

CosmoDerma



Cornu cutaneum – Past and present

Govind Srivastava¹, Gautam Srivastava²

¹Department of Dermatology, Venereology and Dermatologic Surgery, Skin Institute and School of Dermatology, Delhi, India, ²Department of Faculty of Life Sciences and Education, Faculty of Life Sciences and Education, University of South Wales, Cardiff, United Kingdom



Review Article

***Corresponding author:** Govind Srivastava, Skin Institute and School of Dermatology, Delhi, India.

sisdoctor@yahoo.co.in

Received : 30 June 2021 Accepted : 20 July 2021 Published : 18 August 2021

DOI

10.25259/CSDM_31_2021

Quick Response Code:



ABSTRACT

Cutaneous horn has been a source of controversy throughout the history. It evoked interest nearly half a millennium before and continues to do so. Cornu cutaneum has been found to be associated with several premalignant and malignant skin conditions. The pre-malignant conditions commonly include actinic keratoses and Bowen's disease. Uncommonly, it is due to the presence of an underlying squamous cell carcinoma. Thus, an effort is needed to determine the pathology present at the base of the horn for proper management of the disease. Wide excision biopsy is a common denominator in the therapy of this peculiar disorder.

Keywords: Cutaneous horn, Skin cancer, Premalignant lesions, Hyperkeratosis, Photoallergy

INTRODUCTION

Cornu cutaneum (CC) is a fascinating entity. Popularly known as cutaneous horn, it is a slowly developing exophytic protrusion on the skin or mucous membrane. With time, it becomes firm, conical and develops a curvature, resembling an animal's horn. Accepted as a reactive cutaneous lesion, it can be induced by a variety of benign, pre-malignant, or malignant processes.^[1] CC jut out of the skin as hard, dry, yellow, brown, or dark conical projection, on a normal or slightly erythematous, fleshy base. Neglect or personal stigmas often keep such patients from reporting for a proper remedy. As these lesions are largely asymptomatic, this contributes to the delay.^[2] The bigger horns grown over years and decades have been referred to as giant cutaneous horn or "devil's horn." The present review attempts to trace its interesting history and to update on this uncommon entity.

HISTORICAL ASPECTS

Bondeson^[3] has beautifully researched the historical aspects of cutaneous horn. The first recorded case of CC was an elderly Welsh woman, Margaret Gryffith. She was exhibited in circuses and prominent gatherings for money by a showman in London in the year 1588.^[4] In the following decade (1598), another case was discovered by French hunters in the forests near Mezieres. This was Francois Trouvillou, a 35-year-old man who used to stay in seclusion after being banished from the village at a young age due to a CC on his head. Being scared of being killed or imprisoned as a monster, he always covered his head with a hat. After being brought to Paris by his French discoverers, he was shown to King Henry IV, who permitted him to be displayed to people of Paris for money. In those times, the exhibition of humans and animal curiosity/abnormalities earned a lot of interest among local people.^[3,5,6] Although Francois Trouvillou died in the year

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1599 and buried in the Saint-Come cemetery in Paris, the dialog about human horns did not die. Some advanced natural causes for this anomaly while others attributed it to some supernatural cause.^[7] Margaret Gryffith herself had attributed her horn due to her wishful thinking that a horn would grow on her head. She did this in a fit of anger over allegations by her husband.^[3] Francois Trouvillou, however, had no such superstition. He was aware of his horn in his childhood at the tender age of seven, which slowly assumed a finger-like size by the age of seventeen. It grew backward on his scalp and he often had to cut its tip to avoid pain due to it pressing on the skull.^[3,5]

Cutaneous horns remained a nature's mystery. No serious attempts were made to study its scientific basis for several decades. This credit goes to Bartholin, a Danish anatomist who thoroughly studied the previously recorded cases with apt attention.^[8] He was dying to see a living case of CC with his own eyes to research its pathogenesis. In 1646, he happened to see a 70-year-old woman, Margretha Mainers with a cutaneous horn growing on her right temple. On a closer examination, he found that this horn is emerging out of a reddish skin tumor which was itchy and often bled on scratching. Although this woman also attributed this to her bitter quarrels with her prodigal son; Bartholin with his well-researched work cleared the doubts that there is nothing obscure or supernatural about these horns. He vehemently denied any supernatural links to its development and stressed an unknown biological process.^[2,3,8,9] Inspired by Thomas Bartholin, a German scholar, Franck wrote a 31-page treatise entirely devoted to cutaneous horns in the year 1676. He gave arguments both in favor and against the superstitions, with his personal bias toward the supernatural.^[10]

