Linear trichilemmomas: An unusual clinical presentation

Sir,

Trichilemmomas are benign cutaneous neoplasms characterized by cells showing differentiation toward the outer root sheath of hair follicle. We describe a rare clinical variant of this condition.

A 41-year-old female patient presented with multiple, asymptomatic lesions over the posterior aspect of the left pinna and retroauricular area for six to eight months. On examination, there were multiple mildly erythematous to hyperpigmented papular lesions with a verucous surface arranged in a linear pattern on the above-mentioned sites. There was no oozing and the lesions were non-tender. The other ear was spared. Systemic examination was unremarkable. Provisional clinical differential diagnoses included viral warts, epidermal nevus and squamous cell carcinoma [Figure 1].

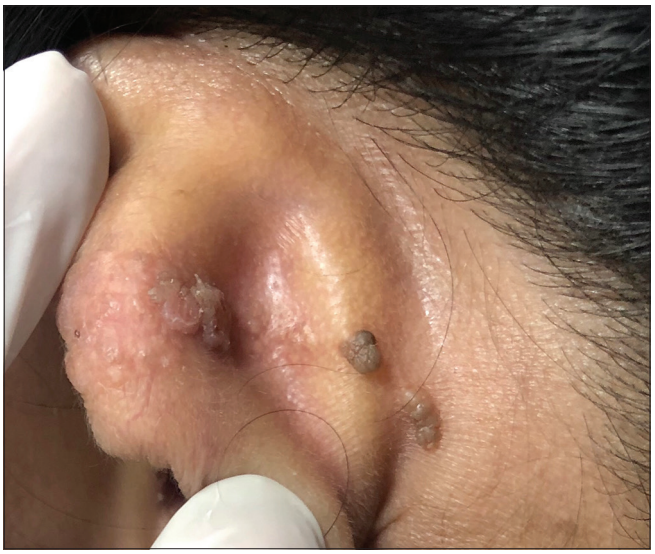

Figure 1: Skin colored to hyperpigmented verrucous surfaced papules in linear pattern

A shave biopsy was taken which showed irregular circumscribed lobular proliferations connected with overlying epidermis [Figure 2]. These proliferations were composed of squamoid and basaloid cells. A thickened and hyalinized basement membrane surrounded the proliferation in most parts. Squamoid cells showed abundant pale and vacuolated cytoplasm whereas the basaloid cells at the periphery showed a palisaded appearance resting on the thickened basement membrane reminiscent of outer root sheath of hair follicle [Figures 3 and 4]. Surrounding stroma showed a sparse lymphoplasmacytic infiltrate. The surface epidermis showed mild papillomatosis and hyperkeratosis. Based on these features, a final diagnosis of trichilemmoma was made. Treatment could not be initiated, as the patient was lost to follow-up.

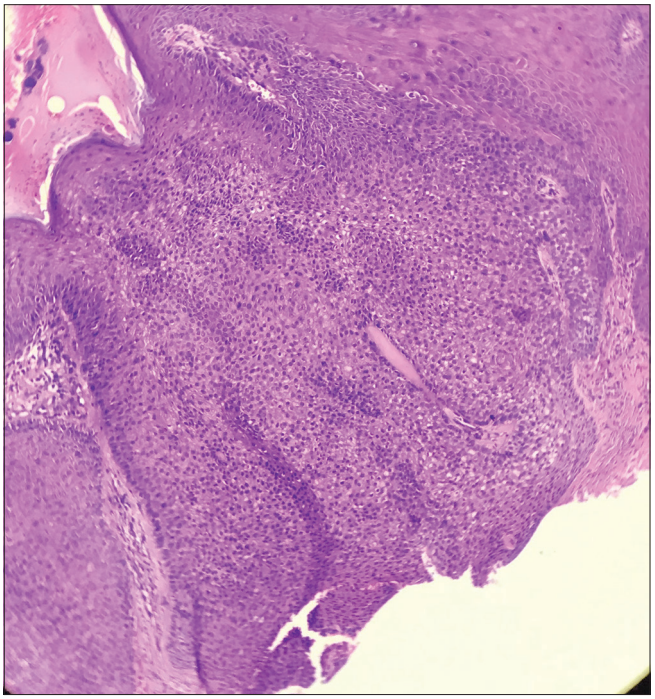

Figure 2: Circumscribed lobular neoplasm connected to overlying epidermis (H and E ×40)

