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Original Research Article

A rare presentation of Intralaryngeal Schwannoma and the study of 50 cases of intralaryngeal Schwannoma from the literature

Raghavendra Prasad K. U.*, Shreyanka M. Doreswamy, Manohar S. Ramanaik

Department of Otorhinolaryngology, Hassan Institute of Medical Sciences, Karnataka, India

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***Correspondence:**
Dr. Raghavendra Prasad K. U.,
E-mail: prasadusha45@gmail.com

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ABSTRACT

Background: Laryngeal schwannoma is a rare, slow-growing, benign neurogenic tumour usually located in the aryepiglottic folds. The aim is to report a rare case of laryngeal schwannoma and to review 50 cases of a similar entity from the literature in terms of age, sex, presenting symptoms, site, size, the nerve of origin, mode of surgery and outcome.
Methods: A retrospective study was conducted at the Department of Otorhinolaryngology, in a tertiary care centre along with a review of cases from the literature. A detailed description of the case of laryngeal schwannoma in a 20-year-old patient, who presented with difficulty in swallowing, dyspnoea and hoarseness in the voice to the department of otorhinolaryngology will be given. Along with the data on a comprehensive literature search on about 50 cases of laryngeal schwannomas.
Results: Out of the 50 cases, 54% were females and the majority of the cases were above the age of 55 years. Aryepiglottic folds were the common site of tumours (38%). Other sites where the tumour was found included the false cords, true vocal cords, ventricle, arytenoids, epiglottis, post cricoid region, trachea, and thyroid gland. All cases were treated surgically.
Conclusions: Laryngeal schwannomas are rare, nerve sheath tumours usually located in the supraglottic larynx. The surgical approach depends on the site, size and extent of the tumour. Sparing the parental nerve depends on the size of the tumour and the nerve of origin.

Keywords: Schwannoma, Larynx, Neurogenic tumour, Supraglottis

INTRODUCTION

Schwannomas are extremely rare neurogenic tumours, about 25-45 per cent of them arising in the head and neck region[1] and account for 0.1 to 1.5 per cent of all benign laryngeal tumours.[2] Most originate from the vagus nerve, with involvement of the parapharyngeal space.[3] In the larynx, 80 per cent are located in the aryepiglottic fold, 20% in the false or true vocal cords.[2] These are the nerve sheath tumour, that emanates from perineural Schwann cells and grows extrinsic to the parental nerve fascicles.[4] They are well-encapsulated tumours that grow submucosally, with a slow annual growth rate of 2.75 mm to 3 mm.[5,6] Few occurrences of polypoidal growth have also been reported.[2] They are presumed to arise from the internal branch of the superior laryngeal nerve.[7] The malignant transformation in the head and neck schwannomas vary from 8 to 13.9 per cent.[8,9] The objectives of our study were to report a rare case of a 20-years old female patient who presented to the department of otorhinolaryngology with difficulty in swallowing and dyspnoea. We would also like to present 50 cases of laryngeal schwannomas reviewed from the literature.

METHODS

The descriptive study was conducted at the department of otorhinolaryngology, HIMS, Hassan, Karnataka, India, from October 2021 to April 2022. A single case of a 20-years old female patient, who was diagnosed with laryngeal schwannoma and managed at our tertiary care centre was included in our study after obtaining informed and written consent from the patient. A broad literature search was carried out online from different indexed journals (using PubMed and Google scholar) during the study period of 7 months and 50 diagnosed cases of laryngeal schwannomas with varied presentations were included. All cases of head and neck schwannomas other than laryngeal presentations were excluded from the study.

The sample size was calculated using a simple random method and the data collected were subjected to statistical analysis using statistical package for the social sciences (SPSS) software 20.2.

The results were expressed in terms of frequency and percentage. The keywords used were larynx and schwannoma.

The clearance was obtained from the institute ethical committee.