Soon, in the year 1680, another old English woman, Marie Davies made the entry to the London show business. Thought to be born in 1591, she recalled that she suffered soreness of the head for 20 years wearing a straight hat. Gradually, she developed a "wen," the size of a hen's egg, which in the course of 5 years became a solid wrinkled horn like Ram's horn. She shed her first pair of horns after 4 years, another set after 4 more years while the third set of horns broke off after she slipped. The shed horns were sold to wealthy noblemen for a price. Her fate and the fate of her last pair of horns, while she was on display in London, was not traced further.^[3,11,12]

There is a mention of another woman, Mrs. French, living near Tenterden, with an 11-inch horn. She is quoted in the Van Rymsdyk and Van Rymsdyk Museum Britannicum.^[13] Nearly at the same time, Elizabeth Lowe, a Scotch woman got her 4-inch horn excised, which is still preserved in the anatomic museum of Edinburgh University.^[14,15] These specimen of CC kept the curiosity alive in the mind of people as well as scientists.

In the year 1791, another authoritative and authentic study on CC was published by a prolonged research by Home and Bondeson^[3,16] Fifty-six-year-old Ms. Lonsdale from Horncastle, Lincolnshire had 4 horns growing out of a cystic scalp tumor. Another fairly young woman, Ms. Elizabeth Allen from Leicestershire, had a prominent horn from an underlying growth. They postulated that an underlying encysted skin tumor or repeated local trauma acted as a trigger for an altered growth of the skin which gradually results in deposition of horny substance forming CC. Apart from the head and face which are difficult to hide, CC can develop on any part of the body.^[16,17]

In the following decades, isolated cases were observed. In 1820, a Mexican man, Paul Rodriguez, revealed a group of three horns with a massive 14-inch circumference - which he kept as a secret for decades. One day, he suffered a head injury, lost consciousness and started bleeding heavily. People came to his rescue, removed his head guard and were shocked to see such massive horns.^[18] Widow Dimanche, an elderly French woman, had an enormous curved horn on her forehead which was 10 inches long 2 inches wide. Although the weight of this CC was fatiguing this lady, yet she dreaded any surgical intervention. She finally consented to surgery in the late 1830s. Her justification for the delay was that she did not want to meet God with such a satanic ornament on her face.^[19] In the year 1834, Giese removed a thick curved horn above the right eye from an 87-year-old abbess of Filzen nunnery of Germany. Despite its successful removal, the wound failed to heal due to the skin tumor beneath and a small horn grew at the same site.^[20]

Lebert, in the year 1864, published an elaborate collection of 109 cases of CC.^[21] After nearly 5 decades, in 1911, Bland-Sutton classified them in four varieties, namely, Sebaceous, Wart, Cicatrix, and Nail horns.^[22] In 1941, Montgomery^[23] classified CC into five types based on appearance, histological structures and causative factors. Since then, several workers have shed more light on its pathogenesis and management.^[24-27] At present, the dermatologic community focuses on the underlying base of CC whose histopathology gives a clue to its precise etiology [Table 1].

EPIDEMIOLOGICAL FACTORS

No specific epidemiological studies are available for this disease. CC is believed to be more common in the Caucasian population when compared to African or Asian population. The first statement pertaining to the epidemiology of CC came from a German scholar, Zacharias Conrad von Uffenbach, who happened to see 3 specimens of giant horn in the collection of the famous antiquary Elias Ashmole. He postulated, rather preposterously, that men generally had their horn on the forehead, while women have them on the back of their head. Further, he stressed that England must

Table 1: Classification of cornu cutaneum.							
S. No.	Bland-Sutton ^[22] (Type I–IV)	Montgomery ^[23] (Type I–V)	Present day concept (Based on pathology of underlying base of the horn)				
1.	Sebaceous horn (Origin from sebaceous cyst on scalp)	Cutaneous horn (Origin from epidermoid cyst)	Benign base				
2.	Wart horn (Usually seen on penis)	Mucosal horn (Origin from mucous membrane)	Premalignant base				
3.	Cicatrix horn (Develops on burn scars)	Verrucous horn (Origin from wart)	Malignant base				
4.	Nail horn (In neglected bedridden patients)	Papillomatous horn (Origin from cornified epithelium)	Reaction to a distant concomitant malignancy				
5.		Filiform horn (Origin from normal/hyperkeratotic skin)					

have a climate specifically predisposing for the development of CC.^[28] However, being an infrequently reported entity, the true incidence, prevalence, age/sex incidence or geographic variation was difficult to estimate. Certain factors, nevertheless, are more consistently observed by several researcher workers: Chronic exposure to sunlight, repeated trauma, fairer skin, and advancing age.^[1,22-24,27]

