Case Series

Lateral Neck Swellings with Unusual Entities: A Case Series in a Tertiary Care Hospital

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ABSTRACT

Introduction

The neck and parapharyngeal space are one of the most vital regions in the body, encompassing multiple major blood vessels, nerves, spine and the airway itself. Lateral neck masses that present to an ENT practitioner may not only include a wide variety of differentials, but may present as emergencies in case of an airway compromise. In such situations, decision making and arriving at the diagnosis becomes important not only from a curative perspective but also from a lifesaving one. Here we discuss a few cases of lateral neck masses that had presented to us with unusual presentation or had a rare diagnosis, along with the line of management that was followed.

Cases

We present a case of a giant pleomorphic adenoma presenting with stridor, an adult onset cystic hygroma, an asymptomatic vagal schwannoma, an isolated infiltrating lipoma and a case of a sympathetic chain schwannoma presenting with dyspnea.

Results

Tumours of the neck and parapharyngeal region have a wide variety as this region contains almost every kind of tissue. Diagnosis of any lesion should be done with caution, using the appropriate history, examination and investigative tools available. Not only the common presentation, but also outliers and uncommon presentations of common tumours should be kept in mind.

Conclusion

Thus regardless of the vaccination status, adherence to personal preventive behaviours is necessary to prevent the further spread of COVID-19 infection. It can be emphasized by the competent authority by maintaining some regulations and the same message can be disseminated by health education materials to the general population with special emphasis on high risk groups.

Keywords

Neck Mass; Parapharyngeal; Schwannoma; Infiltrating Lipoma; Giant Pleomorphic Adenoma.

The neck and parapharyngeal space are one of the most vital regions in the body, encompassing multiple major blood vessels, nerves, spine and the airway itself. A wide spectrum of primary pathologies may present, whose pre operative diagnostic accuracy has greatly improved in recent years with advent of multi-planar radiological imaging such as fine-sliced computerized tomography (CT) and magnetic resonance imaging (MRI) with fat suppression sequences and gadolinium for contrast.[1] Most parapharyngeal neck tumours are benign, with an intraoral swelling being the most common presentation. The most common parapharyngeal space tumor is pleomorphic adenoma arising from deep lobe of parotid gland in the pre-styloid compartment. However, a giant pleomorphic adenoma is

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a much rarer entity. Similarly benign neurogenic lesions are the second most common type of parapharyngeal space tumour, with paragangliomas being the commonest followed by schwannomas.[2] Riffat et al found only 1 case of lymphangioma in a systematic review of 1143 over a period of 20 years, although they do not mention if it was congenital or adult onset,[2] adult onset being the exception rather than the rule. Lipomas, although very common, an isolated infiltrating lipoma of the head and neck region with no other associated symptoms, is an extremely rare entity.[3] Lateral neck masses that present to an ENT practitioner may also present as emergencies in case of an airway compromise. In such situations, decision making and arriving at the diagnosis becomes important not only from a curative perspective but also from a lifesaving one. Here we discuss a few cases of lateral neck masses that had presented to us within a period of 6 months, either with unusual presentation or had a rare diagnosis, along with the line of management that was followed.

Case Series

Case 1

A 29 year old male presented with a swelling on the left parotid region, which had been gradually increasing in size for the past 6 months. He developed dysphagia and dyspnea for the last 3 weeks with an acute exacerbation for 5 days (Fig. 1a). Patient also complained of acute pain and redness over the swelling for last 3 days. The swelling was at the parotid region, extending upto the left submandibular triangle, about 12 x 7 cm, irregular with ill defined margins and with multiple pus points on its surface. It was warm to touch, tender, firm with areas of fluctuation, an irregular surface and mobile in both axes. There was no skin fixity over the swelling and no visible or palpable pulsations.Oral cavity examination showed a significant bulge in the soft palate, obscuring the oral cavity and oropharynx.An emergency tracheostomy was performed to relieve dyspnea, and an incision-drainage was done at the maximum point of fluctuation in the parotid region, to reveal that it was a superadded abscess over a firm mass. The culture showed Burkholderia cepacia which are recognized as important opportunistic pathogens that can cause variable lung infections in cystic fibrosis patients, which result in asymptomatic carriage, chronic infection or 'cepacia syndrome'.[4]

A diagnostic nasal endoscopy (DNE) showed a smooth surfaced globular mass bulging from the lateral wall of the nasopharynx, obstructing the left choana. Plain CT neck with a contiguous contrast enhanced MRI showed a large well encapsulated heterogeneously enhancing space occupying lesion (103 x 57 mm) at the left lateral aspect of upper neck at the submandibular region with extension into oropharyngeal and nasopharyngeal cavity (Fig. 1b, 1c). The mass had areas of necrotic foci and a suspected origin from the deep lobe of parotid with widening of the stylomandibular tunnel and extension into the prestyloid parapharyngeal compartment. A fine needle aspiration cytology (FNAC) from the swelling to reach a cytological diagnosis was inconclusive (suggestive of an inflammatory lesion). A final CT- angiography done showed the mass was supplied by the branches arising from left external carotid artery (predominantly by the ascending pharyngeal, lingual, facial and post auricular arteries).

