Case report

Extra-laryngeal branching of recurrent laryngeal nerve: a possibility thyroid surgeons need to keep in mind to prevent inadvertent nerve injury

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SUMMARY

A 45-year-old woman presented with a neck swelling since the past 6 years. The swelling was painless but slowly and progressively increasing in size. On examination, a thyroid swelling of size approximately 6×4 cm was seen on the left side. Ultrasonography revealed the size to be 6×5 cm with microcalcifications. Fine needle aspiration cytology revealed the mass to be papillary carcinoma. She was planned for total thyroidectomy with central compartment clearance. Preoperative assessment including thyroid hormone profile was normal and the patient was taken for surgery after proper anaesthesia clearance. During the surgery, it was found that there was a branching of the left recurrent laryngeal nerve. The branching was confirmed with nerve monitor and careful dissection was done to prevent injuries. The surgery was completed successfully without any nerve damage and the postoperative period was uneventful. Follow-up assessments revealed no signs of nerve injury like hoarseness of voice. Possibility of abnormal branches and duplication of nerves though uncommon should not be underestimated and extra vigilance and caution must be exercised when operating near a nerve, even by an experienced surgeon familiar with the local anatomy. Any doubts arising during the surgery must be clarified immediately, and nerve monitors should be used to check suspected branching and duplications before proceeding further.

BACKGROUND

General anatomy, though largely remain the same across a whole species, individual variations exist and are mostly unpredictable. Inadvertent nerve injury is always a dreaded complication of any surgery as the chances of nerve regeneration is minimal and good quality repair of nerves need extra resources and skilled hands, which may not be readily available. This is a case of recurrent laryngeal nerve (RLN) branching found during thyroidectomy. Injury to the nerve leads to the loss of vocal fold adduction and abduction leading to dysfunction during phonation, breathing and deglutition. If missed, the patient presents with hoarseness of voice during follow-up visits. With careful dissection, keeping in mind the possibility of anatomical variations and adequate exposure of the area near the nerve, surgeons can identify any aberrations and proceed accordingly. With the current overburdened and stressed out lifestyle of surgeons, it is not

unusual that one would have an urge to finish the surgery early. However skilled or familiar with the local anatomy be a surgeon, extra caution has to be taken when near a nerve and adequate time should be spent to prevent unwarranted injury to the nerve. Instruments like nerve monitors should be made accessible immediately if needed during any surgery with a risk of nerve injury and one should not hesitate to ask for opinion or use nerve monitors in any case of doubt or confusion regarding possible anatomical anomaly of a nerve.

CASE PRESENTATION

A 45-year-old woman came to the office with chief complaints of a painless, progressively enlarging neck swelling since past 6 years. The swelling was mainly on the left side of the neck and had increased in size mainly over the past year. Apart from cosmetic changes and discomfort due to the mass, she had no major complaints. She denied any history of weakness, anxiety, palpitations, anorexia, weight changes, bowel or bladder disturbances and menstrual irregularities. She did not have any history of hypertension, diabetes, thyroid disorders or any allergies. She had no previous surgeries or major hospitalisations. Family history was significant for hypertension in mother and coronary artery disease in father. There was no family history of any thyroid diseases.

On examination, there was a non-tender swelling of size 6×4 cm on the left side of the neck anteriorly. It was firm in consistency and all the borders could be defined. The swelling was moving up with deglutition and no movement was observed with tongue protrusion, suggesting a thyroid swelling. The swelling was mobile along both vertical and horizontal planes. There was no local rise of temperature or nodules on palpation. No bruit was heard on auscultation. The right lobe was normal on palpation. No Significantly enlarged cervical lymph nodes were palpable on either side. Vital signs were normal and a review of system was negative. Complete blood count with thyroid hormone profile was normal. Subsequently an ultrasound was done which revealed a 6×5 cm mass in the left lobe of thyroid with irregular margins and areas of microcalcifications. Fine needle aspiration cytology (FNAC) revealed papillary thyroid carcinoma. She was further planned for a total thyroidectomy with central compartmental clearing. All the basic

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Figure 1 The left recurrent nerve as seen after complete careful dissection.

investigations and anaesthetic clearance was obtained before the surgery.