A 20-years-old female patient presented to the department of otorhinolaryngology at our tertiary care centre complaining of difficulty in swallowing, dyspnoea on exertion, hoarseness of voice for 6 months, and snoring for 1 month. Further medical history was unremarkable. Thorough ear, nose, throat, and head and neck examinations were conducted. Indirect laryngoscopic examination revealed a 3×4 cm well-defined smooth oval-shaped mass at the vallecula towards the left side completely displacing the epiglottis to the right (Figure 1). An X-ray neck lateral view was obtained (Figure 2). Computed tomography (CT) scan showed a 3.5×3.9×3.6 cm solid cystic lesion epicentre at the left paraglottic space extending cranially till the base of the tongue displacing the epiglottis and aryepiglottic fold towards the right side. The mass was found to be completely covering the laryngeal inlet, vocal cords were not visualized. Contrast enhancement of the solid tissue mass was noted. Prophylactic tracheostomy was performed under local anaesthesia, followed by a direct laryngoscopic examination. On direct laryngoscopy, a smooth mass was seen in the oropharynx at the vallecula which pushed the epiglottis to the right side completely. On displacing the tumour to one side, part of the right vocal cord was seen. A biopsy was taken from the mass for histopathology. Histopathological examination under the microscope showed cellular and hypocellular areas composed of elongated spindle cells with wavy nuclei arranged in a palisaded pattern (Antoni A) and scattered patterns (Antoni B) (Figure 4).

Areas of cystic change and thrombosed blood vessels admixed with chronic inflammatory cells were seen. Subsequently, the patient underwent suprahyoid anterior pharyngotomy with schwannoma excision under general anaesthesia (Figure 3). Postoperative recovery was smooth. The patient was on Ryle's tube feeding for 8 days and tracheostomy tube for 5 days. Laryngeal oedema persisted for 4 weeks. Postoperatively laryngoscopic examination showed normal vocal cords bilaterally.

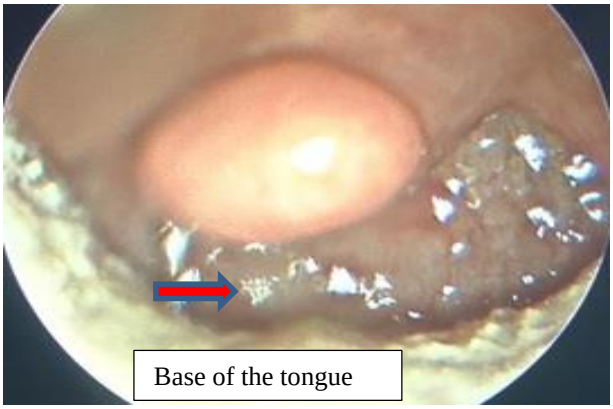

Figure 1: Direct laryngoscopic image showing well-defined smooth mass (arrow) completely obstructing the laryngeal inlet.

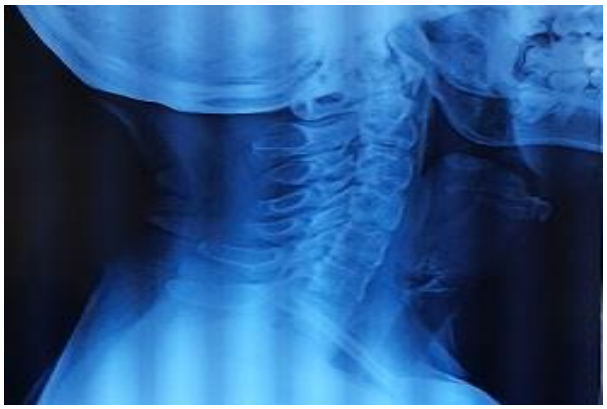

Figure 2: X-ray neck lateral view showing a mass in the larynx.

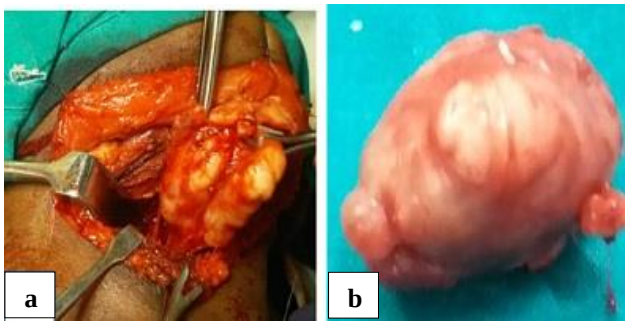

Figure 3: Intra operative images (a) showing the delivery of the Schwannoma, and (b) tumor after excision.