Surgical excision of the mass was done via transparotid-transcervical approach. The submandibular gland was retracted anteriorly and the posterior belly of the digastric muscle was cut to access the entirety of the mass in the parapharyngeal space. Digital counter pressure was applied via the lateral oropharyngeal wall for a better plane of dissection. Although our initial surgical planning included a midline mandibulotomy for better exposure and control(Fig. 1d), the procedure was successfully done without it. Mass was excised in toto(Fig. 1e) but the proximity to the mucosa of the oral cavity resulted in a rent near the retromolar region which was repaired intraoperatively.The specimen was sent for histopathological examination (HPE), and showed a final diagnosis of pleomorphic adenoma involving the deep lobe of the parotid. The patient was discharged after 10 days with no signs of facial palsy, and fully healed oral mucosa.

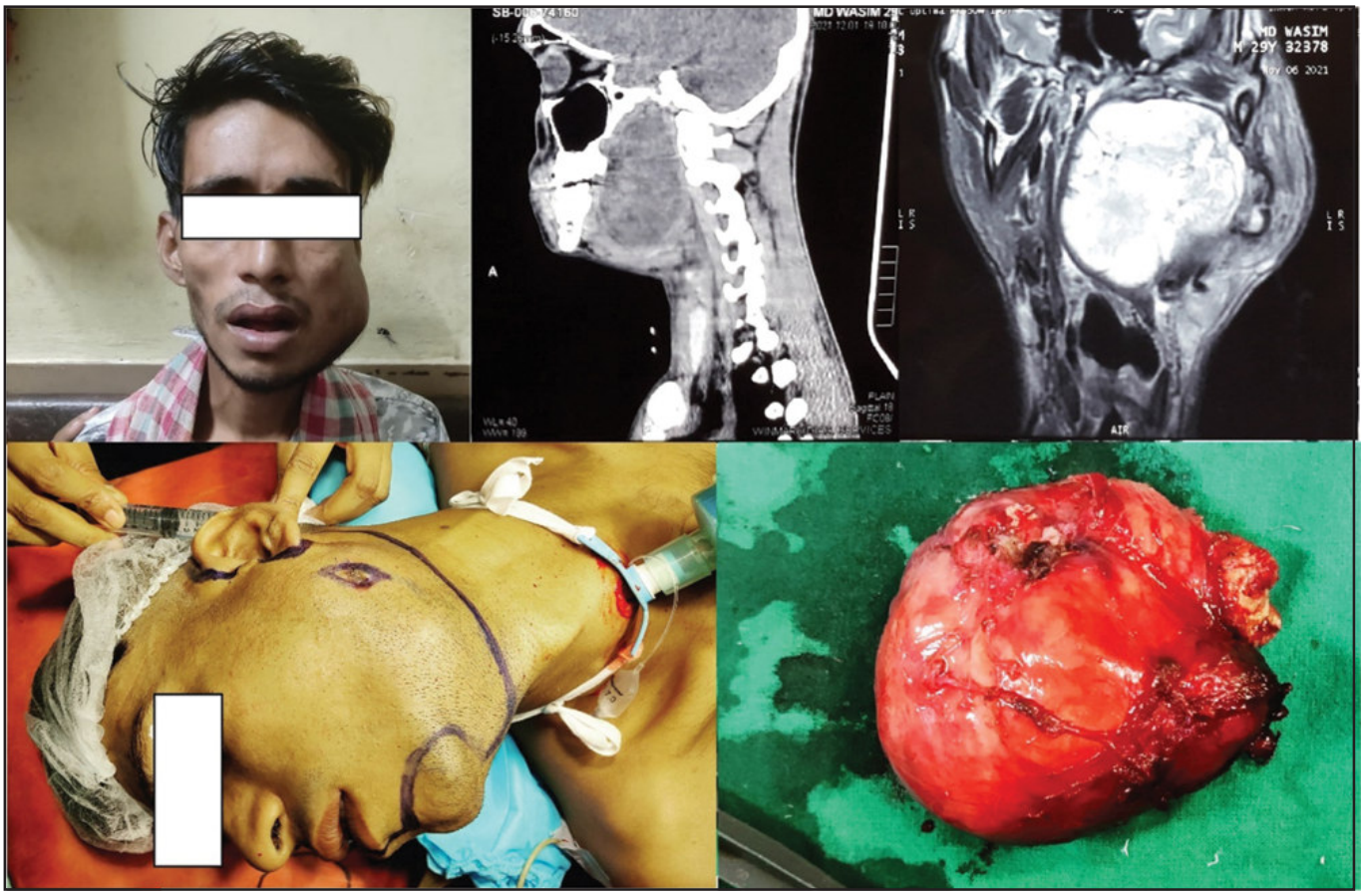

Fig. (left to right, top row) 1a. Clinical Picture of the patient; 1b. 1c. Plain CT with contiguous contrast MRI showing the extent of the lesion; (bottom row); 1d. On table marking showing planned incision for a transcervical- transparotid approach with mandibulotomy; 1e. Intra operative picture showing the lesion after removal.

Case 2

