CASE REPORT

Cutaneous Horns: enigma - Remembering Lady Dimanche

Onkar Singh[1], Vipin V Nair[1], Priya Ranjan[1], S. Gaba[2], Kamlesh K Singh[1]
[1]Armed Forces Medical College, Pune, India
[2]Post Graduate Institute of Medical Education & Research, Chandigarh, India.

Keywords: Cutaneous keratin horn; Cornu cutaneum

Introduction

Cutaneous horns are a rare clinical entity, essentially limited to textbooks in today's surgical practice. Image of Lady Dimanche in Bailey and Love textbook of Surgery with a large cutaneous horn over the forehead is an example of such a rare occurrence (1). We hereby report the case of a plantar horn in a retired Indian soldier.

Case Report

This 56 year old male presented to the department of plastic and reconstructive surgery with a history of growth in the medial instep of the right foot for the last 06 years. It was insidious in onset and gradually progressive, painless and did not offer any sort of hindrance to his ambulation. There was no history of systemic disease. General examination was normal with vital parameters well preserved within normal limits.

Local examination revealed 21 mm long, solitary, hard, non-tender curved horn-like lesion at the junction of the hind and midfoot medially extending into the instep region in non-weight-bearing part of the sole. The diameter of the base was 10mm without inflammation or discharge from the lesion. There was no loco-regional lymphadenopathy. He was diagnosed to have a plantar cutaneous horn and was planned for surgical excision. Haematological and biochemical parameters were within normal limits. Per op fluoroscopic image revealed it to be radio translucent horn. It was excised by an elliptical incision and was primarily closed with interrupted sutures in a linear suture closure. Post-op period was uneventful. Histopathological examination revealed skin epidermis with hyperkeratosis, hypergranulosis, mounds of parakeratosis with extensive 'verruciform orthokeratosis'. Dermis shows sparse mononuclear inflammatory infiltrate (Figure 1).

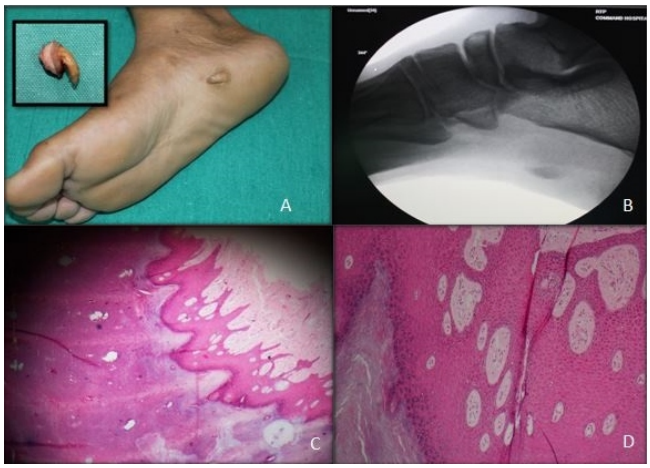

Figure 1. Cornua cutaneum **A**, Lesion before excision and Inset showing Gross photograph after excision **B**, X-ray lateral view showing dense shadow from keratin horn **C**, Low-power view showing skin epidermis with hyper-keratosis, hypergranulosis and extensive verruciform orthokeratosis with dermis showing sparse mononuclear infiltrate. **D**, High-power view demonstrating extensive verruciform orthokeratosis.

Discussion

Image of Dimanche (Madame Dimanche, called Widow Sunday), a French woman living in Paris in the early 19th century, grew a 24.9 cm (9.8") horn in the region of her forehead in six years from the age of 76 before it was successfully removed by French surgeon Br. Joseph Souberbeille (1754–1846). A wax model of her head is on display at the Mütter Museum (The College of Physicians of Philadelphia, US) with long sebaceous horn arising from her forehead and little anecdotes about her and her job, in the first few pages of Bailey and Love textbook of surgery, has fascinated all medical practitioners.

The first case of a cutaneous horn was documented much before Madame Dimanche as early as in the year 1588, wherein pamphlets of Mrs Margaret Gryffith, an elderly Welsh lady, were advertised and she used to be put up as an exhibit for monetary purposes by a businessman. Academic milieu on cutaneous horns in human beings was first given by the Surgeon Everard Home in London in 1791.