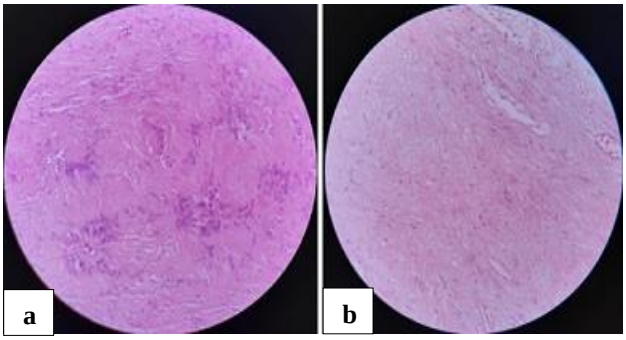

Figure 4: Histopathology (a) 40X image- hypercellularity with Verocay bodies and Antoni A areas and (b) 40X image Antoni B regions.

RESULTS

Summary of the 50 cases of laryngeal schwannoma from the literature review.

Of the 50 cases from the literature, 23 (46 %) were males and 27 (54%) were females. The early presentation was noted at 8 years of age. 16 out of 50 occurred in patients above the age of 55 years followed by 13 cases between the age group of 35-45 years. The most common presentation was hoarseness of voice (68%) followed by dysphagia/ odynophagia (26%) and breathing difficulty (24%). Other symptoms noted were foreign body sensation/ globus sensation in the throat (16%), mass in the neck (8%) and snoring (6%).

Two patients were asymptomatic. The duration of symptoms ranged from one month to more than 20 years. Thirty-two per cent of the cases presented within 1 year and the duration of the symptom was not documented in another 32% of the cases. The aryepiglottic fold is the most common site of the tumour (38%) in our literature search followed by the ventricular fold (22%) and glottis (12%). Other sites of presentation noted were epiglottis (6%), ventricle (2%), trachea (2%), thyroid gland (2%), post cricoid region (2%), arytenoids (6%) and pyriform fossa (6%). One patient (2%) presented with a neck mass with a normal endolarynx.

The size of the tumour noted was ranging from 0.3 cm to 5.8 cm. In 48% of the cases, the size of the tumour was reported to be between 2 to 4 cm. Twenty per cent of the patients had a size less than 2 cm and 10 per cent had more than 4 cm. In 22 per cent of the cases the size of the tumour was not mentioned. The most common site of the tumour was found to be the involvement of aryepiglottic folds (38%) followed by ventricular folds (22%) and glottis (12%). Other sites involved are epiglottis (6%), piriform fossa (6%), and arytenoids (6%). Rare locations like ventricles (2%), trachea (2%), thyroid gland (2%), post-cricoid region (2%).

One case was reported with a neck mass with endolarynx normal. All the patients were treated surgically except for one patient, where the diagnosis was made incidentally at the time of autopsy. Most of the cases were operated through an endoscopic approach (40%) followed by an open approach (22%) and lateral thyrotomy (16%). Two cases (4%) were approached through median thyrotomy, and 1 case through lateral pharyngotomy. One patient underwent sternotomy and excision of the tumour. Four patients (8%) underwent laryngectomy. Transoral robotic surgery (TORS) was reported in 2 cases.

In the majority of cases, the nerve of origin was not mentioned and, in a few cases, it was presumed to be arising from branches of the superior laryngeal nerve or recurrent laryngeal nerve. Follow-up was done for a variable period. No recurrence was reported in 70% of the reviewed cases. Eight per cent of the cases reported recurrence and in 22% of the cases, follow-up information was not documented. Six cases were reported with multiple schwannomas.

Table 1: Summarizing 50 cases of laryngeal schwannomas reviewed (age, sex).

No. of cases	Age (in years), N (%)						Sex	
	<15	15-25	25-35	35-45	45-55	>55	Male	Female
50	1 (2)	6 (12)	9 (18)	13 (26)	5 (10)	16 (32)	23 (46)	27 (54)

Table 2: Summarizing cases of laryngeal schwannoma (presenting symptoms, duration of the symptoms).