A 24 year old female presented with a mass on the right side of the neck, gradually progressing in size for last 2 years(Fig. 2a). It an approximately 8x8 cm swelling, insidious inonset, gradually progressive, along the right side of the neck, extending from the base of the mandible superiorly up to the third tracheal cartilage inferiorly. It was an ovoid shaped, non tender, cystic swelling with well-defined margins, and mobile in all axes with no other skin irregularity over its surface. The swelling was fluctuant with no translucency, no visible or palpable pulsations.

A contrast enhanced MRI of the lesion showed a 7.2 x 8.2 x 2.5 cm lobulated cystic mass with internal septations and regular post contrast peripheral enhancement from the level of carotid bifurcation to the lower pole of right lobe of the thyroid, deep to the sternocleidomastoid muscle. A CT angiography of the neck vessels showed no obvious feeding vessel to the mass, with only anterior displacement of the right internal jugular vein (Fig. 2b, 2c). An FNAC of the lesion showed mature lymphocytes with proteinaceous material in the background, consistent with a lymphatic cyst.

The patient underwent a complete excision of the mass

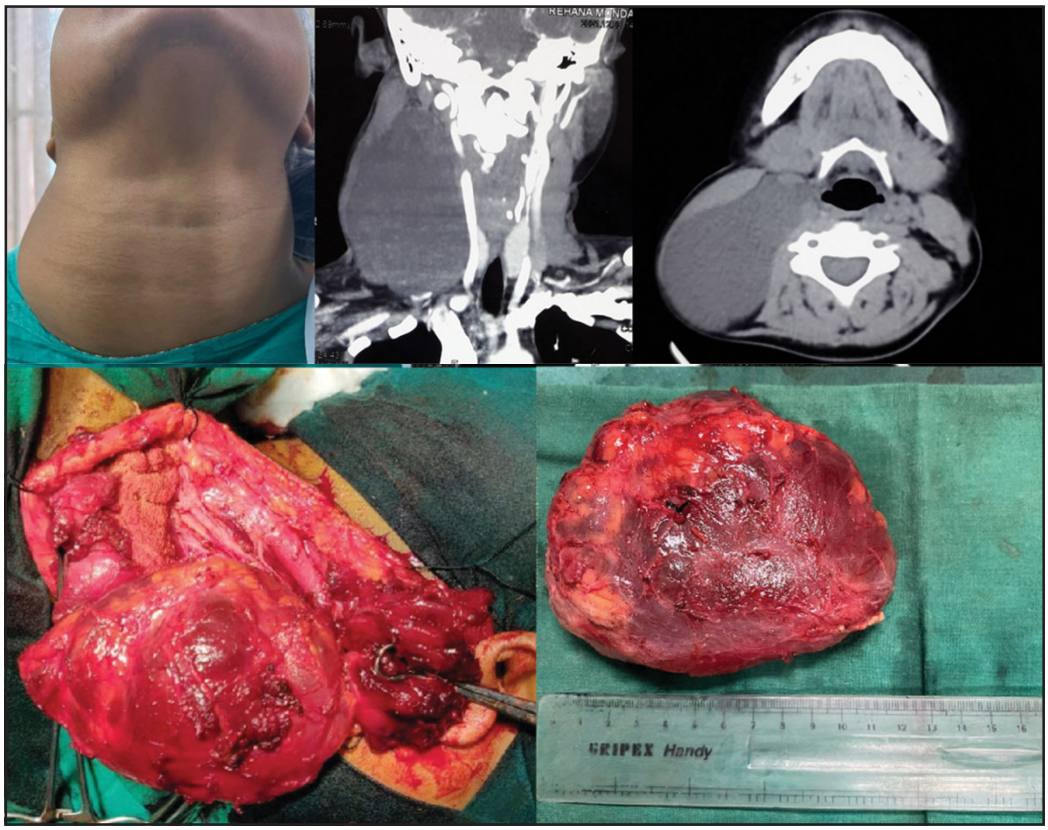

Fig. (left to right, top row) 2a. Clinical Picture of the patient; 2b., 2c. CT angiography of the lesion in coronal and axial view; (bottom row) 2d. Intra operative picture showing the lesionafter dissection; 2e. Post operative specimen.

