



Non recurrent laryngeal nerve with right aberrant subclavian artery in recurrent case of papillary carcinoma of thyroid: an interesting clinical entity.

*Sudhir M Naik Purshottam Chavan Rajshekar Halkud Ashok M Shenoy Sumit gupta
Latif zameer*

Kidwai Memorial Institute of Oncology Bangalore

Abstract:

Background/objectives: A nonrecurrent laryngeal nerve is a rare anatomical variant and a routine preoperative imaging studies are not indicated. 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. Case report: The patient presented with a swelling in the neck for 4 months which is slowly increasing in size. Patient gives history of a surgery done in the neck with an old midline suprasternal scar which was done 40 years earlier. laryngoscopic mirror examination was normal with moving vocal cords. Rest of the oral and oropharyngeal examination along with complete blood picture and biochemical investigations were normal. The patient was not a known diabetic or hypertensive.

Conclusion: Accurate knowledge of frequent variations of the nerve along with the non recurrent variant reduces the risk of intraoperative injury. A nonrecurrent laryngeal nerve(NRLN) is a rare anatomical variant and a routine preoperative imaging studies are not indicated. 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.

Introduction:

Thyroid surgery is a common surgical procedure in the Indian subcontinent.^{1,2} Complications such as bleeding, hypoparathyroidism and recurrent laryngeal nerve(RLN) injury represent nearly half of all the complications of thyroid surgery.³⁻⁵ The nerve injury after thyroidectomy, although infrequent can jeopardize the quality of life.⁶ In addition to the hoarseness that occurs with unilateral RLN injury, bilateral RLN injury leads to dyspnea

and often life-threatening glottal obstruction.^{7,8} The nerve injuries are more during revision surgeries and advanced thyroid malignancies of the thyroid and vascular thyroid of Graves disease.^{9,10} Ensuring the integrity of the RLN is to always identify the nerve during all surgical procedure on thyroid and parathyroid glands.^{11,12} The RLN usually branches off the vagus and loops under the subclavian artery on the right and under the ligamentum arteriosum on the left innervating all the intrinsic laryngeal muscles except the cricothyroid muscle.¹³ The relative long course of the nerve in the neck makes the nerve vulnerable to iatrogenic injury in numerous procedures involving the cervical and upper thoracic regions.¹³ Accurate knowledge of frequent variations of the nerve along with the non recurrent variant reduces the risk of intraoperative injury.¹⁴ A nonrecurrent laryngeal nerve(NRLN) is a rare anatomical variant and a routine preoperative imaging studies are not indicated.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 CT scan of the chest.¹⁴ High resolution ultrasonography of the neck can give an hint towards the variations of the nerve if particularly investigated.¹⁵ We report a case of NRLN in a 72 year old woman during total thyroidectomy and modified radical neck dissection done with diagnosis of papillary carcinoma of thyroid with neck metastasis.

Case Report:

The patient presented with a swelling in the neck for 4 months which is slowly increasing in size. Patient gives history of a surgery done in the neck with an old midline suprasternal scar which was done 40 years earlier.laryngoscopic mirror examination was normal with moving vocal cords. Rest of the oral and oropharyngeal examination along with complete blood picture and biochemical investigations were normal. The patient was not a known diabetic or hypertensive.(fig 1) T3 was 0.492ng/ml, T4 was 6.140 µg/dl and TSH was 5.4 µIU/ml.

On contrast enhanced CT a heterogeneously enhancing mass lesion at level II, III measuring 3.5x3 cm with multiple areas of necrosis seen. Superiorly extend to cricoids and inferiorly to left supraclavicular region. The lesion displacing the left IJV and carotid anteriorly and left EJV laterally. Both the thyroids, larynx, and pharynx were normal. A diagnosis of metastatic lymphadenopathy was done. (fig 2) On fine needle biopsy it was reported as metrastatic papillary carcinoma of the thyroid.