No. of cases	Symptoms, n (%)							Duration (years), n (%)			
	Hoarseness of voice/ dysphonia	Dysp-noea	Dysphagia/ odynopha-gia	Snori-ng	Foreign body/ globus sensation in the throat	Mass in the neck	Asympto-matic	<1	1-6	>6	Not menti-oned
50	34 (68)	12 (24)	13 (26)	3 (6)	8 (16)	4 (8)	2 (4)	16 (32)	13 (26)	6 (12)	16 (32)

Table 3: Summarizing cases of laryngeal schwannoma (size and location of the tumour).

No. of cases	Size of the tumour (cm)				Location of the tumour, n (%)										
	<2	2-4	>4	Not mentioned	Aryepiglottic fold	Glottis	Ventricular fold	Arytenoid	Trachea	Epiglottis	Pyriform fossa	Ventricle	Postcricoid	Thyroid gland	Neck mass
50	10 (20)	24 (48)	5 (10)	11 (22)	19 (38)	6 (12)	11 (22)	3 (6)	1 (2)	3 (6)	3 (6)	1 (2)	1 (2)	1 (2)	1 (2)

Table 4: Summarizing cases of laryngeal schwannoma (surgical approach and outcome).

Surgical approach, n (%)									Outcome, n (%)		
Median thyrotomy	Lateral thyrotomy	Lateral pharyngotomy	Laryngectomy	DLscopy/MLS excision	TORS	Sternotomy	Open approach	Not mentioned	No recurrence	Recurrence	Not mentioned
2 (4)	8 (16)	1 (2)	4 (8)	20 (40)	2 (4)	1 (2)	11 (22)	1 (2)	35 (70)	4 (8)	11 (22)

DISCUSSION

Schwannomas were first identified by Virchow in 1908 and later reported by Vercoy in 1910 and termed them as neurinomas.[10] It was Stout in 1949, recognized the Schwannian derivation and termed it as schwannoma.[11] Two types of neurogenic tumours must be distinguished: Schwannomas and neurofibromas. Schwannomas are the nerve sheath tumour,that emanates from perineural Schwann cells. These are well-encapsulated tumours that grow extrinsic to the parental nerve fascicles.[2] Neurofibromas are unencapsulated neurogenic tumours arising from perineural fibrocytes that are typically entangled with the parental nerve fascicles.[3,4] There is a variable predilection for sex. However, in our review literature male to female ratio was found to be 1:1.17 which is almost equal and consistent with Wong et al.[17] Schwannomas have been described among all age groups, but few authors have reported an increased incidence in the sixth and seventh decade.[18] Our reported case was a 20-year-old female patient. And in the literature review, the age ranged from 8 years to 79 years with 32% of the cases above 55 years and 2% of the cases below 15 years.

The clinical symptoms of the disease are those usually associated with a slow-growing nature of the tumour, nerve of origin, location and its extent.[12] The patient develops hoarseness of voice, globus sensation in the throat, dysphagia and dyspnoea with inspiratory, occasionally biphasic stridor with time.[2] There has been one incidence of asphyxial death caused by laryngeal schwannoma.[12] The most common presentation of schwannoma in the head and neck region is mass in the neck.[13] However, in our case, the patient had no neck swelling and instead presented with difficulty in swallowing, dyspnoea on exertion, and hoarseness of voice with recently developed snoring. Plantet et al reported a case of schwannoma with a lateral neck mass and normal endolarynx on examination.[19] One patient had an asymptomatic presentation.[20]

It mostly involves the aryepiglottic folds, but the involvement of the ventricular folds or arytenoids has also been described.[2] This is consistent with our findings in which aryepiglottic folds were more commonly involved site (38%) followed by ventricular folds (22%). Glottis was involved in 12%. Other rare sites like ventricle, epiglottis, tracheal mass, mass in the thyroid gland, neck mass, post cricoid region, and piriform fossa were noted. The tumour in our case originated from the left paraglottic space and caused airway obstruction which required tracheostomy.

