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Case report: Trichilemmal cyst involving submandibular gland

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Abstract
Trichilemmal cysts are rare adnexal lesions, commonly occurring on scalp. We present a case of 13 year old female patient presenting with left sided neck swelling since 3months, swelling was 20×20 mm in size, soft to firm freely mobile, non-tender in the left submandibular region. USG showed an anechoeic cystic lesion with hyperechoeic content in the left submandibular gland. Surgical excision was done; histopathology confirmed it to be trichilemmal cyst.

Keywords: submandibular gland cysts, trichilemmal cysts, adnexal tumors

Introduction
Trichilemmal cyst are benign adnexal tumors usually occurring in hair bearing areas such as scalp, groin, thigh especially in elderly women. They arise from the outer root sheath of the hair follicle. The cyst contains keratin and outlined by stratified squamous epithelium. Trichilemmal cysts may be inherited as an autosomal dominant trait, there can be familial predisposition with patients presenting at younger age and with multiple lesions. Occurrence of trichilemmal cyst at submandibular region has been rarely reported.

Case Report
A 13 year old female patient presented to ENT OPD with complains of swelling over left submandibular region since 3 months. The swelling was insidious in onset, gradually progressive in size, on examination it was 20×20 mm in size, soft to firm in consistency, mobile, nontender involving the left submandibular region. USG showed thick wall small anechoic cyst containing internal echoes and bright echogenic component producing acoustic shadow noted in submandibular region measures 14×9×11mm.
Surgical excision of the swelling was done under general anaesthesia and the specimen was sent for histopathological examination. Report was suggestive of a circumscribed lesion, cyst composed of squamous epithelium showing abrupt keratinisation and abundant keratin, a diagnosis of trichilemmal cyst was made.

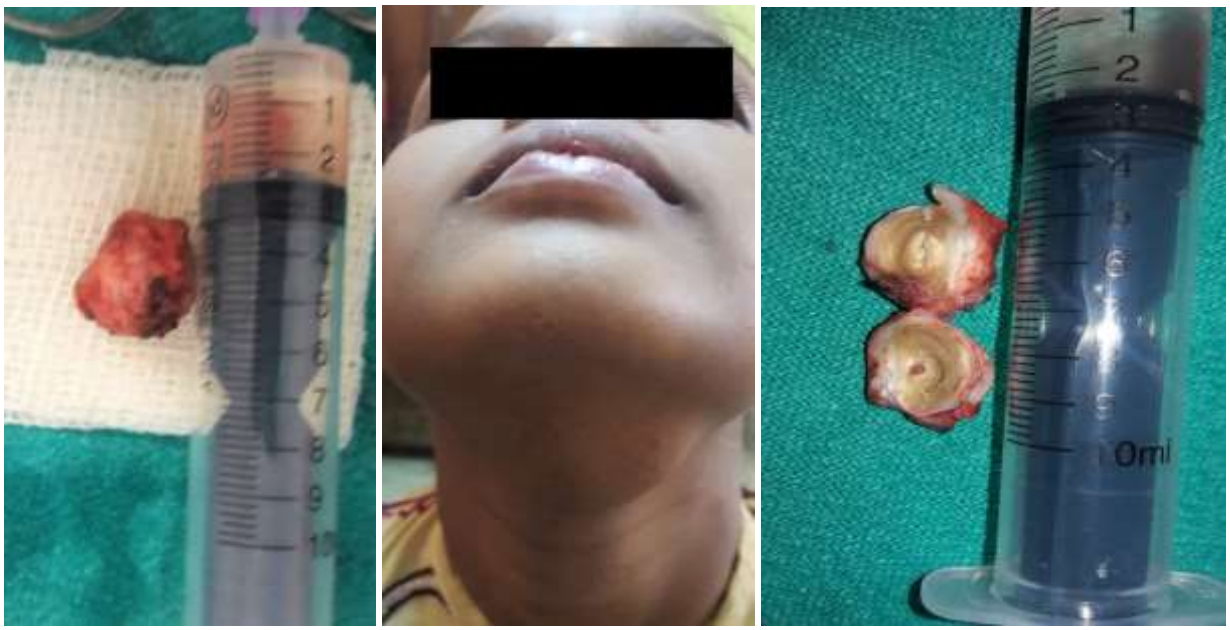

Fig 1

Discussion

Trichilemmal cysts are intradermal cysts that arise from the epithelium located between the sebaceous gland and arrector pili muscle. They are lined by stratified squamous epithelium without granular cell layer, similar to outer sheath of hair follicle. These cysts are composed of keratin and its breakdown products. Trauma is considered to be the causal agent, but definitive etiology is yet unknown.

Perez *et al*. [1] reported intraoral presentation of trichilemmal cyst, El-Bahy and Ishak [2] presented a trichilemmal cyst occurring in temporal and infratemporal fossae. They performed a frontotemporal craniotomy and orbitozygomatic osteotomy and confirmed that the histology was that of a trichilemmal cyst.

Yoo DJ *et al*. [3] reported occurrence of trichilemmal cyst in submandibular gland. Surgical excision of cyst along with submandibular gland was done. Our case was similar to their as the cyst was present in submandibular gland.

In terms of oncological transformation Saida *et al*. [4] described three stages of trichilemmal tumor formation:

Adenomatous stage (trichilemmal cyst), Epitheliomatous stage (proliferating trichilemmal cyst) and Carcinomatous stage (malignant proliferating trichilemmal tumor).

Trichilemmal cysts have malignant potential and surgical excision is usually recommended.

The differential diagnosis of cystic swellings in the submandibular triangle include: branchial cleft cysts, cystic hygroma, submandibular gland mucocele. Second branchial cleft cysts are commonly seen in late childhood or early adulthood, it presents with painful tender swelling in submandilar region deep to and along the anterior border of sternocleidomastoid muscle. They are soft, doughy, variable sized cystic swellings, their remanent tract may course between carotid branches anterior to glossopharyngeal and hypoglossal nerves, and enter into oropharynx. On CT imaging an uncomplicated branchial cleft cyst has a thin, smooth rim and low-density mucoid contents. Treatment is surgical excision of the cyst along with its tract.

Trichilemmal cysts are surgically removed, limited surgical resection is sufficient for cure. Their involvement of submandibular gland mey or may not require gland removal. Although it is a rare condition but long term follow up for the results are needed.

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