Trichilemmoma was first described by Headington and French in 1962, as a neoplasm showing differentiation toward the outer root sheath of a hair follicle.[1] Ackerman and Wade in 1977 and 1980[2] contented that trichilemmomas are just aged viral warts, however, this was not supported by many dermatopathologists.

Trichilemmomas are either solitary or multiple. Linear pattern in blaschkoid distribution has also been reported previously on the nose[3] and ankle.[4] Lesions are usually skin colored, verrucous surfaced or smooth, small papular lesions

How to cite this article: Desai CA, Shah N. Linear trichilemmomas: An unusual clinical presentation 2022;88:104-6.

Received: April, 2020 **Accepted:** April, 2021 **EPub Ahead of Print:** September, 2021 **Published:** December 2021

DOI: 10.25259/IJDVL_552_20 **PMID:** 34623051

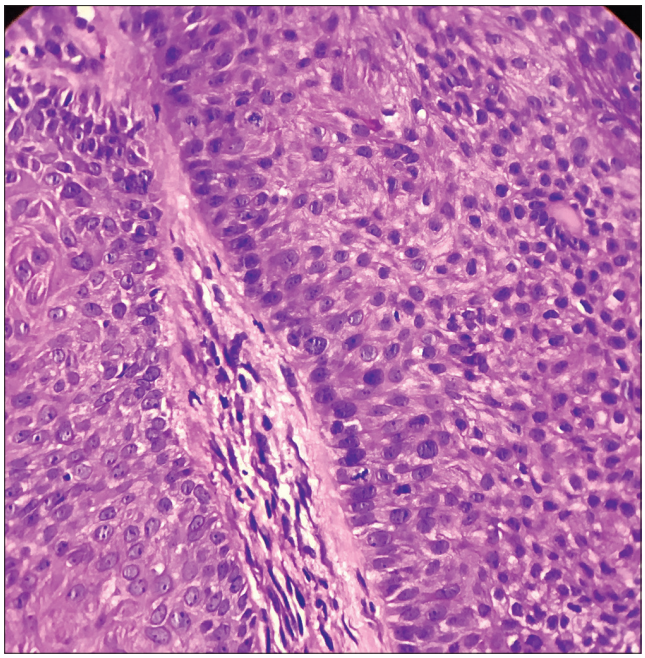

Figure 3: Basaloid cells arranged in a palisade and resting on thickened basement membrane (H and E ×400)

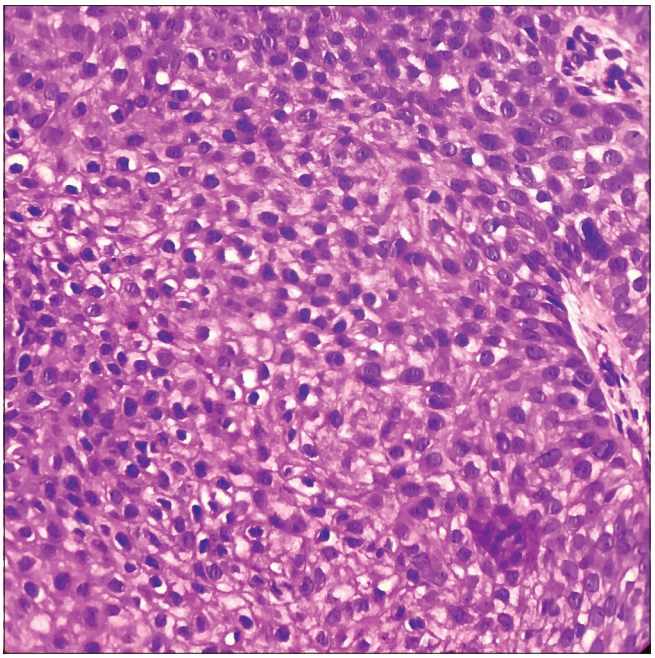

Figure 4: Clear or pale vacuolated cells resembling the outer root sheath of hair follicle (H and E ×400)