Investigations

- ▶ Routine blood work including complete blood count and basic metabolic panel: normal.
- ▶ Thyroid hormone profile: TSH: 2.4 mIU/L, free T4: 0.9 ng/dL, free T3: 356 pg/mL.
- ▶ Ultrasonography: a 6×5 cm mass with irregular border seen in the left thyroid lobes with mildly increased vascularity. Multiple foci of microcalcifications were noted. No evidence of retrosternal extension was seen. Suggestive of: papillary thyroid carcinoma.
- ▶ FNAC: papillary thyroid carcinoma.
- ▶ Other investigations like ECG and chest X-ray needed for anaesthesia clearance was normal.
- ▶ Postoperative radioactive iodine uptake scan: no evidence of residual thyroid tissue was seen.

TREATMENT

The patient was planned for total thyroidectomy with central compartmental clearance. The surgery went uneventfully until the dissection of the left thyroid lobe. The left RLN was identified and carefully separated. While dissecting the area surrounding the nerve, another structure was found which looked like a nerve. Since the left RLN was already identified, this finding was unexpected. A nerve monitor was used to check the structure and it was found to be a branch of the left RLN with some motor fibre components. Further dissection was done with caution and the nerves were traced superiorly and inferiorly. Now the two

Figure 2 Two branches of the left recurrent laryngeal nerve.

Figure 3 Extra-laryngeal branching point of the left recurrent laryngeal nerve.

nerves were clearly defined (figure 1). Two branches of the RLN were seen on the left side with the lateral branch at the cricothyroid junction and the medial branch just lateral to midline piecing the cricothyroid membrane (figure 2). The branching point was seen approximately 2 cm from the inferior rim of the thyroid cartilage (figure 3). Extra caution was taken to avoid any inadvertent injury to either branch. The anatomy on the right side was as expected and dissection was done as usual. The remaining surgery was uneventful and the patient was shifted to the inpatient ward.

OUTCOME AND FOLLOW-UP

The postoperative period was uneventful and follow-up visits were scheduled after the discharge. Radioactive iodine uptake study revealed no residual tumour. She was started on 150 µg levothyroxine to keep the thyroid-stimulating hormone (TSH) suppressed. She was followed up for 3 months. No complications like hoarseness of voice were reported. Fibre optic laryngoscopy done revealed normal vocal cord movements on either side.

DISCUSSION

A meta-analysis conducted by Henry *et al*¹ in 2016 on extra-laryngeal branching (ELB) of the RLN mentions an overall pooled prevalence of 60.0% (95% CI 52.0 to 67.7). The study further revealed a 39.2% (95% CI 29.0 to 49.9) prevalence of ELB as reported from intraoperative studies. Of the 19 studies analysed, 61.3% (CI 49.1 to 72.4) had no branching. Bifurcation was the most common pattern with a 37.6% (CI 26.2 to 49.4) prevalence intraoperatively. Trifurcation and multiple branches were less common with an intraoperative prevalence of 1.0% (CI 0 to 4.1) and 0.1% (CI 0 to 1.7), respectively. The study further goes on to report that, when present, the branching occurred commonly within 2 cm from the inferior rim of cricothyroid joint (90.2%). Five studies reported the presence of a motor signal in both branches of the bifurcated RLN with a prevalence of 99.9% (CI 99.7 to 100) positive motor signal obtained from the anterior branch. Whereas, a positive motor signal was obtained only in 1.5% (CI 0.1 to 3.9) of the posterior branches. The study concludes with the message that there were significant differences in the prevalence of ELB between cadaveric studies (73.3%) and intraoperative studies (39.2%) suggesting the possibility of gross underestimation of the prevalence of ELB intraoperatively. Though almost 100% of the anterior branches had a positive motor signal, and only 1.5% of the posterior branch, it was found on further analysis that up to 3.9% of the posterior branches could contain motor fibres and hence care must be taken to protect all the branches whenever possible.

