Case Report

A rare case of a solitary laryngeal xanthoma and its clinical implications in a developing country like India

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INTRODUCTION

Xanthomas are localized infiltrates of lipid-containing histiocytic foam cells that usually are found within the dermis or tendons. Xanthomas frequently arise in the presence of a disturbance of lipoprotein metabolism. They arise due to reactive proliferation of fat laden histiocytes and are not considered as true tumours. There are only 4 reported cases so far of isolated laryngeal xanthomas in English literature so far.

CASE REPORT

We report a rare case of polypoidal xanthoma of vocal cord in a 42 year old male, non-smoker, known case of type 2 diabetes mellitus and hypertension on medication, with history of voice abuse, who presented with complaints of hoarseness and change in voice for 1 year. Direct laryngoscopic examination showed a smooth textured yellowish mass arising from the anterior aspect of the left vocal cord (Figure 1).

Figure 1: Smooth textured yellowish mass arising from the anterior aspect of the left vocal cord.
No abnormal lesions were found in other parts of the larynx or on the skin (Figure 2).

![Figure 2: No other abnormal lesions found on the skin or any other part of the body.](image)

Routine blood investigations showed deranged blood lipid values. Total cholesterol, low-density lipoproteins (LDL) and triglyceride values were grossly increased (497 mg/dl, 164 mg/dl and 1441 mg/dl respectively). High-density lipoproteins (HDL) cholesterol values were decreased at 38 mg/dl.

Following clinical diagnosis of left vocal cord polyp, microlaryngeal excision surgery was done under general anaesthesia (Figure 3) using a coblator and the excised tissue was sent for histopathological examination.

![Figure 3: Microlaryngeal excision under general anaesthesia.](image)

Histopathology showed fragments of stratified squamous epithelium with sub-epithelial displaying sheets of foamy cells. Also seen were mild lymphocytic infiltrates, areas of haemorrhage and necrosis (Figure 4). Stains for any organisms were negative. Immunohistochemistry of the lesion was positive for CD68, and negative for S-100 protein, and cytokeratin.

![Figure 4: Stratified squamous epithelium with subepithelium showing a well-defined lesion displaying sheets of foamy cells.](image)

Patient was called for follow up after one week to evaluate vocal cord status and to start speech therapy, after which he was started on appropriate statins for dyslipidemia.

Follow up at the end of 6 months showed no recurrence (Figure 5). No additional lesions were seen on the skin and the patient had normal speech quality, with alleviation in all pre-operative symptoms.

![Figure 5: No recurrence on follow up.](image)

DISCUSSION

In the larynx, the most common disorder exhibiting apolypoid appearance is the laryngeal polyp. Rarely, polypoid hamartomas in the larynx have been reported.2 Vocal cord polyps tend to show subepithelial oedema along with a combination of varying degrees of organization (fibrosis) and/or degeneration (basophil and hyaline degeneration), with some polyps showing underlying sub-epithelial haemorrhages.6

Hamartomas on the other hand show a disorganized architectural pattern with mesenchymal derivatives alone, or with superadded epithelial elements. Hamartomas of the larynx are mainly composed of cartilage and fibromuscular tissue.7

Neither of these pathologies are known for containing foamy histiocytes. Of the laryngeal masses containing foamy histiocytes, the various differentials that can be
considered are: verruciform xanthoma of the larynx, xanthoma disseminatum involving the larynx, fibrous histiocytoma, and laryngeal xanthoma.2

Verruciform xanthoma is a relatively uncommon hyperplastic condition of the epithelium affecting primarily the oral mucosa, anogenital mucosa and skin. Microscopically, they present as hyperplastic, parakeratinized squamous epithelium lining the papillary projections with elongated rete ridges of relatively uniform depth and the parakeratin filling the clefts between the epithelium projections. The hallmark of this lesion is the presence of numerous large macrophages with foamy cytoplasm (xanthoma cells) typically confined to the connective tissue papillae, which extend high into epithelium.8 The absence of any papillary projections or rete ridges ruled out the diagnosis of verruciform xanthoma in our case.

Xanthoma disseminatum on the other hand, primarily affects the skin, causing multiple red-yellow papules and nodules. Mucous lesions, which are present in 40-60% of the cases, appear along with the skin lesions.9 The absence of any cutaneous xanthomatous lesions in our patient on either presentation or on follow up ruled out this diagnosis also.

Finally, the absence of any large pleomorphic spindle cells, storiform growth pattern or multinucleated giant cells precluded the diagnosis of fibrous histiocytoma.10

Following the histopathological analysis, the diagnosis of isolated laryngeal xanthoma was arrived at.

