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MAIN ARTICLE

A novel technique to identify the nerve of origin in head and neck schwannomas

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Abstract

Objective: Identifying the nerve of origin in head and neck schwannomas is a diagnostic challenge. Surgical management leads to a risk of permanent deficit. Accurate identification of the nerve would improve operative planning and patient counselling.

Methods: Three patients with head and neck schwannomas underwent a diagnostic procedure hypothesised to identify the nerve of origin. The masses were infiltrated with 1 per cent lidocaine solution, and the patients were observed for neurological deficits.

Results: All three patients experienced temporary loss of nerve function after lidocaine injection. Facial nerve palsy, voice changes with documented unilateral same-side vocal fold paralysis, and numbness in the distribution of the maxillary nerve (V2), respectively, led to a likely identification of the nerve of origin.

Conclusion: Injection of lidocaine into a schwannoma is a safe, in-office procedure that produces a temporary nerve deficit, which may enable accurate identification of the nerve of origin of a schwannoma. Identifying the nerve of origin enhances operative planning and patient counselling.

Key words: Head; Neck; Schwannoma; Neurilemmoma

Introduction

Schwannomas are peripheral nerve sheath neoplasms arising from Schwann cells, and up to 45 per cent are found in the head and neck.[1] It is rare for patients to present with nerve deficits that indicate the nerve of origin, which may include hoarseness with schwannoma of the vagus nerve, or Horner's syndrome with schwannoma of the cervical sympathetic chain.[2] The management of these tumours is generally surgical, but surgery carries a risk of permanent deficit of the nerve of origin, even with conservative enucleation and preservation of nerve fibres.[3–5]

Pre-operative diagnosis of the nerve of origin is important for surgical planning and patient counselling. Patient counselling is especially critical when there is a risk of permanent deficits to critical structures such as the vagus nerve or facial nerve. In the case of cervical schwannomas, radiological evaluation of the pattern of great vessel displacement can enable diagnosis of the nerve of origin in most, but not 100 per cent of cases.[6–8] Additionally, schwannomas may arise from other cranial nerves, the brachial plexus or peripheral nerves, and the nerve of origin may be less clear in these cases.

In this study, a novel technique for identifying the nerve of origin was investigated. It was hypothesised that injection of local anaesthetic directly into a schwannoma would produce a temporary palsy of the nerve of origin. This may help in identification of the nerve of origin; in addition, patients would be able to experience the nerve deficit in question and make a fully informed decision regarding potential surgery. Objectives included describing the efficacy and safety of this technique.

Materials and methods

This paper describes a retrospective case series of three patients with head and neck schwannomas who presented to our institution from 2014 to 2016. Patients presented with head and neck masses. The diagnosis of schwannoma was confirmed by needle biopsy in two cases and highly suspected by imaging characteristics in the third case. All patients who underwent lidocaine injection evaluation were included, and no patients were excluded. Patient charts were reviewed for examination of: presenting characteristics, radiological evaluation findings, pathological diagnosis, neurological deficits and adverse effects of lidocaine

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injection. This study was approved by the University of Nevada School of Medicine Institutional Review Board.

Technique

After obtaining proper informed consent, masses were infiltrated with 1–2 ml of plain 1 per cent lidocaine. In the case of superficial parotid or cervical masses, injection was performed by direct palpation of the mass and to a depth based on available imaging. In the case of a nasal cavity mass, injection was performed under direct visualisation using a flexible laryngoscope with an injection port. Patients were then observed for symptoms of neurological deficits and for return of neurological function. In the case of cervical schwannoma, hoarseness prompted evaluation of laryngeal function through flexible fibre-optic laryngoscopy. Repeat flexible laryngoscopy was performed after 30–45 minutes to ensure complete return of function. In the case of sensory deficits, thorough sensory testing was performed to determine the distribution of the neurological deficit.

Results

Patient one

A 39-year-old male presented with a history of a slow-growing, asymptomatic mass of the right parotid gland. On examination, there was no weakness of the facial nerve.

A computed tomography (CT) scan showed a well-circumscribed, $5.5 \times 2.5 \times 2.1$ cm cystic right parotid lesion, with a possible diagnosis of first branchial cleft cyst (Figure 1). There was no hypermetabolic uptake on a positron emission tomography scan conducted prior to referral. Ultrasound-guided fine needle aspiration was ordered after evaluation in our department.