via a transcervical route (Fig. 2d). Dissection was carried out carefully so as not to puncture the mass, while avoiding the structures it has displaced such as the carotid sheath (Fig. 2e). A post op HPE showed features consistent with cystic hygroma. At 1 year follow up, the patient has not had any recurrence.

Case 3

An 18 year old female presented with a 5 x 4 cm right sided asymptomatic neck lump which was gradually progressive in size,non tender,firm, smooth surfaced, mobile in both axes and non-pulsatile without any skin or muscle fixity (Fig. 3a). There were no engorged vessels or any bruit present.

CECT suggested that a well-defined mildly enhancing hypodense lesion seen in right carotid space suggestive of vagal nerve schwannoma. CT Angiography of Neck Vessels reported that a well-defined soft tissue lesion sized 4.0 x 3.4 x 4.0 cm was noted in right carotid sheath without splaying of internal carotid artery (ICA) and external carotid artery (ECA) (Fig. 3c). MRI of neck reported that a well-defined space occupying lesion is seen in the right carotid space (measuring 45 x 48 mm). The lesion was iso to hypointense compared to the muscle on T1 and heterogenous compared to the muscle on T2 (Fig. 3b). FNAC suggested benign neural neoplasia.

The patient underwent excision of the mass via the traditional transcervical route. The lesion was beneath the carotid artery, displacing the IJV posterolaterally. The carotid was meticulously separated from the mass, whose