It has been reported that the mean age of patients with a benign etiology of horn base is 8.9 years younger than those with a premalignant/malignant base pathology.^[25,27] No sex predilection has been observed in benign CC. However, in cases of premalignant or malignant CC, males distinctly outnumber females. Males have a history of more outdoor occupations leading to excessive exposure to ultraviolet light which predisposes them to several premalignant/malignant cutaneous disorders – ultimately resulting in a higher incidence of CC. Such horns are mostly located on the head, face, neck, and upper extremities of older males and have a broad base.^[25] However, CC are known to occur anywhere in the body, including the penis.^[22,23,25,29]

Poor socioeconomic status, old age, isolated living conditions all favor development of giant CC. These people are victims of neglect and the medical facilities largely unamenable to them. They are scared to disclose the condition fearing further social boycott.^[24,30]

ETIOPATHOGENESIS

CC is a clinical term collectively ascribed to the lesions which are circumscribed, conical, markedly hyperkeratotic, where the height of the keratotic mass is at least half of its largest diameter.^[24] It is essentially a reaction pattern and not a specific lesion.^[31] The exact pathogenesis of this entity is obscure. However, certain factors such as photodamage, cellular aging, and chronic irritation are believed to trigger this type of reaction. The previous classifications of CC by Bland-Sutton^[22] or Montgomery^[23] are historical but no longer in vogue. Now, it is well-established that the horn *per se* is an insignificant, obnoxious mass of dead keratin. It is the base of the horn where the underlying pathology lies, which affects the prognosis.^[26] This etiology of CC may be benign, premalignant, or malignant [Table 2].

Yu *et al.*^[25] studied the histopathological details of 643 cutaneous horns. They found that 65% of them were benign and caused by epithelial hyperplasia or cutaneous warts, 20% by premalignant conditions, predominantly solar keratosis, and Bowen's disease, while the remaining 15% by malignant skin lesions, chiefly squamous cell carcinoma (SCC).

CLINICAL FEATURES

CC is a slow-growing exophytic growth that is usually asymptomatic. Some patients give a preceding history of some bumps or changed skin texture for a variable duration. Once the horn forms, it has no tendency of resolution; it may keep growing and may attain an enormous size. It is asymptomatic but its base in some cases may be tender and itchy [Figure 1]. The length and diameter of CC are variable. Bland-Sutton^[22] recorded that the sebaceous horn originating from a sebaceous cyst involves chiefly the scalp and attains large proportions. It reveals a sebaceous cyst at its base in histopathology. Wart horns usually develop on the penis but lack encysted structures at their base.^[32-34] Nail horns were the most common horns, according to him, which usually developed in the nails of big toes of old, neglected, and bedridden patients. The length of the horn can vary from less than a cm to over 25 cm, while the diameter at the base varies from a few mm to a few cm.^[35-39] Small horns are always straight; as the horn grows, it develops a curvature and a darker hue. The surface of the horn can vary from smooth to corrugate longitudinally or transversely. Multiple horns have been recorded in a single patient. After surgical/traumatic removal, some of them may re-grow.[1,2,24,26,37] Giant horns are often associated with malignancy, though it is not a rule.^[23,30,40-42]

Table 2: Etiology of cornu cutaneum (According to the pathology of the underlying base). [1,24-27,31-39]							
Benign base	Premalignant base	Malignant base	Reaction to distant malignancy				
Angiokeratoma Chronic friction Epidermal nevus Haemangioma HPV Juvenile xanthogranuloma Lichenoid keratoses Molluscipox virus Pilomatricoma Pyogenic granuloma Rhinosporidiosis Sarcoidosis Sebaceous adenoma Seborrhoeic keratoses Tricholemmoma	Actinic keratoses Arsenic keratoses Bowen's disease Keratoacanthoma Micaceous balanitis Pseudoepitheliomatous keratoses	Basal cell epithelioma Carcinomas Kaposi's sarcoma Squamous cell carcinoma Verrucous carcinoma	Renal cell carcinoma				
HPV: Human papilloma virus							



Figure 1: Cornu cutaneum seen on the neck of a 64-year-old woman. The skin at the base was erythematous and itchy. The growth was present for the past 5 years and the patient refused surgical intervention due to the growth being "lucky" for her. She refused skin biopsy for histopathological examination.

Pyne *et al.*^[43] studied clinical and dermoscopic features of 163 consecutive CC patients and correlated them with the histopathology of the base of the horn. Microscopic study revealed 49 benign keratoses, 21 actinic keratosis, 37 SCC *in situ*, and remaining 56 invasive SCC. They concluded that the invasive SCC had a significantly less terrace morphology, but a higher incidence of base erythema as well pain, when compared with other three entities. Further, invasive SCC horns had a height less than their base diameter.