On routine administration of plain 1 per cent lidocaine into the skin and subcutaneous tissues, and the superficial portion of the mass, the radiologist observed immediate facial nerve paralysis. Concerned, the procedure was aborted. The patient was observed, and, 1 hour later, complete recovery of the facial nerve palsy was noted. He was brought back for a second attempt. Paralysis of the facial nerve again developed after lidocaine injection, but fine needle aspiration biopsy proceeded.

Cytology showed spindle cells, and immunohistochemistry was positive for S-100 and negative for smooth muscle actin, consistent with schwannoma.

The facial nerve was the suspected nerve of origin. The patient was counselled that a permanent facial nerve deficit was a risk of surgery, and he elected for observation rather than resection.

Patient two

An 18-year-old female presented with a painless right neck mass, which had been slowly enlarging for the past year. She desired removal for cosmesis.

FIG. 1

Axial, contrast-enhanced computed tomography image of patient one, a 39-year-old male with a right parotid mass. After lidocaine injection, the patient had immediate, temporary right facial nerve paralysis.

Ultrasound showed a $3.9 \times 3.1 \times 2.5$ cm, well-circumscribed mass with small cystic areas. Fine needle aspiration findings raised suspicion of schwannoma. Immunohistochemical stains were strongly positive for S100 and negative for cytokeratin AE1/AE3. The CT and magnetic resonance imaging (MRI) characteristics were consistent with schwannoma (Figure 2). On CT, there was some separation of the internal jugular vein and common carotid artery, which is most consistent with schwannoma of the vagus nerve.

Lidocaine injection was performed to attempt to further confirm the nerve of origin. Prior to the procedure, flexible fibre-optic laryngoscopy was performed, and this showed normal vocal fold motion bilaterally. After anaesthetising the skin, the schwannoma was grasped firmly and immobilised, while a 30-gauge needle was advanced into its capsule. Two millilitres of 1 per cent lidocaine were infiltrated into the mass. The patient began coughing immediately, and her voice was suddenly hoarse. Repeat flexible fibre-optic laryngoscopy revealed paresis of the right vocal fold. Her hoarseness resolved after 20 minutes, and subsequent laryngoscopy showed normal vocal fold motion (Figure 3).

After discussion of resection and potential future vocal fold medialisation, the patient decided to forego surgical management.

Patient three

A 55-year-old female presented with a 4.1 cm mass of the left pterygopalatine fossa, with extension to the middle cranial fossa and left nasal cavity.

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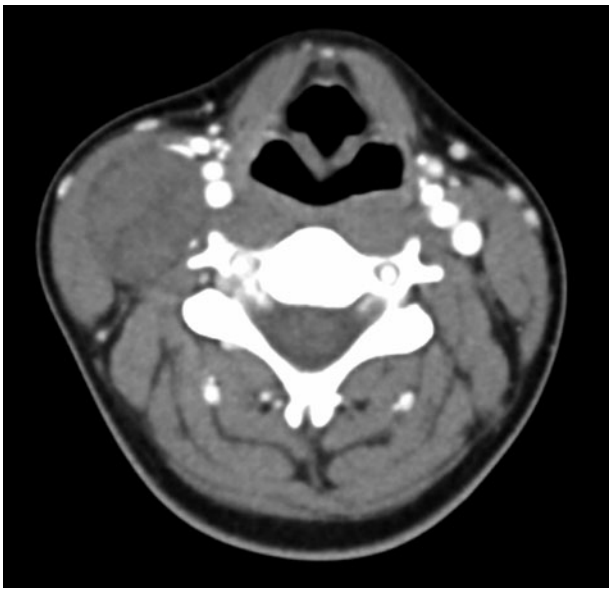

FIG. 2

Axial, contrast-enhanced computed tomography image of patient two, an 18-year-old female with a 4 cm right neck mass. When this mass was injected with lidocaine, the patient had immediate, temporary right vocal fold paralysis.