which sometimes coalesce to form small plaques. Usual location is on the head-and-neck region of older individuals. Solitary lesions may arise in naevus sebaceous either singly or it may be associated with other neoplasms. Multiple lesions are a cutaneous marker for Cowden disease, which is a multisystem disorder characterized by occurrence of multiple hamartomas and neoplasia with a high incidence of thyroid and breast malignancies. It is an autosomal dominant condition having a female preponderance and with a mutation in the PTEN gene on chromosome 10q23.31. Here, multiple facial trichilemmomas are associated with acral keratoses, palmar pits and mucocutaneous papillomatous papules. Furthermore, vitiligo, lipomas, hemangiomas, neurofibromas, schwannomas and xanthomas may be variably present. Visceral manifestations such as carcinomas of thyroid, breast and endometrium, macrocephaly, dysplastic cerebellar gangliocytomas and renal cell carcinomas have been reported.[4,5]

Classical histopathology of trichilemmomas depicts well-circumscribed lobular neoplasm connected to the epidermal or follicular epithelium at multiple points and comprising of pale to clear vacuolated cells centrally with peripheral cuboidal cells arranged in palisade and resting on a thick glassy basement membrane. Clear or pale cells contain glycogen. Squamous eddies may be seen. Sometimes, partly this tumor may have pseudo-infiltrative pattern with epithelial cords or islands interspersed with a thick, fibrous stroma. Such a variation is called desmoplastic trichilemmoma.

Histological mimickers of this tumor include trichilemmal carcinoma, clear cell variants of basal cell carcinoma and squamous cell carcinoma. However, absence of atypia, mitoses, necrosis, deep infiltrative margins and other features of malignancy sufficiently rules these out. Peripheral palisade of cells resting on a thickened basement membrane and absence of ductal differentiation rules out the possibility of hidroacanthoma simplex. Inverted follicular keratosis can also be excluded as it has more number of squamous eddies, a resemblance to irritated seborrheic keratosis, some spongiosis and absence of typical feature of palisade of cuboidal cells resting on a thick basement membrane. Trichilemmomas are CD34 positive which can also be used when in doubt.[5] Treatment is usually by surgical excision or carbon dioxide laser ablation.

In our case, typical histopathology excluded all other clinical and histological differentials. We report this case for its uncommon linear pattern of presentation following a blaschkoid distribution.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

Chirag A. Desai, Neha Shah[1]

Divya Sparsh Skin & Hair Clinic, [1]Skin Vogue Clinic, Mumbai, Maharashtra, India

Corresponding author:
Dr. Chirag Desai,
Divya Sparsh Skin & Hair Clinic, Dadar (E), Mumbai, Maharashtra, India.
83.chirag@gmail.com

References

1. Headington JT, French AJ. Primary neoplasms of the hair follicle. Histogenesis and classification. Arch Dermatol 1962;86:430-41.
2. Ackerman AB, Wade TR. Tricholemmoma. Am J Dermatopathol 1980;2:207-24.
3. Jha AK, Prasad S, Sinha R. Linear trichilemmoma following a blaschkoid pattern: A clinical dilemma. J Eur Acad Dermatol Venereol 2016;30:299-301.
4. Gonzalez JL, Plotnikova N, Seymour A, Mannion CM, Danialan R, Rosmarin D. Linear trichilemmomas on the ankle of a 28-year-old female. J Cutan Pathol 2017;44:93-7.
5. Patterson JW. Tumours of cutaneous appendages. In: Patterson JW, editor. Weedon's Skin Pathology. 4th ed. Edinburgh: Churchill Livingstone/Elsevier; 2016. p. 908-10.