Clinical implications

Prevalence of dyslipidemia in India trends from 10 to 15% in rural to 25-30% in urban populations. But a major shortcoming of such studies in India is the lack of large studies to detect patterns of dyslipidemia, as a result of which many of the patients may have undetected deranged lipid profiles.

Abnormalities of various cholesterol lipoprotein lipids are important risk factors for coronary heart disease (CHD). There is a strong pathophysiological association of raised cholesterol with initiation and progression of coronary atherosclerosis.

Moreover, a country like India is undergoing a rapid epidemiological transition with increasing population, economic prosperity, urbanization and aging, along with associated risk factor transition. As a result of this, Indian populations are more prone to cardiovascular events and hypercholesterolemia than before. These derangements are also associated with increase in adverse lifestyles such as greater smoking and tobacco use, change in nutritional habits with greater intake of unhealthy diets and increasing sedentary lifestyle. All these factors have contributed to the rising burden of non-communicable diseases, especially CHD. Even in rural areas of India non-communicable and chronic diseases have become the leading causes for death.11

Hence in such cases, when a patient presents with an isolated xanthoma of the larynx, apart from the excision of the lesion itself, it is imperative to look for underlying derangements in the lipid profile and start the patient on the statins, to prevent any future risk of cardiac events.

This is especially true in a country like India, where a rapidly evolving population has predisposed the people to a higher risk of non-communicable diseases like dyslipidemia which, apart from causing xanthomas that may obstruct the airway, also poses a serious risk of coronary heart disease.

CONCLUSION

All patients presenting with hoarseness of voice should undergo laryngoscopic evaluation. In case any vocal cord polypoidal lesion with yellowish tinge is visualised, vocal cord xanthoma should be considered as a potential diagnosis. The patient should be evaluated for any other lesions in the body while keeping in mind that the absence of any such lesion does not exclude the diagnosis of xanthoma. After microlaryngeal excision of the lesion, a thorough histopathological analysis should be done, followed by immunohistochemistry if required. If a diagnosis of laryngeal xanthoma is arrived at, lipid profile and diabetic or hypertensive status should be checked for any underlying pathology. Ruling out any underlying dyslipidemias is especially important in a developing country like India, where the general population is rapidly undergoing a change from rural type to urban type, leading to an increase in undiagnosed non communicable diseases. Not only will this help to estimate the burden of the disease, but appropriate medication, if started for the patient on time, will help to prevent morbidities such as cardiac events in the future.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

REFERENCES

1. Parker F. Xanthomas and hyperlipidemias. J Am Acad Dermatol. 1985;13(1):1-30.
2. Matsumoto T, Nobukawa B, Kobayashi K, Watanabe M, Hosokawa A, Tomaru K, Ichikawa G. Solitary polypoid xanthoma in the larynx. Histopathology. 1999;34(5):475-7.
3. Perrone G, Zagami M, Casale M, Salvinelli F, Morini S, Rabitti C. Immunohistochemistry and differential diagnosis of a solitary flat laryngeal xanthoma: a case report. In vivo. 2007;21(1):119-21.
4. Jain AK, Jain S, Katarkar A, Alam N, Lakum N, Shah P, Modh D. A Rare Case of Sessile Polypoidal Xanthoma of Vocal Cord. 2012;2(2):66-8.
5. Vera-Sempere F, Collado-Martín D, Vera-Sirera B. Solitary Polypoid Laryngeal Xanthoma. Case Rep Otolaryngol. 2013;967536.
6. Kambič V, Radšel Z, Žargi M, Ačko M. Vocal cord polyps: incidence, histology and pathogenesis. J Laryngol Otol. 1981;95(6):609-18.
7. Rinaldo A, Mannarà GM, Fisher C, Ferlito A. Hamartoma of the larynx: a critical review of the literature. Annals of Otology, Rhinol Laryngol. 1998;107(3):264-7.
8. Hegde U, Dodawad VG, Sreeshyla HS, Patil R. Verruciform xanthoma: A view on the concepts of its etiopathogenesis. Journal of oral and maxillofacial pathology: JOMFP. 2013;17(3):392.
9. Park M, Boone B, Devos S. Xanthoma disseminatum: case report and mini-review of the literature. Acta dermatologica et venerologica Croatica. 2014;22(2):150-5.
10. Van Laer C, Hamans E, Neeten I, Van Marck E. Benign fibrous histiocytoma of the larynx: presentation of. J Laryngol Otol. 1996;110:474-7.
11. Gupta R, Rao RS, Misra A, Sharma SK. Recent trends in epidemiology of dyslipidemias in India. Ind Heart J. 2017;69(3):382-92.

Cite this article as: Basavarajaiah BM, De KS, Rao RBS, Rudrappa BA. A rare case of a solitary laryngeal xanthoma and its clinical implications in a developing country like India. Int J Otorhinolaryngol Head Neck Surg 2021;7:385-8.