A biopsy was positive for S-100 immunohistochemical staining, and negative for cluster of differentiation 1a and carcinoembryonic antigen. On MRI, the mass had internal cystic spaces, was of intermediate intensity on T1-weighted imaging, with avid enhancement, and was heterogeneously hyperintense on T2-weighted imaging (Figure 4). These findings were consistent with a diagnosis of schwannoma. The patient only complained of left nasal congestion, with no cranial nerve deficits.

A flexible laryngoscope with a side port for injection was used to directly visualise the mass protruding into the nasal cavity. Under visualisation, 1 ml of 1 per cent lidocaine was injected into the mass. The patient experienced burning followed by numbness in the maxillary (V2) distribution of the trigeminal nerve.

A biopsy was not performed, but a presumptive diagnosis of trigeminal schwannoma was made in light of the imaging characteristics, extension of the mass to the middle cranial fossa, and the V2 nerve deficit on lidocaine injection.

The patient has elected for observation of the mass at this time.

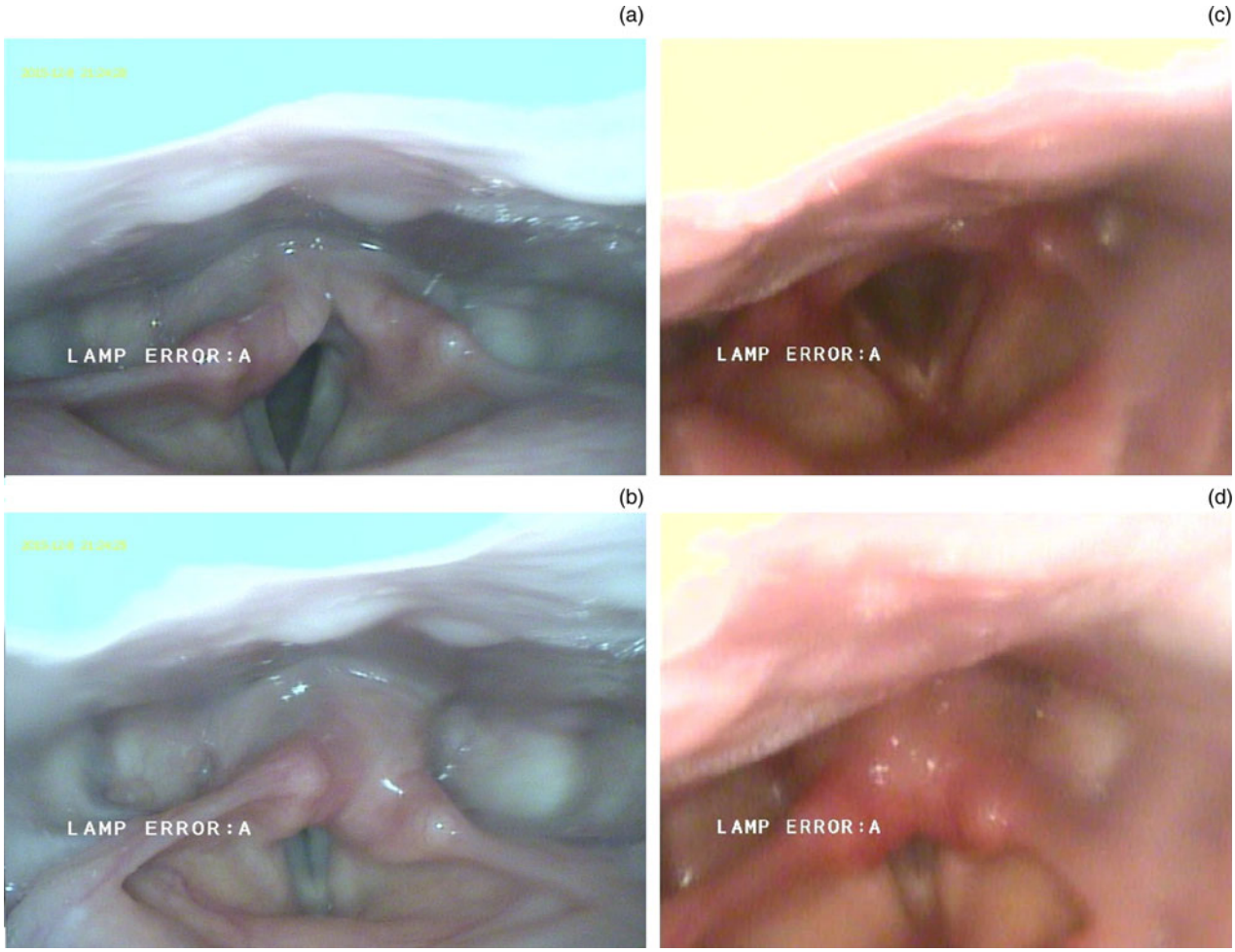

FIG. 3

Flexible laryngoscopy in patient two with right neck mass consistent with vagal schwannoma. Abduction (a) and adduction (b) of true vocal folds less than 5 minutes after lidocaine injection of the neck mass, indicating paralysis of the right vocal fold. Complete return of laryngeal function and resolution of hoarseness occurred 45 minutes after lidocaine injection (c and d). Note the symmetry of the arytenoids on adduction in (d) compared to (b) with return of vagus nerve function.

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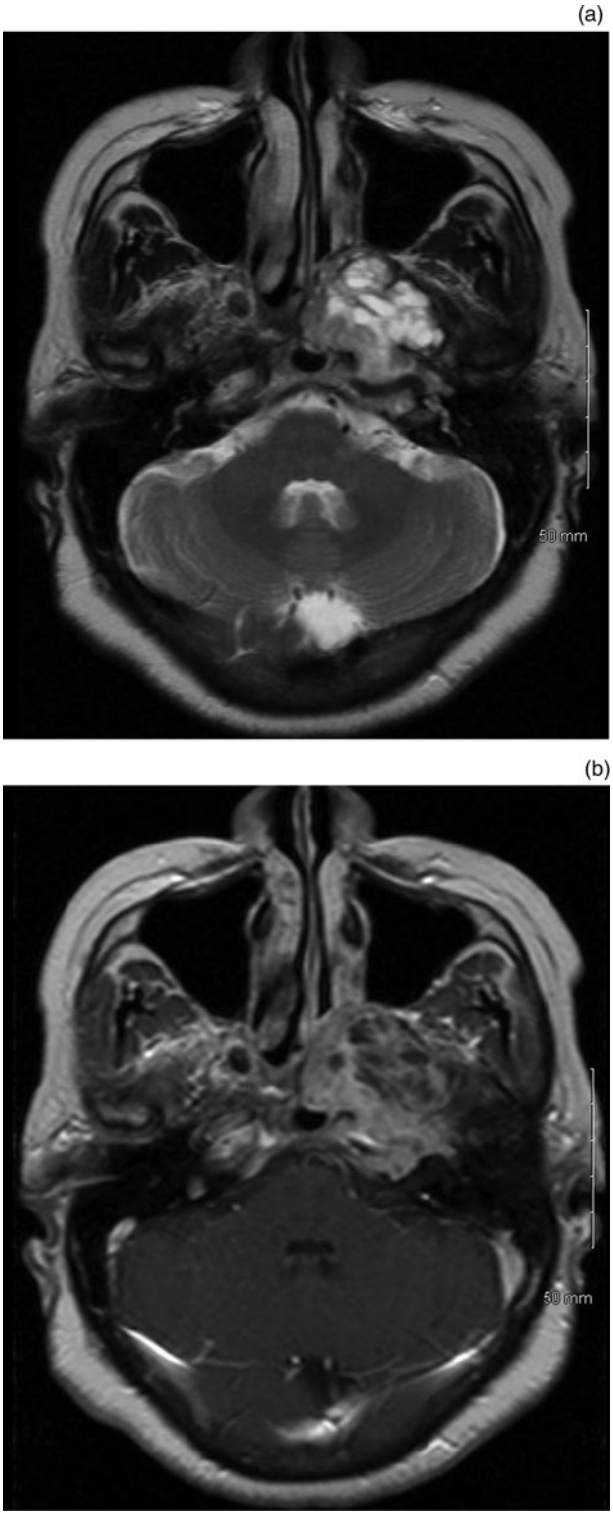

FIG. 4

Axial, magnetic resonance imaging (MRI) of patient three, a 55-year-old female with a 4.1 cm mass of the left pterygopalatine fossa. (a) T2-weighted MRI of the mass showed heterogeneous hyperintensity. (b) T1-weighted post-gadolinium MRI showed enhancement with internal cystic spaces, consistent with schwannoma. After lidocaine injection, the patient had immediate, temporary paraesthesia and numbness in the maxillary distribution of the trigeminal nerve.