Echocardiography showed sclerotic aortic valve with mild aortic regurgitation, with normal cardiac chambers, with normal left ventricular function and no regional wall motion abnormalities. Total thyroidectomy was done with left modified neck dissection on the left side with the removal of the nodes and and sternomastoid muscle. (fig 3)The left recurrent laryngeal nerve was traced and saved. On the right a non recurrent laryngeal nerve was seen with its direct branch from the vagus at the level of the level of the isthmus entering the cricothyroid muscle 3 cm away from the vagus without a recurrent course. It was graded as right sided non recurrent laryngeal nerve of type Ia.(fig 4) The patient developed respiratory distress four hours after surgery and was intubated orally and extubated the next day. She was symptomless for 40 hours and later developed stridor. She was relieved of the distresss by tracheostomy and an endoluminal bronchoscopy done and no major abnormalities were seen in the tracheal lumen.

Post operative chest xrays and lateral soft tissue of the neck revealed normal neck anatomy but of CT chest an anomaly of aberrant right subclavian artery(arteria lusoria) coursing behind the esophagus was seen.(fig 5,6) The pathology was confirmed as papillary carcinoma of the thyroid and foci was seen in the thyroid tissue. The patient was sent for large dose iodine scan and radioablation was done. Histopathological examination revealed foci of papillary carcinoma of thyroid in the right lobe and rest of the left and right lobe shows hashimotos thyroiditis. Left sided lymph node mass shows a large lymphnode with metastasis of papillary carcinoma.(fig 7) Rest of the 11 lymph nodes show reactive changes.

Discussion :

Steadman in 1823 reported the first case of NRLN and its clinical importance during surgery was reported in early 1920s.¹⁴ In 1935, Berlin reported this anomaly on the left side.¹⁶ 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 recurrent laryngeal nerve 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.¹⁷

Rarely the fourth arch artery on the right disappears and so the right subclavian originates directly from the aortic arch.¹⁴ In such cases the aberrant right subclavian traverses behind the esophagus in 85% cases, between the esophagus and trachea in 15% and in front of the trachea in 5% of the cases.¹⁴ This variation is known as arteria lusoria.^{15,16,18,19}

Three types of NRLN have been described in literature, type IA: the nerve has a straight course at the level of superior thyroid pedicle, type IB: (most common) the nerve runs transversely at the level of isthmus, type II: nerve has a downward course and loops upwards before reaching trachea esophageal groove.²⁰ 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 mimicks the same.²¹

NRLN have a variable incidence ranging from 0-3.9%.^{22,23} Henry et al reported an incidence of 0.54% (17 cases in 3098) on the right side and 0.07% on the left (2 cases in 2846) when compared to the worldwide incidence of 0.32%.²⁴ 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 recurrent laryngeal nerve caudally when the cardia descends and neck elongates during embryonic development.^{25,26} 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 upward to the right behind the esophagus.²⁷ 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.²⁷ On the left side NRLN is seen in cases of dextrocardia.²⁷

Pre-operative diagnosis of a NRLN is difficult using Computerised tomography (CT) scan or magnetic resonance imaging (MRI) but visualising the coexisting arterial anomaly arteria lusoria gives a lead to possible diagnosis.²⁸ 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.²⁹ Incidences of RLN injuries during thyroid surgeries worldwide is around 1-2% and the total thyroidectomies have replaced other conservative procedures for thyroid malignancies.³⁰

The nerve injuries are minimal with expertise and less with capsular dissection for benign diseases compared to lateral dissection for malignant diseases.³¹ The nerve injury manifests as irregular hoarseness and can be better avoided by identifying and carefully tracing the path of the recurrent nerve.³² Factors affecting the injuries are surgeon's expertise, histopathologic diagnosis, previous thyroid surgeries, technique adapted and anatomical variations.³³ The nerve can be injured in various ways like partial transection, traction, rough handling, contusion, crush, burn, clamping, misplaced ligature, and compromised blood supply.^{34,35} The vocal folds do not approximate each other in unilateral injuries and traction injuries to the axons lasts for six months while transection of the nerve due to cutting ligature or cauterization lasts more than 6 months.^{34,35} Bilateral RLN injuries leads to severe airway compromise warranting an emergency

tracheostomy.³⁰ The upper 2 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.^{34,35} 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.³⁰

Hazem et al reported favourable 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."³⁰ They reported 7.6% RLN injury with nerve not dissected and significantly lesser with complete dissection where verifying its anatomic integrity and extra-laryngeal ramifications is possible.³⁰