Horns of various sizes and shapes are a common character for certain species of animals. These grow from the bones of these animals, occupy the same anatomic area and act as a weapon of defense. The distinction between CC and animal horn is given in Table 3.

HISTOPATHOLOGY

CC is essentially a benign outgrowth, no different than a dead piece of wood. However, the pathology of the underlying base must be found out while treating these patients. For the same reason, it is imperative to treat the giant horns with adequate margin, for they have a greater chance of malignancy at their base. Thus, histopathology of the base of CC is mandatory to find out its precise etiology. Irrespective of the pathology of the base, CC of any origin displays dead keratin. By and large, the horns reveal a greatly thinned stratum corneum, overlying compact, redundant hyperkeratosis with some pockets of parakeratosis. The underlying dermis, if any, is scanty and fibrotic.^[25,26,44] Cystic structures lined by trichilemmal-type epithelium can be seen in older horns.^[44]

DIAGNOSIS

The diagnosis of CC is largely clinical. However, the histopathology of the underlying base is required in each case to clinch the etiology [Figures 2-4]. As a wide variety of benign, premalignant and malignant conditions are encountered at the base of CC, their diagnosis will guide in the management of such lesions.^[1,24-28,45]

MANAGEMENT

Irrespective of its size, a cutaneous horn needs an excisional biopsy. Its base has to be included for histopathological evaluation. It is prudent to excise it with a 3–10 mm margin depending on the clinical suspicion. If the base is broad,

Table 3: Difference between animal horn and cornu cutaneum. [26,42,44]						
Features	Animal horn	Cornu cutaneum				
Nature	Physiological, species specific, area specific	Pathological, rare				
Morphology	Typical horns, firmly attached to bones	Variable size, no attachment to bone				
Base of horn	Bone	Underlying diseased skin				
Central bony core	Present, provides strength and can be used as a weapon	Absent				
Association with disease	Not associated	Underlying skin disease				
Histopathology	Compact hyperkeratosis and dermis encircling a central, well-developed bone. Absence of cystic	Greatly thickened stratum corneum, scanty fibrotic underlying dermis.				
	structures/trichilemmal type epithelium	Cystic structures lined by typical trichilemmal type epithelium				



Figure 2: Cutaneous horn on the back resembling a fingernail; a yellowish-whitish, curved 4 cm protrusion, representing a keratoacanthoma. (Fernandes *et al.*, Acta Dermatoven APA Vol 18, 2009; reprinted with permission).



Figure 3: Cutaneous horn shown in Figure 2; a keratoacanthoma with typical hyperkeratosis with associated parakeratosis, overlying a keratoacanthoma (hematoxylin-eosin $\times 100$). (Fernandes *et al.*, Acta Dermatoven APA Vol 18, 2009; reprinted with permission).

the resultant defect may need a split-skin graft or Z-plasty. For penile horns, a pre-operative MRI can give a clue to the



Figure 4: Edge of the keratoacanthoma, at the base of the cutaneous horn, with proliferating squamous epithelial cells (hematoxylineosin \times 100). (Fernandes *et al.*, Acta Dermatoven APA Vol 18, 2009; reprinted with permission).

status of infiltration.^[46] Smaller CC can be removed using electrocautery or radiofrequency. Newer modalities include Nd YAG laser and CO₂ laser gives good cosmetic results. Thappa *et al.*^[47] believe that CC is first-stage of a two-stage diagnosis where the second stage determines the nature of the underlying lesion clinically or histologically. This may accordingly modify the management practice. A follow-up is recommended in patients with giant horns as they are known to recur.

PROGNOSIS

Early lesions and many cases with benign histopathology of the base have an excellent prognosis and fewer chances of recurrence.^[30,40,48] The patients who report late worsen their prognosis and increase chances of recurrence. The underlying pathology of the base may also influence the outcome.

CONCLUSION

CC is a rare and unusual condition with a historically rich background. For almost half a millennium, its origin and

peculiarity have intrigued humankind. Several hypotheses have been put out for its occurrence ranging from natural causes to supernatural powers. CC is essentially a benign lesion originating from a benign, premalignant, or malignant base. Ultraviolet light is thought to be the chief perpetrator in the etiopathogenesis of this disease. An excisional biopsy with the base of the lesion reveals the underlying cause, which helps to formulate a treatment plan. With the advent of the modern healthcare system and greater awareness among people, giant cutaneous horns are seldom seen and will soon become a marvel of the old world.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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How to cite this article: Srivastava G, Srivastava G. Cornu cutaneum – Past and present. CosmoDerma 2021;1:33.