Complications

There were no long-term sequelae or complications associated with these procedures. Each patient tolerated the injection well, with minimal discomfort.

Discussion

Head and neck schwannomas are often asymptomatic and present a diagnostic challenge. Even if a high suspicion for schwannoma is present based on cytology and imaging characteristics, determination of the nerve of origin may be difficult. In the head and neck, possible extracranial nerves of origin include the cervical sympathetic chain, brachial plexus, and the Vth, VIIth, Xth, XIth and XIIth cranial nerves. The major consideration in resection or enucleation of schwannomas is permanent paralysis of the nerve of origin.

In this study, a novel technique of pre-operative lidocaine injection is presented, which may be a reliable method to identify the nerve of origin. In the three cases presented, patients tolerated the procedure well, with no complications. In addition to identification of the nerve of origin, patients are able to temporarily experience the possible nerve deficit first-hand, which may allow them to make a fully informed decision regarding surgery. Furthermore, unexpected nerve deficits after superficial lidocaine injection into a head and neck mass may make the clinician more suspicious of a schwannoma diagnosis, thereby more optimally guiding imaging and immunohistochemistry investigation.

The patients in this series decided to defer surgery, and therefore the nerve of origin could not be confirmed by intra-operative findings or post-operative nerve paralysis. However, there are several findings that support the validity of this technique as an accurate identifier of the nerve of origin. First, the onset of the nerve deficit was within minutes in each case, with a low volume injection of 1 per cent lidocaine, which makes it unlikely that lidocaine simply diffused towards the nerve in question. Second, in each case there was a complete deficit of a single nerve distal to the nerve in question, rather than a partial deficit or deficits of limited branches of the nerve. Lastly, the injection in each case was not performed in close proximity to the location of the presumed nerve of origin. For example, in the case of the schwannoma of the vagus nerve, lidocaine was injected about 1–2 cm from the skin surface, which was not deep within the mass. The fact that such a superficial injection, distant from the normal location of the vagus nerve, created a vocal fold paralysis is evidence that the mass is arising from or directly continuous with the vagus nerve.

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Several studies have identified radiological patterns for differentiating the nerve of origin in cervical schwannomas, which most commonly arise from the vagus nerve or sympathetic chain. In 1996, Furukawa *et al*. proposed that vagal nerve schwannomas separate the internal carotid artery and internal jugular vein, while sympathetic chain schwannomas do not.[6] The nerve of origin was accurately diagnosed in all nine patients analysed using that criteria. Since then, multiple studies have found that this criteria may diagnose the nerve of origin in most but not all cervical schwannomas.[5,7,9] A recent large series and meta-analysis by Graffeo *et al*. found that the internal carotid artery/internal jugular vein splaying criterion has a 75–87 per cent probability of identifying the nerve of origin correctly.[8] However, the authors proposed other imaging criteria, in a combined model, to increase this accuracy.

- **Surgical management of head and neck schwannoma carries a risk of permanent injury to the nerve of origin**
- **The nerve of origin may be difficult to definitively identify using imaging techniques**
- **A diagnostic technique of lidocaine injection into schwannomas was performed in a small number of patients**
- **Lidocaine injection produced temporary deficit of a single nerve, consistent with the expected nerve of origin based on imaging**
- **This technique may help to confirm or identify the nerve of origin in head and neck schwannomas**

This study has several limitations. As described above, we were unable to confirm the nerve of origin in these patients intra-operatively or by post-operative deficits, which would be the 'gold standard' method for identification. Future studies would be required to further confirm the accuracy of this technique. This technique has only been conducted in a small number of patients, but we have shown that it can be performed in a variety of locations where head and neck schwannomas are found. Lidocaine injection in this manner can be performed easily and safely in the otolaryngology out-patient clinic.

Conclusion

Injection of lidocaine into a suspected schwannoma is a safe, in-office procedure that produces a temporary single nerve deficit, which may enable accurate identification of the nerve of origin of a schwannoma. Identifying the nerve of origin can enhance both operative planning and patient counselling.

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