The RLN injury rates worldwide on complete dissection and identification is 0 - 2.1%, while in revision cases its higher upto 2-12% and marginally higher if the nerve is not dissected at all.³⁰ The use of nerve monitoring devices during surgery detecting vocal cord movement when the recurrent laryngeal nerve is stimulated has not decreased the RLN injury rates as it is already less with meticulous dissection.³⁶ RLN injury are higher during thyroidectomies for thyroid carcinoma, hyperthyroid (toxic) goiter and recurrent goiter cases as nerve identification and hemostasis are difficult here.³⁶ Post operative adhesions, anatomical displacements and neo-vascularization of the gland makes the nerve more vulnerable in revision surgeries.³⁶ Echternach et al reported more intubation related laryngeal injuries than due to nerve injuries of larynx during thyroid surgeries.³⁷ Nerve identification during surgery reduces the risk of its damage and intraparenchymal dissection and subtotal excision has no role in present day thyroid surgeries.^{33,34,35}

Conclusion:

Accurate knowledge of frequent variations of the nerve along with the non recurrent variant reduces the risk of intraoperative injury. A nonrecurrent laryngeal nerve(NRLN) is a rare anatomical variant and a routine preoperative imaging studies are not indicated. 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.



Fig 1 Scar of previous surgery in the neck

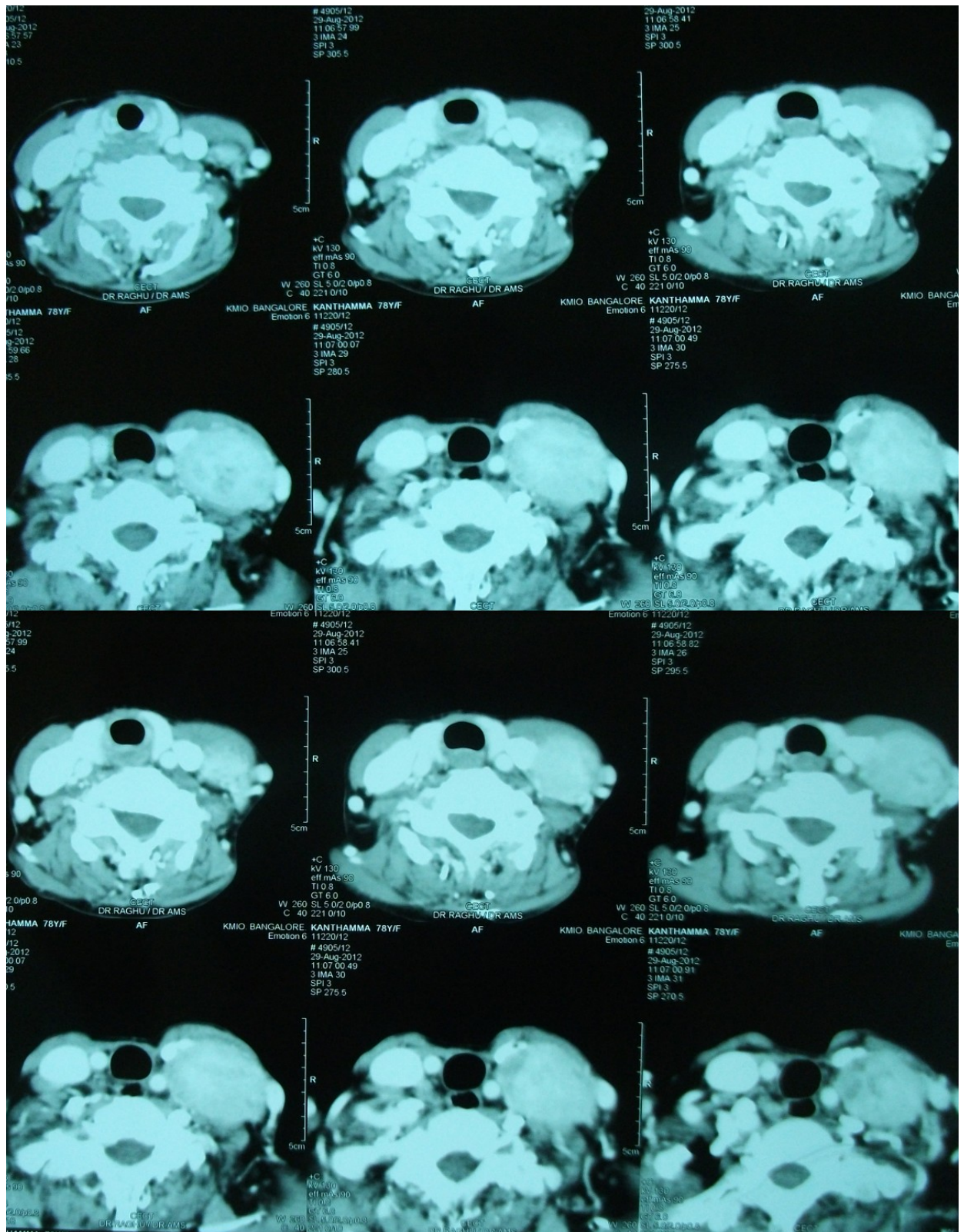


Fig 2 Axial CT scan showing (L) level IV lymph node mass with a small thyroid

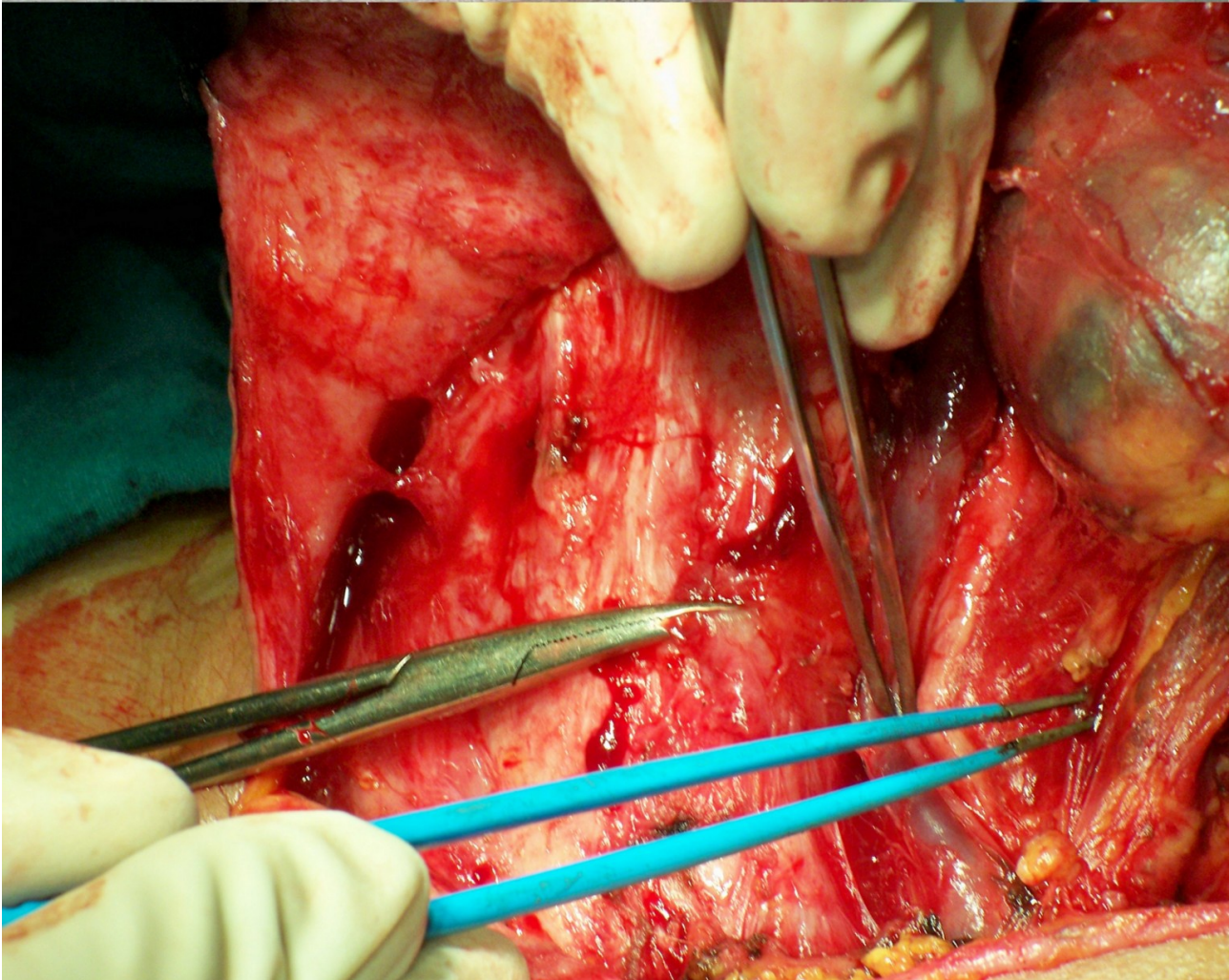
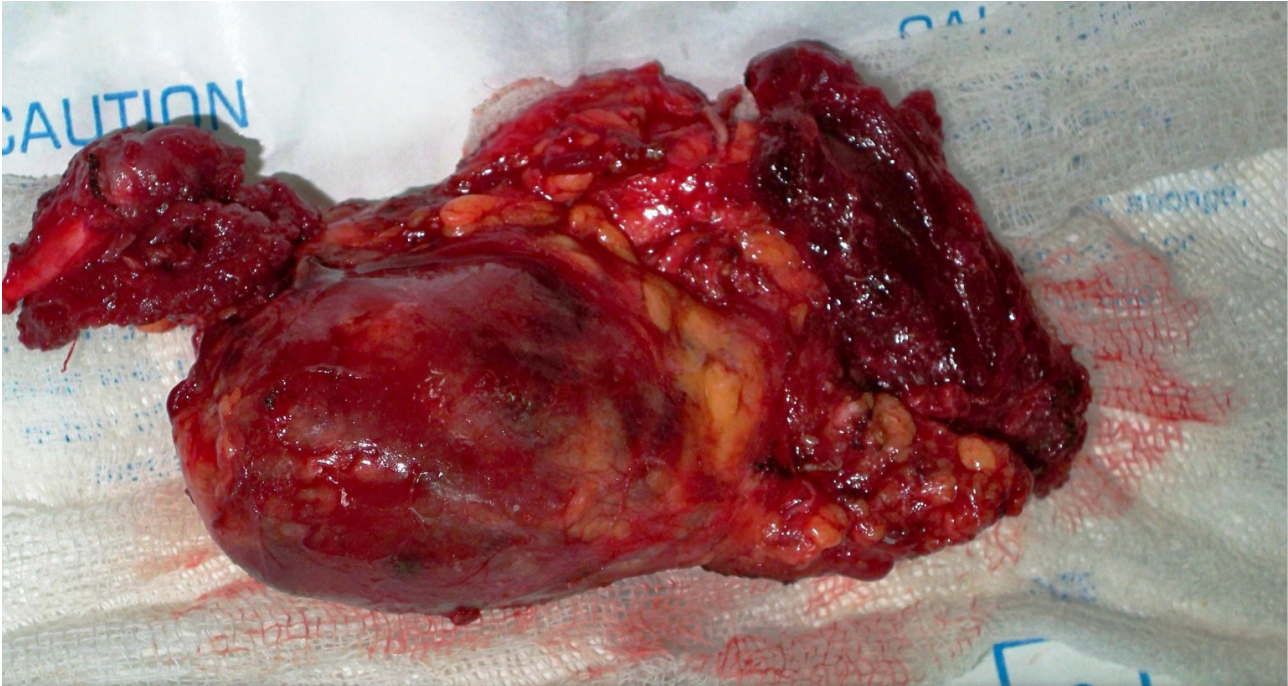