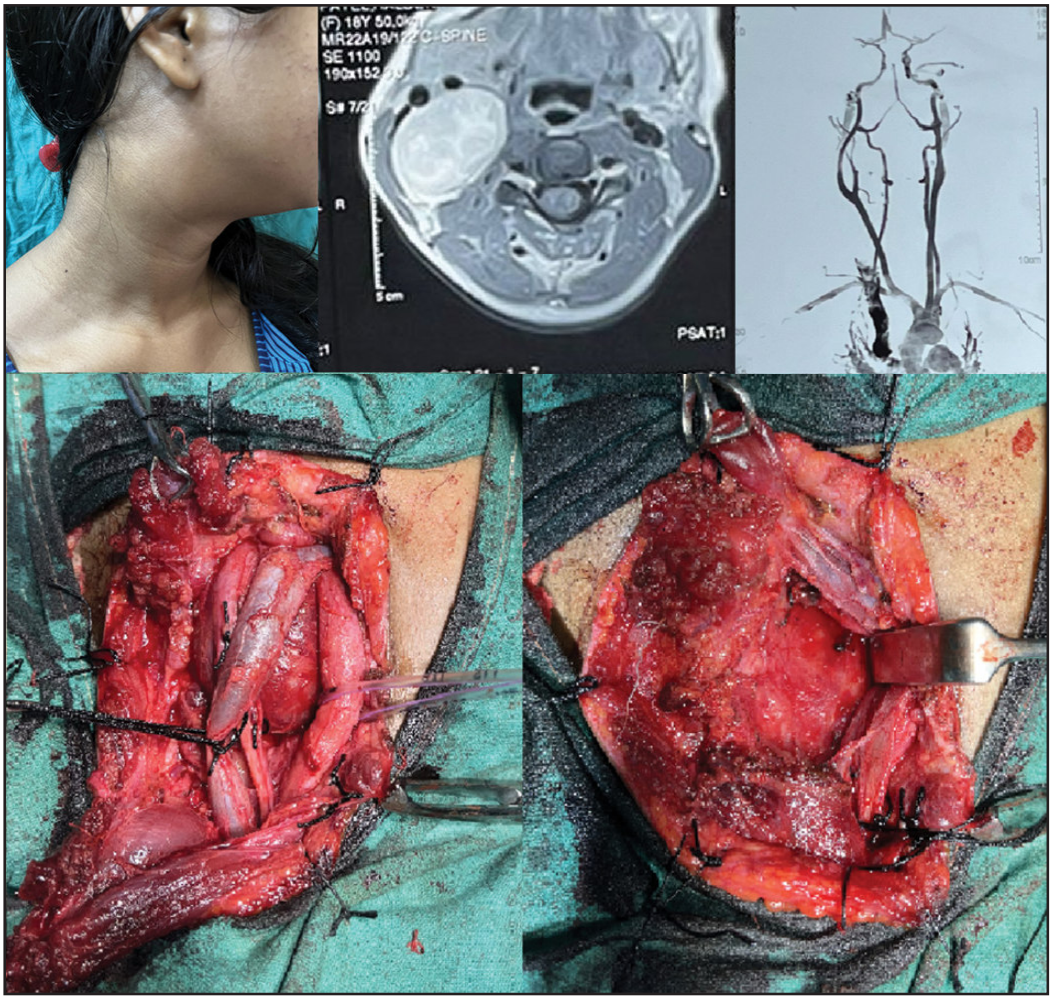

Fig. (left to right, top row) 3a. Clinical Picture of the patient; 3b. Contrast enhanced MRI of lesion at the level of supraglottis in axial section; 3c. CT angiography of the lesion in coronal view; (bottom row) 3d., 3e. Intra operative picture showing the lesion after dissection and after removal.

superior and inferior end appeared to be in continuity with the vagus. An operating microscope was used to carefully dissect out the tumour from the nerve sheath, and the mass was dissected out keeping the trunk of the vagus intact. A post op HPE showing schwannoma (Fig. 3d, 3e). At follow up, after 6months of surgery the patient was well without evidence of recidivism clinically. The patient had mild hoarseness but no pooling of saliva, no facial deviation, no abnormality of tongue movement orshoulder shrugging.

Case 4

A 35 year old male presented with a 9x7 cm swelling at the left angle of mandible since 6 months, slowly progressing in size, smooth surfaced, mobile in both axes, non-pulsatile and without overlying skin fixity (Fig. 4a). There were no visible vessels or bruit.He also had dyspnea for 1 week. Rigid laryngoscopy revealed a mass almost completely obstructing the airway, originating from the parapharyngeal space (Fig. 4b).

CECT scan of neck showed a 92 x 70 x 58 mm well defined heterogeneously enhancing soft tissue mass lesion at the left submandibular region producing significant extrinsic compression at the left lateral aspect of oropharynx with involvement of left aryepiglottic fold and narrowing of pharyngeal airway, extending from the posterior margin of the soft palate to the prevertebral space, with lateral displacement of carotid sheath structures (Fig. 4c). Contrast MRI showed a large well circumscribed lesion with altered signal intensity and heterogenous enhancement in left carotid space with splaying of the ICA and ECA. CT angiography of neck vessels showed no obvious feeding vessels (Fig. 4d). FNAC of the mass was inconclusive.

Although the patient was planned for a transcervical-transmandibular approach for tumour excision, the surgery was successfully completeled via a transcervical approach alone after a tracheostomy (Fig. 4e). An operating microscope was used to carefully delineate the major nerves such as the vagus, ansa cervicalis, greater auricular and hypoglossal to search for nerve of origin, but no major nerve sheaths were involved. Post op HPE showed a diagnosis of schwannoma, which was probably of sympathetic origin, since no major nerves were involved in the tumour. Post operatively the patient did not have any symptoms of any major nerve injury.