Correspondence: Vipin V Nair
E-mail: vipinvenugopalnair@gmail.com
Received: 27-11-2019 Accepted: 28-12-2019
https://orcid.org/0000-0001-6903-6368
DOI: http://doi.org/10.4038/sljs.v37i4.8639

Bondeson gave an overview and historical perspectives regarding cutaneous horns. He described superstitious beliefs prevalent in European population as regards cutaneous horns and how some enterprising individuals made money exhibiting patients with such ailments (3).

Cutaneous horns also are known as 'Cornu cutaneum' in Latin have been enigmatic rare entities, with an incidence of 0.3-1.3% (4). Most of them are seen in fair-skinned individuals because of their increased prevalence and the likelihood of occurrence with sun exposure and actinic keratoses though they are not uncommon in dark-skinned, as sporadic cases have been reported from East Africa, Sudan, Arab and India. These horns are usually of small size but at times they can attain large size. J W Gould described its association with verruca Vulgaris (5). Cutaneous Horns can occur in any part of the body but more than 30% are seen in the head and neck region. These lesions were found both on sun-exposed areas like forehead, hand, eyelids, scalp and trunk. They are also reported on non-sun exposed areas like the penis, lacrimal sac, nasal vestibule and sole. (6).

Cutaneous horns are well-circumscribed, conical projections formed of densely laid adherent keratin. They are hyperkeratotic, owing to the hyperproliferation of basal keratinocytes with compactly layered keratin mimicking a horn. Risk factors for progression to malignancy are male sex, advanced age, sun-exposed surface, and more height to base ratio along with pain and perilesional skin changes.

The association of cutaneous horn with malignant or premalignant potential is found to be around 20-38.9% with squamous cell carcinoma being the most common histopathological variant owing to their increased association with keratin formation (4, 7). Pathogenesis is unclear but they generally arise in the backdrop of actinic keratoses which is a known precursor of squamous cell carcinoma. Underlying lesions can be benign (benign nevus, pyogenic granuloma, seborrheic keratoses, lichen simplex, etc.), premalignant (leukoplakia, kerato-acanthoma, actinic keratoses etc.) or malignant (squamous cell carcinoma, basal cell carcinoma, Bowen's disease, Kaposi sarcoma and melanoma, etc.) (8).

E Copcu et substantiated their association with dermatological ailments like solar keratoses, actinic keratoses, kerato-acanthoma, squamous cell carcinoma and basal cell carcinoma and increased likelihood of horns occurring in sun-exposed areas (7). Peter M Nthumba described a giant cutaneous horn in hypopigmented burn scar of more than 20 years duration in a black African lady, nicely concealed by scalp hairs, comprehensively ruling out the possibility of Marjolin's ulcer (9). Olugbenga et al described cutaneous horn in the sole for the first time in the literature in 2011 in a black African lady (2). Ramji et al gave an account of cutaneous horn in yet another sun-protected area in the volar aspect of the forearm with evidence of basal cell carcinoma at its base (10).

While dealing with cutaneous horns, it is necessary to rule out an association with malignant lesions. Our patient's histopathology revealed it to be keratin horn with no evidence of malignancy. He did not show any evidence of recurrence in one year of follow up.

All authors disclose no conflict of interest. The study was conducted in accordance with the ethical standards of the relevant institutional or national ethics committee and the Helsinki Declaration of 1975, as revised in 2000.

References

1. The Mütter Museum. Corkscrew-balloon.com (2003-05-26). Retrieved on 2010-10-27.
2. O Oludiran, V. Ekanem & P. Umebese, "Plantar Cornu Cutaneum Associated with Cutaneous Myxoma," Surgical Science, Vol. 2 No. 1, 2011, pp. 13-15. https://doi.org/10.4236/ss.2011.21004
3. Bondeson J. Everard Home, John Hunter, and cutaneous horns: a historical review. Am J Dermatopathol. 2001 Aug;23(4):362-9. https://doi.org/10.1097/00000372-200108000-00014
4. Yu R C, Pryce D W, Macfarlane A W, Stewart T W. A histopathological study of 643 cutaneous horns. Br J Dermatol. 1991;124:449-452. https://doi.org/10.1111/j.1365-2133.1991.tb00624.x
5. J. W. Gould and R. T. Brodell, "Giant Cutaneous Horn Associated with Verruca Vulgaris," Cutis, Vol. 64, No. 2, 1999, pp. 111-112.
6. V.Shanmugasundaram. Cutaneous Horn in Actinic Keratosis-a Case Report. Journal of Evolution of Medical and Dental Sciences 2013; Vol 2, Issue 24, June 17; Page: 4291-4293, DOI: 10.14260/jemds/834 https://doi.org/10.14260/jemds/834
7. E. Copcu, N. Sivrioglu, N. Culhaci, "Cutaneous Horns: Are These Lesions as Innocent as They Seem to be?" World Journal of Surgical Oncology, Vol. 2, No. 18, 2004. https://doi.org/10.1186/1477-7819-2-18
8. Popadić M. Squamous cell carcinoma presenting as a giant cutaneous horn of the lower lip. Indian J Dermatol Venereol Leprol. 2014;80:74-6. https://doi.org/10.4103/0378-6323.125469
9. P.M. Nthumba, "Giant Cutaneous Horn in an African Woman. A Case Report," Journal of Medical Case Report. 2007; 1:170. https://doi.org/10.1186/1752-1947-1-170
10. Ramji AN. Cutaneous horn in a sun-protected site harbouring unusual malignancy. Int Surg J. 2019 Apr;6(4):1415-1417. https://doi.org/10.18203/2349-2902.isj20191291

