.*, 4(2):237-40, 2013.
- Miranda, J. A.; Sartini, A. L. & de Carvalho Borges, M. H. Nervio Laríngeo Inferior Não - Recorrente: Relato de Caso. *Arq. Int. Otorrinolaringol.*, 10(4):318-21, 2006.
- Moore, K. L. & Persaud, T. V. N. *Embriología Clínica. El Desarrollo del Ser Humano*. 7th ed. Madrid, Elsevier. Madrid, 2004. pp.361-6.
- Morais, M.; Capela-Costa, J.; Matos-Lima, L. & Costa Maia, J. Nonrecurrent Laryngeal Nerve and Associated Anatomical Variations: The Art of Prediction. *Eur. Thyroid J.*, 4(4):234-8, 2015.
- Ngo Nyekia, A. R.; Njockb, L. R.; Miloundja, J.; Evehe Vokwelyd, J. E. & Bengono, G. Recurrent laryngeal nerve landmarks during thyroidectomy. *Eur. Ann. Otorhinolaryngol. Head Neck Dis.*, 132(5):265-9, 2015.
- O'Neill, J. P.; La Femina, J.; Kraus, D. & Shaha, A. R. The nonrecurrent laryngeal nerve in thyroid surgery. *World J. Endocr. Surg.*, 3(1):1-2, 2011.
- Obaid, T.; Kulkarni, N.; Pezzi, T. A.; Turkeltaub, A. E. & Pezzi, C. M. Coexisting right nonrecurrent and right recurrent inferior laryngeal nerves: a rare and controversial entity: report of a case and review of the literature. *Surg. Today*, 44(12):2392-6, 2014.
- Ongaro, D.; Elia, S.; Cazzaniga, R. & Taglietti, L. Right non-recurrent inferior laryngeal nerve discovered during carotid endarterectomy: A case report and literature review. *Int. J. Cardiovasc. Thorac. Surg.*, 2(4):29-33, 2016.
- Pardal-Refoyo, J. L. Usefulness of neuromonitoring in thyroid surgery. *Acta Otorrinolaringol. Esp.*, 63(5):355-63, 2012.
- Pemberton, J. J. & Beaver, M. G. Anomaly of the right recurrent laryngeal nerve. *Surg. Gynecol. Obstet.*, 54:594-5, 1932.
- Peña, E.; Zúñiga, J. & Baena, G. Simultaneous occurrence of three anatomical variations: anomalous right subclavian artery, non-recurrent inferior laryngeal nerve and right thoracic duct. *Int. J. Morphol.*, 31(4):1181-4, 2013.
- Plaza Rivas, F. Historia de la nomenclatura de los nervios craneales. *Rev. Soc. Venez. Hist. Med.*, 57(1-2):7-16, 2008.
- Raffaelli, M.; Iacobone, M. & Henry, J. F. The "false" nonrecurrent inferior laryngeal nerve. *Surgery*, 128(6):1082-7, 2000.
- Rojas, M.; Quijano, Y. & Luque Bernal, R. M. Variaciones anatómicas del nervio laríngeo recurrente en una muestra de población colombiana. *Rev. Fac. Med. (Bogotá)*, 64(2):207-13, 2016.
- Rouvière, H. & Delmas, A. *Anatomía Humana: Descriptiva, Topográfica y Funcional*. Tomo 1: Cabeza y Cuello. 11th ed. Barcelona, Masson, 2005. pp.308-16.
- Sanders, G.; Uyeda, R. Y. & Karlan, M. S. Nonrecurrent inferior laryngeal nerves and their association with a recurrent branch. *Am. J. Surg.*, 146(4):501-3, 1983.
- Sarma, M. K.; Kakati, K.; Sharma, K. & Goswami, S. Ch. Recurrent laryngeal nerve injury (RLNI) in thyroid surgery and its prevention. *Int. J. Res. Med. Sci.*, 3(7):1632-6, 2015.
- Shoja, M. M. & Tubbs, R. S. The history of anatomy in Persia. *J. Anat.*, 210(4):359-78, 2007.
- Simon, M. M. Recurrent laryngeal nerve in thyroid surgery - Triangle for its recognition and protection. *Am. J. Surg.*, 60(2):212-20, 1943.
- Stedman, G. W. A singular distribution of the nerves and arteries of the neck and the top of the thorax. *Edin. Med. Surg. J.*, 19:564-5, 1823.
- Tateda, M.; Hasegawa, J.; Sagai, S.; Nakanome, A.; Katagiri, K.; Ishida, E.; Kanno, R.; Hasegawa, T. & Kobayashi, T. Nonrecurrent inferior laryngeal nerve without vascular anomaly as a genuine entity. *Tohoku J. Exp. Med.*, 216(2):133-7, 2008.
- Testut, L. & Latarjet, A. *Tratado de Anatomía Humana*. Tomo III. Barcelona, Salvat Editores, 1984. pp.169-72.
- Toniato, A.; Mazzarotto, R.; Piotto, A.; Bernante, P.; Pagetta, C. & Pelizzo, M. R. Identification of the nonrecurrent laryngeal nerve during thyroid surgery: 20-year experience. *World J. Surg.*, 28(7):659-61, 2004.
- Wang, Z.; Zhang, H.; Zhang, P.; He, L. & Dong, W. Preoperative diagnosis and intraoperative protection of nonrecurrent laryngeal nerve: a review of 5 cases. *Med. Sci. Monit.*, 20:233-7, 2014.
- Yalçin, B. Anatomic configurations of the recurrent laryngeal nerve and inferior thyroid artery. *Surgery*, 139(2):181-7, 2006.
- Yetis,ir, F.; Özkardes, A. B.; Dündar, H. Z.; Birkan, B.; Çiftci, A. B. & Kılıç, M. Non-recurrent laryngeal nerve. *Ulus. Cerrahi Derg.*, 30(2):112-4, 2014.
- Yetis,ir, F.; Salman, E.; Özkardes, A. B.; Aydın, S. M. & Kılıç, M. Fusion of a cervical sympathetic ganglion with the recurrent inferior laryngeal nerve: a case of false positive non-recurrent inferior laryngeal nerve. *Ulus. Cerrahi Derg.*, 29(3):150-2, 2013.

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