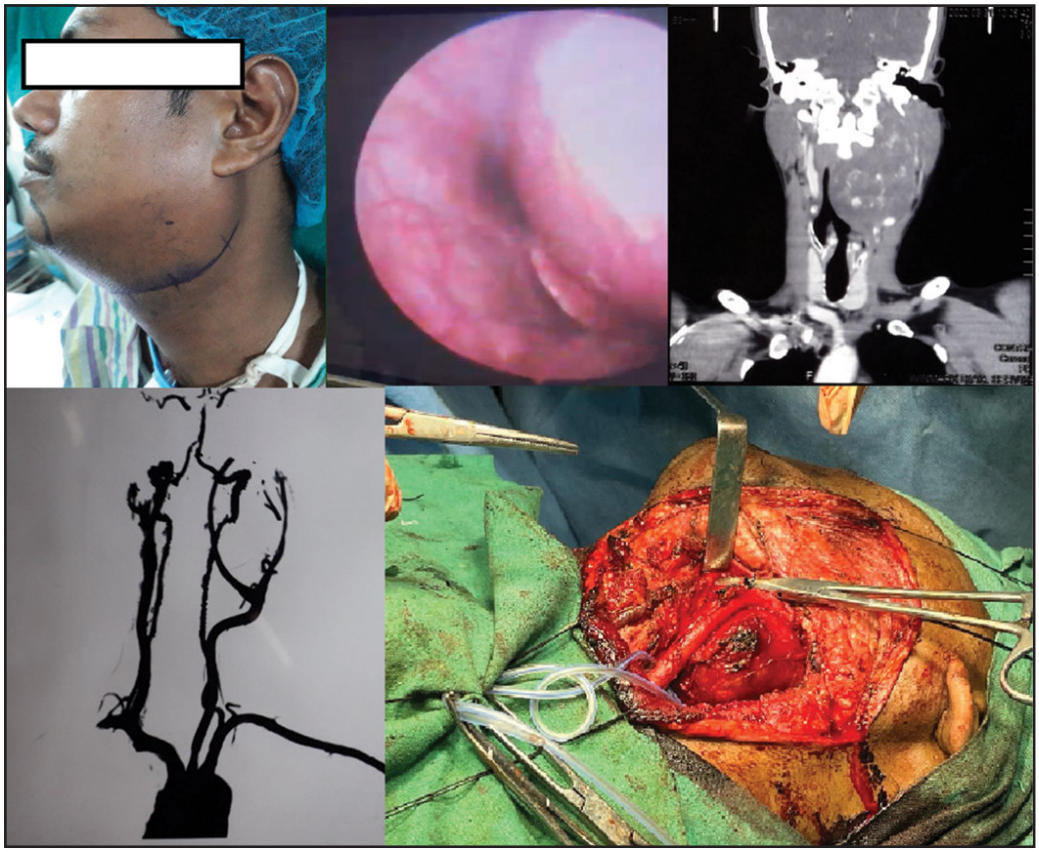

Fig. (left to right, top row) 4a. Clinical Picture of the patient; 4b. Rigid laryngoscopy showing a mass completely occluding the airway in the supraglottic region, with the epiglottis visible anteriorly and a shadow of the right pyriform sinus seen posteriorly; 4c. CE- CT of the lesion in coronal view; (bottom row) 4d. CT angiography of neck vessels showing splaying of ICA and ECA; 4e. Intra operative picture showing the lesion after dissection.

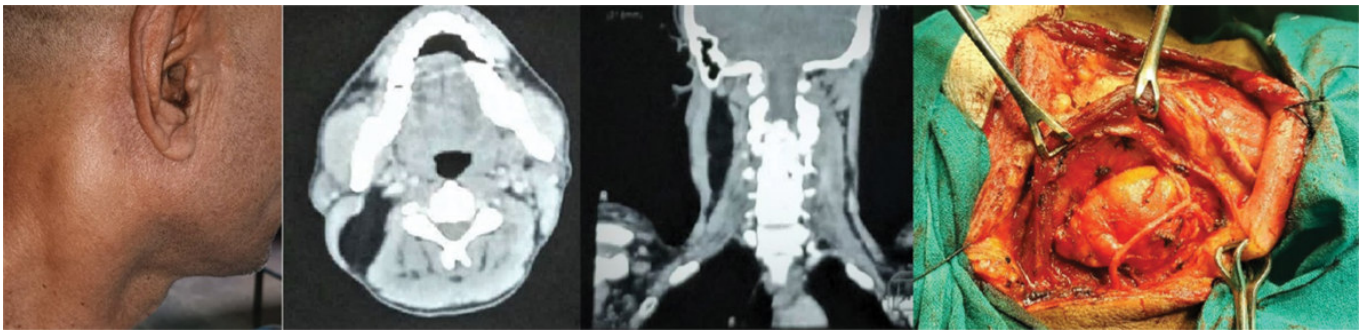

Fig. (left to right) 5a. Clinical Picture of the patient; 5b, 5c. CE- CT of the lesion in axial and coronal view; 5d. Intra operative picture showing the lesion after dissection.

Case 5

A 55 year old male patient presented with an asymptomatic 2 x 2cm ill-defined swelling in the right posterior triangle of neck, which had been slowly progressing in size in the last 3 months (Fig. 5a). The swelling was soft and mobile in all axes with no fixity to the overlying skin. USG was suggestive of a lipoma and FNAC of the swelling showed mature adipocytes in a predominantly lipoid background, again suggestive of a lipoma. Contrast enhanced CT of the neck showed a 75 x 47x 32 mm fat density lesion in the right posterior triangle of neck deep to the sternocleidomastoid (Fig. 5b, 5c). The lesion was excised by the traditional transcervical route (Fig. 5d), and post op HPE showed that it was indeed a lipoma.