Learning Points:

- Cutaneous horns are rare entities normally seen in sun-exposed areas of the body.
- They can also be found in sun protected areas like the sole of foot and penis.
- They can be associated with benign, premalignant and malignant lesions.
- It is imperative to suspect and rule out malignancy in all cases of cutaneous horns.

Answers to images in surgery (from page 36)

1. The chest X-ray shows a simple pneumomediastinum and pneumopericardium with air trapped in the pericardium and tracking along the medial border of the aorta. Blunt and penetrating chest trauma are the commonest aetiological factors, whereas respiratory diseases may also give rise to this condition (1, 2). Subcutaneous emphysema over the anterior chest wall extending to the neck is found to be commonly associated with pneumomediastinum (2) which is also demonstrated in the X-ray. Tension pneumoperi-cardium and tension pneumothorax are life threatening conditions characterized by haemodynamic instability. Nonetheless our patient was haemodynamically stable.

2. Injury to the airway is the commonest reason for the occurrence of concomitant pneumomediastinum and pneumopericardium, and unless diagnosed early it may contribute to a significant mortality (3). Our patient had a carinal injury extending to the right bronchus which was detected on the subsequent CECT chest. Pneumomediastinum occurs when air leaks into the mediastinum via the ruptured peribronchial sheath whereas pneumoperi-cardium results in when the air enters the pericardium through the concomitantly damaged perivascular sheath (2). However, oesophageal and bowel injuries are also associated with pneumomediastinum (2, 3).

3. The initial step of management is to carefully assess the airway, breathing and circulation and to stabilize the patient if there is any haemodynamic compromise while simultaneously liaising with the thoracic surgical team. Pericardial aspiration should be performed if there is evidence of tension pneumopericardium leading to cardiac tamponade (4). CECT chest and bronchoscopy will aid in localizing the injured site. Upper gastrointestinal endoscopy should be performed if an oesophageal injury is suspected. The majority of the cases with simple pneumomediastinum and pneumopericardium can be managed conservatively with careful monitoring given that there is no significant underlying pathology (1). However, if an airway injury is found it needs to be repaired surgically. This patient underwent a right thoracotomy and the tear was repaired with a pericardial patch. She had an uneventful recovery following the surgery.

All authors disclose no conflict of interest. The study was conducted in accordance with the ethical standards of the relevant institutional or national ethics committee and the Helsinki Declaration of 1975, as revised in 2000.

References

1. Kouritas, V.K., et al., Pneumomediastinum. Journal of thoracic disease, 2015. 7(Suppl 1): p. S44.
2. Muckart, D.J.J., T.C. Hardcastle, and D.L. Skinner, Pneumomediastinum and pneumopericardium following blunt thoracic trauma: much ado about nothing? European Journal of Trauma and Emergency Surgery, 2018: p. 1-5. https://doi.org/10.1007/s00068-018-0960-5
3. Nwashilli, N., O. Irowa, and E. Ohanaka, Pneumoperitoneum, pneumomediastinum, pneumopericardium and subcutaneous emphysema following colonoscopy-a case report. Ann Biomed Sci. Vol, 2016. 15(1).
4. Ladurner, R., et al., Pneumopericardium in blunt chest trauma after high-speed motor vehicle accidents. The American journal of emergency medicine, 2005. 23(1): p. 83-86.