Fig 3 Left modified radical neck dissection with specimen

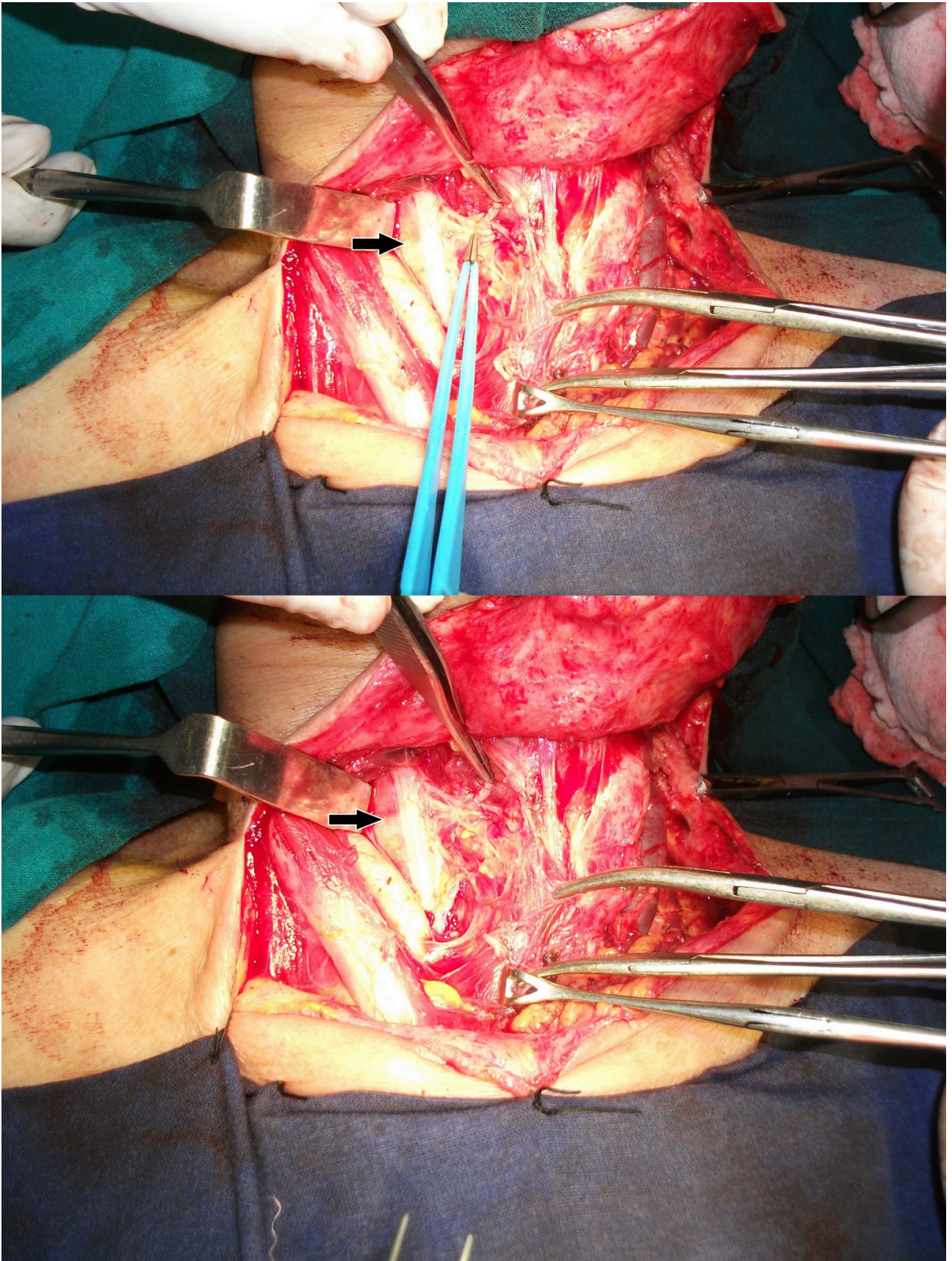


Fig 4 Type Ia NRLN branching directly from the (R) vagus