Discussion

Lateral neck masses present a challenge in that the differential diagnosis encompasses a wide variety of benign and malignant lesions. One of the main locations may be in the parapharyngeal space. It is a potential neck space in the shape of an inverted pyramid with its base near the skull base, and divided into the pre styloid and post styloid compartment by the aponeurosis of Zuckerkandl and Testut. The prestyloid compartment contains adipose tissue, lymphatics, ectopic salivary gland tissue and small nerves and vessels, while the post styloid compartment contains the internal carotid artery, internal jugular vein, cranial nerves IX to XII, the cervical sympathetic chain, lymph nodes and glomus bodies. The parapharyngeal space is the location of 0.5% of all neck masses.[1]

As is quoted in the 8th edition of Scott-Brown's Otorhinolaryngology Head and Neck Surgery,[1] Riffat et al in a systematic review of 1143 cases of parapharyngeal tumours over 20 years, found that most of these tumours were benign (82%). Salivary gland tumours accounting for the majority (45%) of all tumours, followed by neurogenic (41%), miscellaneous (12%) and metastatic deposits (3%). Miscellaneous included tumours such as carotid artery aneurysms, branchial cleft cysts, haemangiomas, meningiomas, lipoma, cystic hygroma and inflammatory pseudotumours among others.[2]

Frequently, they tend to present as asymptomatic lateral neck or intraoral masses when their size grows beyond 2.5-3 cm, or may be found on routine neck examination. Others may present with a mass effect secondary to the tumour, with symptoms ranging from voice change, nasal obstruction, foreign body sensation, hearing loss due to eustachian tube dysfunction, dysphagia, snoring or even stridor. Tumour invasion of adjacent nerves (VII to XII) may also lead to concurrent symptoms raging from cough, hoarseness, dysphagia or dysarthria.[5]

The imaging modality of choice in such masses is either a CT or an MRI, because of the lack of an adequate sonographic window to perform ultrasonography. Contrast CT is easier done specially in patient whom an

MRI is contraindicated, but is far inferior to a gadolinium enhanced MRI.[6]

The lesion may further be investigated with MR angiography or carotid angiography for vascularity and relation to the neurovascular bundles.[1]

Fine needle aspiration cytology for a pre operative diagnosis is a mainstay of most of these tumours with a generally accepted low rate of false positive and false negative results, but Arnason et al showed that they may tend to show a low specific diagnosis rate (36%) and a high nondiagnostic rate (31%).[7]

This is further supplemented by similar studies by Farrag et al and Oliai et al who had similar non diagnostic rates of 21% and 25% respectively.[8,9] Similarly in our series, 40% (2 out of 5) FNAC reports were inconclusive.

Most (95%) of these tumours are removed by definitive surgical excision.[2]

Surgical resection techniques described in the literature are classified as transoral, transcervical, transparotid-transcervical, transcervical-transmandibular or infratemporal and the correct choice between them depends upon the accurate information on mass size and location, its relationship with the surrounding vessels and nerves and its nature. Moreover, newer techniques such as endoscopic endonasal approaches (EEA) for tumours of the upper parapharyngeal space, and endoscope assisted trans oral excision have started gaining popularity because of the superior cosmetic outcomes. However, they are not without their drawbacks and hence should be used in select cases because of the risk of haemorrhage, cranial nerve damage or spillage of tumour cells. Other future avenues showing promise include resection by Trans-oral robotic surgery (TORS) which provides high local control, an excellent 3D view and less complications if patients are selected appropriately.[10,11,12]

Adjuvant radiotherapy is preferred for cases that are malignant, or in case of recurrent benign lesions. Chemotherapy is limited to specific tumour pathologies like rhabdomyosarcoma or in cases with involved margins, aggressive histology, perineural and lympho-vascular spread.[1]

Radiotherapy is the primary modality in cases with either unresectable lesions or patients not amenable to surgery.[2]

Pleomorphic Adenoma :

Pleomorphic adenoma (PA), also known as benign mixed tumour, is the most common salivary tumour, constituting up to two-thirds of all salivary gland neoplasms.[13]

They are generally discovered during routine physical examination, as an asymptomatic mass, which is glandular in origin in the head and neck region and usually manifests as a mobile, slowly progressive, asymptomatic firm swelling that does not cause ulceration of the overlying mucosa. The majority of these tumours measure 2-6 cm in size when excised. However, large to giant tumours may be seen as a single, irregular nodular mass stretching the overlying skin or mucosa. In our case the excised tumour was 9 x 8 x 6 cm.[14]

Although facial nerve dysfunction at initial presentation is almost always an indication of a malignant lesion, it is rarely observed in patients with locally recurrent parotid pleomorphic adenomas. Approximately 3% to 4% of pleomorphic adenomas may become carcinoma ex-pleomorphic adenomas, an aggressive malignancy that may metastasize and result in death.[15]

Usually with tumours of < 4cm size, a transparotid transcervical approach is used which is especially helpful in identifying and preserving the facial nerve, external and internal carotids, internal jugular vein, cranial nerve IX, X, XI, XII and the sympathetic plexus while for masses > 4 cm in size, a transcervical transmandibular approach is preferred, which not only provides a wider surgical field, but also aids in delivery of the mass.[16]

However we were able to successfully deliver the mass in toto via the transparotid transcervical approach without any breach of the tumour capsule.