Lepromatous leprosy with laryngeal involvement

Sir,

Leprosy,caused by *Mycobacterium leprae,* classically affects the skin and peripheral nerves, while involvement of the testes, eyes, lymph node, liver, spleen, bone, bone marrow, muscle and larynx is uncommon. Laryngeal involvement presents with cough, hoarseness of voice, dyspnea and rarely, life-threatening airway obstruction.[1] Implementation of multidrug therapy has further reduced the occurrence of airway obstruction due to laryngeal leprosy, except a few sporadic cases.[2] In the Philippines, newly diagnosed leprosy cases reached 1908 in 2017, almost 90% being multibacillary. Among them 1.9% had grade 2 disabilities at presentation, thus suggesting a relatively high rate of delayed detection.[3]

A 48-year-old Filipino male with longstanding lepromatous skin lesions presented with progressive hoarseness and dysphonia, subsequently followed by difficulty in breathing. The patient had been delaying treatment due to economic constraints and finally approached us with worsening symptoms. Cutaneous examination demonstrated leonine facies, madarosis, saddle nose deformity, nodules and ulcerations on bilateral ears and deformities affecting his feet [Figure 1]. Monofilament test revealed impaired sensation over all digits, although no peripheral nerve was enlarged. Motor test, laboratory tests and chest X-ray were unremarkable. Skin biopsy and Fite-Faraco stain confirmed lepromatous leprosy [Figure 2].

Indirect laryngoscopy demonstrated a fungating epiglottic mass, enlarged arytenoids and markedly narrowed glottic opening due to thickened vocal cords [Figures 3a and 3b]. We observed a friable fungating mass extending from the arytenoids to the true vocal folds on direct laryngoscopy. [Figure 3c]. A computerized tomography scan of the neck demonstrated narrowed glottic opening due to thickened epiglottis, aryepiglottic folds and false vocal cords [Figure 3d]. Histopathology of the arytenoid mass with Fite-Faraco stain showed numerous acid fast bacilli [Figure 3e], confirming the diagnosis of lepromatous leprosy associated laryngeal involvement. Emergency tracheostomy was performed by the otorhinolaryngology surgeons. Post-procedure, we started multidrug therapy for multibacillary leprosy. While on his 2nd blister pack, the patient underwent fiberoptic endoscopic evaluation to detect lack of vocal cord mobility and impaired swallowing.

After his completion of 11th blister pack, we noticed the resolution of ulcers on both ears, without any new skin lesions or sensory or motor deficits. Re-evaluation by the otorhinolaryngology department demonstrated persistent thickening of his epiglottis and vocal folds, thus mandating maintenance tracheostomy. After six months, the otorhinolaryngology department shall reassess the patient to determine feasibility of decannulation.

In all reported cases of leprosy with laryngeal involvement, lepromatous leprosy with widespread skin and cutaneous nerve involvement preceded their laryngeal symptoms. Laryngeal lesions develop insidiously, with an asymptomatic phase for years. Symptoms include hoarseness, dry cough, sore throat, difficulty in breathing, dysphagia, dysphonia, aphonia, odynophagia, raspy breathing, stridor and pain when speaking.[1,4] Laryngeal involvement is confirmed by histopathologic demonstration of acid-fast bacilli.[1]

Usually, laryngeal leprosy starts from the epiglottis.[4,5] This occurs as inspired air flows across the posterior choanae

How to cite this article: de Guia CE, Tablizo AV. Lepromatous leprosy with laryngeal involvement. Indian J Dermatol Venereol Leprol 2022;88:106-9.

Received: April, 2021 **Accepted:** July, 2021 **EPub Ahead of Print:** October, 2021 **Published:** December 2021

DOI: 10.25259/IJDVL_381_2021 **PMID:** 34672475