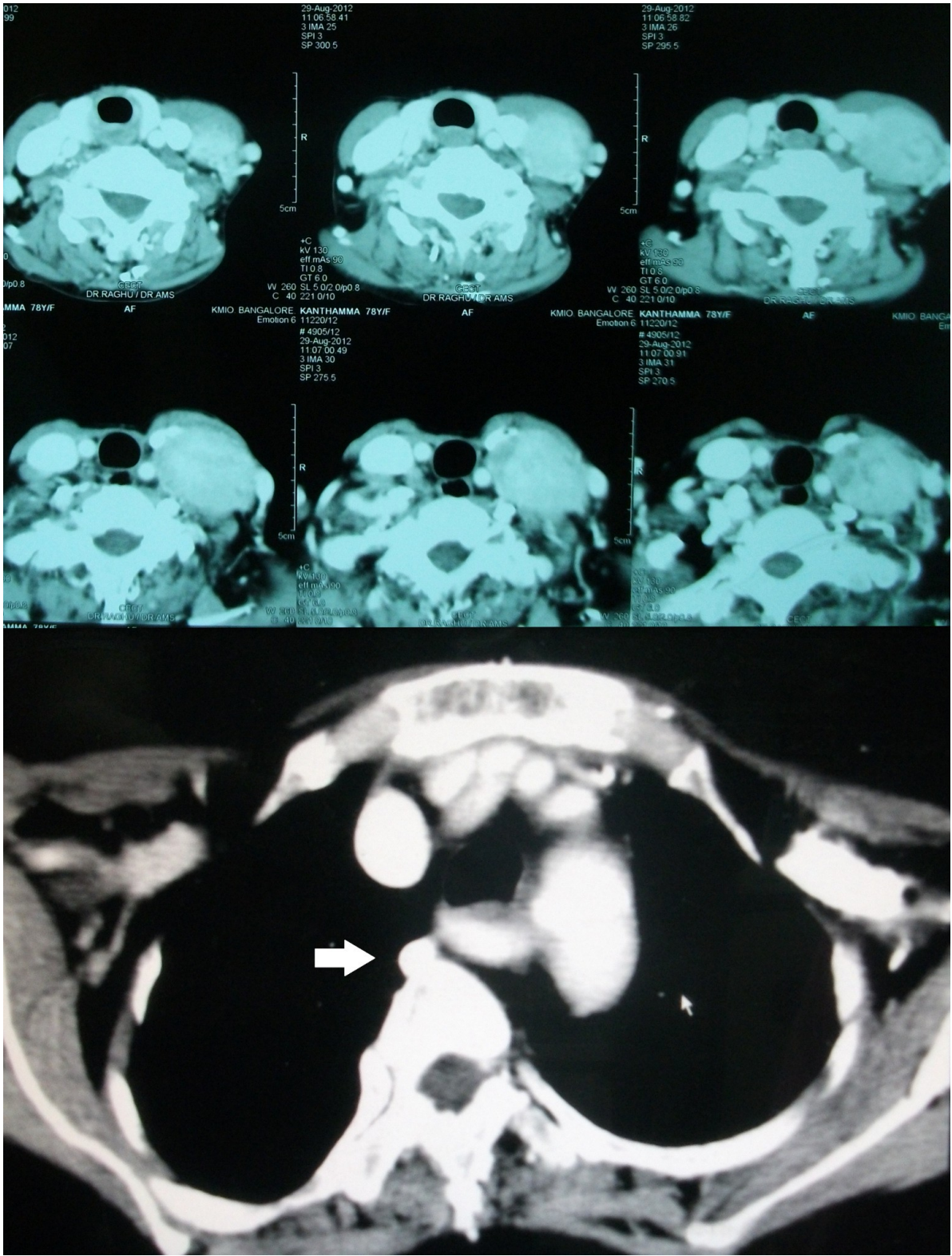


Fig 5 CT scan of chest reviewed with arteria lusoria

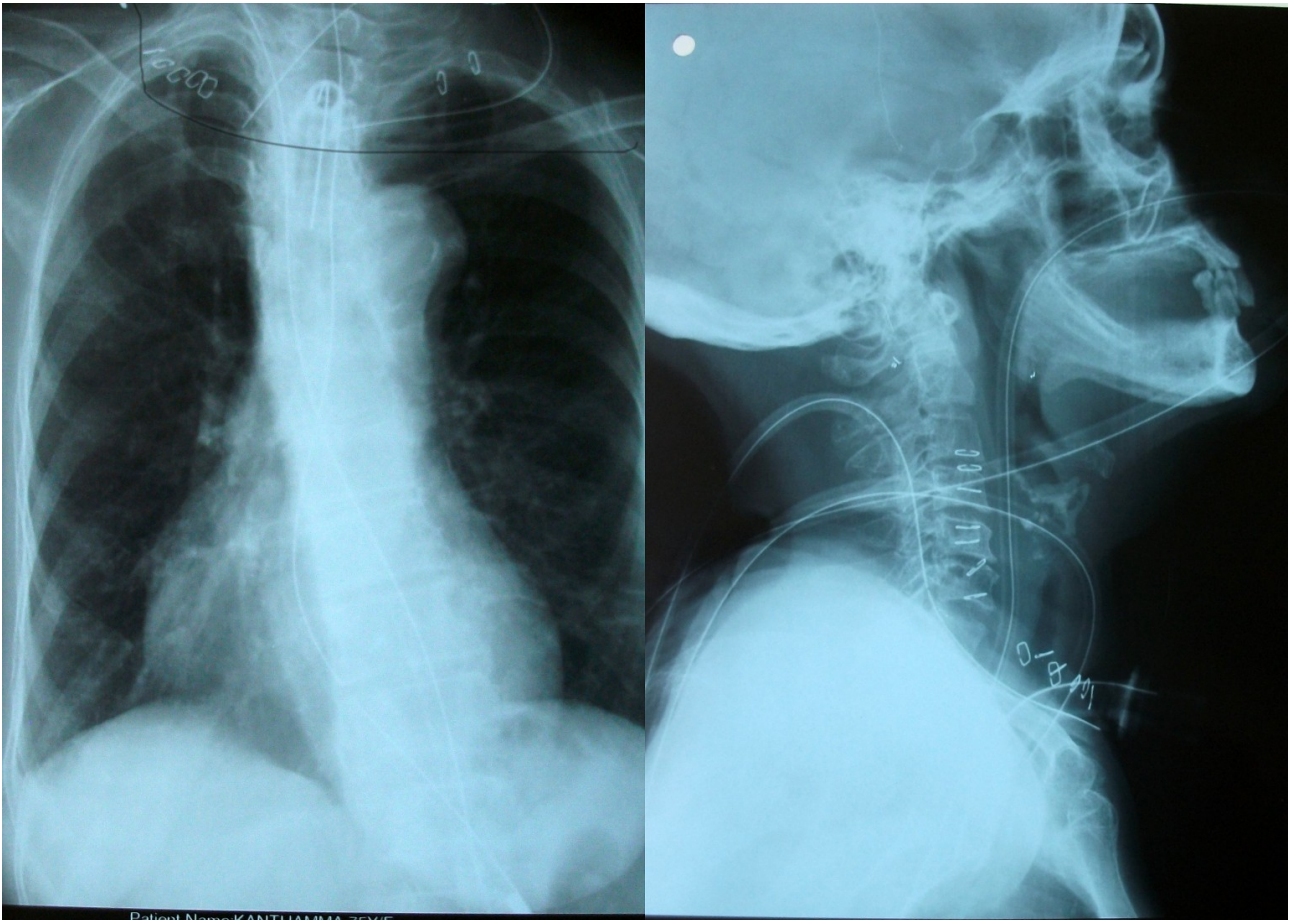


Fig 6 Post operative chest and neck evaluation being normal.