Cystic Hygroma:

Lymphangiomas are uncommon benign malformations of the lymphatic system, that are categorised as either superficial or deep based on the depth of the lymphatic vessels involved. The deep variety is either classified as

a cavernous hemangioma or a cystic hygroma. They occur due to obstruction of the lymphatic system during development and can be associated with genetic disorders such as trisomy 13, Noonan disease or Down's syndrome, among others.[17] Although well recognized in paediatric practice, acquired cases of lymphangioma in adults are exceedingly rare, with only about 100-150 cases reported in text and reviews of literature up to as recently as 2022.[18,19] The 7th edition of Cumming's Otolaryngology, Head and Neck surgery[20] also clearly states that most lymphatic malformations present at birth, and only few may appear later in childhood in response to trauma or infection.They occur due to blockage of previously normal lymph channels. Cystic Hygroma are typically well circumscribed, occurring in the neck. They are soft and slow growing. Other differentials that they may be confused with include thymic cysts, bronchogenic cysts, cystic teratoma, which should be kept in mind while approaching such a case. The management is complete surgical excision along with wide local excision of the affected lymphatic channels, failing which there may be recurrences. The recurrence rates have been reported to be as high as 23%. Other modalities include CO2 laser, ND YAG lasers, cryotherapy and sclerotherapy with sodium tetradecylsulfate, doxycycline or ethanol.[17]

Schwannoma:

They comprise of about 27% of all neurogenic tumours of the parapharyngeal space.[2] They are benign tumours of the nerve sheath which can arise from any nerve which have schwann cells except optic and olfactory nerves, as they lack those cells. Head and neck are the most commonly affected regions (25–45%) with the lateral neck being the frequently involved site. MRI appears to be the investigation of choice for the diagnosis and identification of the nerve of origin. Schwannomas are resistant to radiotherapy and the treatment of choice is conservative surgical excision with regular follow up. Nerve preservation should be attempted whenever possible.[21] Many authors though, believe that close adherence of the vagus nerve to the tumour capsule renders nerve function preservation impossible and advocate nerve transection along with immediate reanastomosis or vocal cord medialization. Others have shown results with nerve sparing techniques such as enucleation and extracapsular removal. Nerve sparing dissection is aided with microsurgical dissection using an operating microscope and intraoperative nerve monitoring.[22] Nerve preservation aside, micro dissection has been shown to aid in completion of resection and prevent recurrences.[23] Multiple recurrences after excision are often associated with neurofibromatosis II.[21]

Lipoma:

Although solitary lipomas are not generally of clinical interest, with 13% of lipomas occurring in the head and neck region, rarely they have been known to infiltrate surrounding tissues (infiltrating lipoma). The most common tissue infiltrated by a lipoma is a skeletal muscle, and they can either exist in an intermuscular or an intramuscular form. The lesion most likely arises from intermuscular fascial septae and infiltrates the surrounding muscle. Such infiltrating lipomas are extremely rare (1.9% of lipomas), and when present, arise in the upper and lower extremities. Isolated infiltrating lipomas in the head and neck region are considered extremely rare. The treatment entails either wide excision through neck incisions or liposuction.[3, 24] It may be difficult to define the exact location of origin, especially with an intermuscular lipoma which secondary infiltrated the adjacent muscle. As many clinicians are unaware of the significance of a lipoma arising within the muscle, knowledge of this pathology can help the physician to provide counselling and appropriate treatment for the patient.[25]

Conclusion

Tumours of the neck and parapharyngeal region have a wide variety as this region contains almost every kind of tissue. Diagnosis of any lesion should be done with caution, using the appropriate history, examination and investigative tools available. Not only the common presentation, but also outliers and uncommon presentations of common tumours should be kept in mind.

This case series provides a brief overview of the type of tumours that can present to an ENT practitioner and the relevant investigations to be done. Most of the lesion require excision via neck incision, while a few such as lymphomas need chemotherapy. Rare presentations such as the ones presented here which include giant pleomorphic adenoma, adult onset non congenital cystic hygromas, and intramuscular lipomas should be kept in mind while reaching a diagnosis and planning the treatment of the patients.

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