In most cases, the RLN courses between the two branches of the inferior thyroid arteries; but due to anatomic unpredictability, the inferior thyroid artery cannot be considered as an anatomic landmark to identify the RLN. A report by Rustad² mentions ELB of RLN to be highly unpredictable and that the branching point can occur at any point peripheral to its point of origin from the parent vagus nerve. The report further reiterates that there is no consistency in the nerve patterns between the

Patient's perspective

It has been roughly 6 years since this swelling has been in my neck. It did not cause much trouble to me and so I did not go to a doctor to get it checked up. However, my relatives and I have noted that the swelling has been increasing in size recently, maybe over the past year or so. I've started to get some discomfort with it like a weird feeling when swallowing and I was uncomfortable with the way it looked on my neck it could be clearly seen to other people. When I went to the doctor for my checkup, he did a detailed physical examination and asked me about any other symptoms that I might be having. I mentioned that I was fine except for the discomfort caused by the mass. So as discussed, we decided for and ultrasonography. I was quickly notified by my doctor that the ultrasonogram was concerning for possible cancer and hence a fine needle aspiration was to be done. When the results of the cytology came, I was informed that it was suggestive of cancer and the thyroid gland had to be removed. It was kind of a shock since I was feeling all fine. The doctors informed me that we possibly caught it early and we could take an aggressive surgery to ensure complete removal. They also informed me of the need to take thyroid supplements lifelong after the surgery and I agreed since taking a supplement was much better than risking cancer spread and complications. Therefore, we proceeded for the surgery as planned. After the surgery while in recovery, the doctors informed me of the interesting finding during the surgery. They told me that one of the nerves in my neck was having an abnormal branch not usually seen in other people. They told me that the nerve was responsible for my voice and an accidental injury to the abnormal branch could have impaired my voice. I was glad when they explained to me how they took extra time and effort to prevent injuries while at the same time being aggressive enough to remove all possible cancer. The doctors asked me about sharing this incident with the medical community by writing a case report and showed me the photos they took during the surgery. I gave them my consent for the same and I felt good that this report could reach other doctors throughout the world and would help other patients too. After the discharge, a nuclear iodine imaging was done and the doctors informed me that there was no trace of any residual cancer. I started taking the thyroid supplements and was asked to keep a check on my voice to look for any changes. During the follow-up visits, they examined my voice box and reassured me it was working as expected. We were very delighted that the surgery went smoothly and there was no accident. I feel better now and is relieved that I made the right choice in going with the surgery.

Learning points

- ▶ Injury to the recurrent laryngeal nerve (RLN) is one of the most challenging and dreaded complications in thyroid surgery. 6% experience temporary deficit and 1% experiencing permanent nerve palsy.^{4,5}
- ▶ With almost over half of patients having some element of extra-laryngeal branching, all possible prevention and utmost care has to be taken to avoid iatrogenic nerve injury to any of the branches.
- ▶ Though not all surgeons recommend complete dissection of the RLN as it may require more invasive procedure,⁶ we recommend complete exposure of the RLN along with any branches be attempted during the surgery.
- ▶ Intraoperative nerve monitors should be readily available and used in all instances when in doubt or due to restricted visualisation of the nerve due to underlying pathologies like large goitre or inflammation.
- ▶ Extra caution and high vigilance are needed during minimal invasive thyroid surgeries keeping the possibility of abnormal nerve branching.

right and left side and that even secondary/tertiary branching of the RLN can occur extra-laryngeally. Similar result was obtained in another study by Sun *et al*³ which reiterate that the relations of the RLN to the inferior thyroid artery, the inferior horn of the thyroid cartilage and the thyroid gland are inconsistent.

To conclude, the branching and course of the RLN is highly variable and unpredictable. Hence, utmost care should be taken by surgeons while operating to correctly identify the RLN and its extra-laryngeal branches to prevent any injuries.

Contributors SS was the consultant who performed the surgery and was assisted by AN. GG, intern observed the surgery. The overview of the case with intraoperative findings was explained by SS. GG wrote the case description with contributions from SS and AN. The final draft was proofread by SS and AN.

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