Indirect and fiberoptic laryngoscopy used as a part of the diagnostic process, generally reveals a submucosal mass in the specified location.[2] CT or MRI is a valuable technique for delineating the anatomical extent of the lesion.[7] A definitive diagnosis, however, may only be made histologically. Schwannomas are characterized by spindle cells with large, oval nuclei and unclear cell membranes. The Antoni A pattern is characterized by compact, spindle-shaped cells, with their nuclei aligned in parallel rows resembling a palisade. With vacuoles and spindle-shaped nuclei, the Antoni B type is less cellular and loosely organized.[14] Fibrillary cell processes separate two rows of compactly arranged nuclei in Verocay bodies. Schwannomas are generally devoid of axons.[2]

According to Enzinger, the diagnosis of schwannoma is made by three criteria: the tumour has a capsule; it contains Antoni-A and Antoni-B areas; and the S-100 reaction is positive.[15]

The variable diagnoses for laryngeal tumours of neurogenic origin include chondroma and adenoma. Also, laryngeal cyst, intralaryngeal lipoma and internal laryngocele should be taken into consideration.[2]

Since schwannomas are benign, well-encapsulated, radioresistant tumours surgical excision is the mainstay of treatment. The approach for each case differs according to its site, presentation and extent of the tumour. Endoscopic excision can be done for small tumours, especially for a true vocal cord tumour. External approaches such as the laryngofissure approach, medial or lateral thyrotomy, lateral pharyngotomy, and laryngectomy have been described for large tumours.[14] Plantet et al, reported a case of schwannoma excision by lateral pharyngotomy approach.[19] Nagata et al reported a case of tracheal schwannoma excision by median sternotomy approach.[22] Four patients in our literature review had undergone laryngectomy (total/partial).[19,23-25] Excision of mass by transoral robotic surgery (TORS) reported by Kayhan et al and Millas et al.[26,27] The suprahyoid pharyngotomy approach was used for our reported case, as it offered the most direct route for tumour excision, avoiding the risk of injury to the vocal fold.

Schwannomas grow in the eccentric pathway and as it enlarges can compress the maternal nerve fibres which grow over the tumour capsule. As the tumour size increases, the nerve fibres splay and become thinner. Schwannoma does not engulf the nerve fibres of origin, but as the tumour grows, it results in splaying of the maternal nerve fibres. Schwannoma originated from all the cranial nerves except the olfactory nerve and optic nerve.[21] Schwannomas can also arise from the small distal branches/peripheral nerves, in which conditions identifying and preserving the nerve is difficult. In cases of laryngeal schwannomas, the nerve of origin is presumed to be an internal branch of the superior laryngeal nerve.[28] Origin of schwannomas from recurrent laryngeal nerve was also been hypothesized in the literature.[3]

Nagato described the possibility of its origin from the anastomotic branches between the internal branch of the superior laryngeal nerve and recurrent laryngeal nerve.[29] In the case of small tumours originating from the known and larger nerves, an attempt can be done to preserve the nerve of origin. Hence, in cases of large schwannomas often the nerve of origin could not be preserved and in cases of a small tumour, it is difficult to identify the nerve of origin which could result in sacrificing the nerve. Incomplete excision can lead to rapid regrowth and potentially fatal consequences. There is a time consensus given the duration of follow-up. No recurrence was found in our reported case during the follow-up. However, 4 cases from our literature review reported recurrence. Recurrence is rare if the tumour is removed completely. The limitations of our study are, it is a single case report with a review of 50 cases from the literature. Also, more cases should be included for better understanding and management of the patients. Age of the patient, presence of any comorbidities and during and post-surgery complications should be considered before planning the line of management. Regular and long-term follow-ups should be considered in view of recurrences and rehabilitation of the patients.

CONCLUSION

Laryngeal schwannoma is a rare, slow-growing, benign neurogenic tumour. The commonest location of schwannoma in the larynx is the aryepiglottic fold. The only histological examination can provide a definitive diagnosis. Complete surgical excision is the sole curative therapy option. The nerve of origin usually cannot be made, when it arises from smaller distal nerves. Sparing the nerve depends on the size of the tumour and the nerve of origin.

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