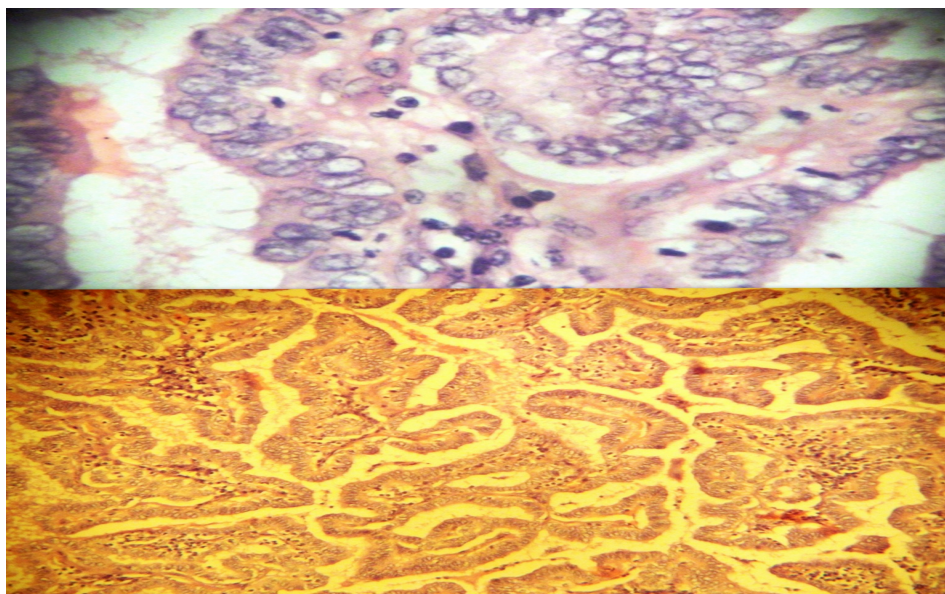


Fig 7 The node showing papillary carcinoma in low and high magnification

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