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Cutaneous horn occurring on the oral tissues: report of a case and review of literature

Corno cutâneo nos tecidos orais: relato de caso e revisão da literatura

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ABSTRACT

The aim is to describe the clinical-pathological and treatment data of a patient with a cutaneous horn (CH) arising on oral tissues and to compare this with a literature-review. This case affects the lower-lip of an 86-years-old female. After excisional biopsy, the histopathological examination revealed a CH associated with actinic cheilitis. Only 23 similar cases have been reported in literature. At this site CH mainly affects the lower-lip of white males in a wide-age range. Excision should be the treatment of choice to enable pathological examination of the underlying lesion, since most cases were associated with pre-malignant/malignant lesions.

Key words: cheilitis; mouth; lip; epidemiology.

RESUMO

O objetivo deste relato é descrever os dados clinicopatológicos e de tratamento de uma paciente, 86 anos, com corno cutâneo (CC) nos tecidos orais (lábio inferior) e compará-los com outros estudos publicados, fazendo uma revisão da literatura. Após biópsia excisional, o exame histopatológico revelou CC associado a queilite actínica. Apenas 23 casos semelhantes já foram relatados. Durante nossa pesquisa, verificamos que o CC afetou principalmente o lábio inferior de homens brancos em uma ampla faixa etária. A excisão deve ser o tratamento de escolha, pois ela possibilita a realização de exame histopatológico da lesão subjacente, já que a maioria dos casos foi associada a lesões pré-malignas/malignas.

Unitermos: *queilite; boca; lábio; epidemiologia.*

RESUMEN

El objetivo es describir los datos clinicopatológicos y de tratamiento de una paciente de 86 años con un cuerno cutáneo (CC) en los tejidos orales (labio inferior) y compararlos con una revisión de la literatura. Tras la biopsia escisional, el examen histopatológico reveló un CC asociado a queilitis actínica. Solo se han informado 23 casos similares en la literatura. El estudio ha revelado que el CC afecta principalmente al labio inferior de los hombres blancos en un amplio rango de edad. La escisión debe ser el tratamiento de elección para permitir el examen patológico de la lesión subyacente, ya que la mayoría de los casos se asociaron con lesiones premalignas/malignas.

Palabras clave: *queilitis; boca; labio; epidemiologia.*

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INTRODUCTION

Cutaneous horn (CH) is a clinical term for describing a yellowish or brownish conical, hyperkeratotic protrusion on the mucous and/or skin surface that clinically resembles the horn of an animal. It may be straight or curved and twisted and could vary from a few millimeters to several centimeters in length. The lesional base may be flat, nodular, verrucous, or crateriform[1-5].

It is important to notice that this term does not indicate any specific pathological diagnosis, since it can develop over a wide array underlying lesions[1-5]. CH is a relatively unusual lesion, affecting mostly the sun exposed areas of Caucasian mid-age patients (≥ 50 years of age), without a clear gender predilection[1-5].

The horn itself is characterized by hyperproliferation and increased cohesiveness of keratin due to an unknown mechanism. Most cases develop from an underlying benign lesion, however about 40% of all CH will arise from pre-malignant or even malignant diseases[1-5]. CH must be treated by surgical excision, followed by an accurate determination of the nature of the condition at the base of the lesion. The surgical excision should be as conservative as possible and should also guarantee a free-lesional margin[1-5].

Since there were very few CH cases arising in oral tissues, the goal of this study was to describe the sociodemographic data, histopathology, treatment, follow-up, and outcomes of one patient affected with CH on the lower lip and compare this data with a comprehensive review of the current literature.

CASE REPORT

An 86-years-old, non-smoker, Caucasian female was referred by a private clinician to our Stomatology Service, for evaluation of a solitary, asymptomatic, slowly enlarging, and cylindrical growth in the lower lip. She denied any history of previous sunlight chronic exposure, trauma, spontaneous bleeding, or ulceration in the affected area.

In the anamnesis, patient complained that this injure persisted for more than 10 years and was very uncomfortable, mainly due to the aesthetic reasons. Her past medical history revealed controlled hypertension and diabetes. The measurements of the blood pressure, pulse and glycemia recorded at first appointment was within normal range.

Extra oral physical examination, revealed a well-circumscribed, exophytic, conical shaped, sessile and consistent nodule, with yellowish and brownish coloration. The lesion was located in the vermilion of the left lower lip and measured approximately 1.5 × 1 cm at its largest diameters (**Figure 1A** and **1B**). There was no induration or erythema at the base of this lesion. It is also possible to note thickened and scaly plaques in her lower lip, compatible with actinic cheilitis. Additionally, there was no palpable lymph node in the neck. Intraoral examination revealed long term partial edentulous jaws rehabilitated by removable prosthodontics.

A clinical diagnosis of CH of undetermined nature was achieved and an excisional biopsy was performed under local anesthesia (**Figure 1C**). The defect was partially closed and healed by second intention uneventfully (**Figure 1D**). The material obtained was conditioned in 10% formaldehyde and subsequently referred to the surgical pathology service.

The histopathological examination depicted a diffuse conical hyperkeratosis and parakeratosis consistent with CH (**Figure 2A**). At the base of the lesion, the stratified squamous epithelium exhibited hyperkeratosis, spongiosis, acanthosis and mild-to-moderate epithelial dysplasia (**Figure 2B** and **2C**). The underling stroma showed a moderate inflammation composed of lymphocytes, plasma cells, and neutrophils (**Figure 2B** and **2C**). The definitive diagnosis was CH associated with actinic keratosis. No clinical relapses were detected after 2 years of follow-up (**Figure 1E**). The patient was advised to wear labial sun protection and to keep regular clinical follow up.

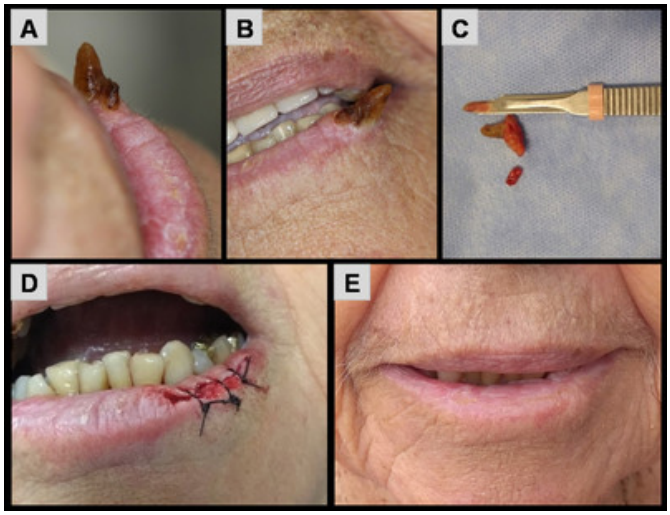

FIGURE 1 – *Clinical image shows: A) solitary, asymptomatic, slowly enlarging, and cylindrical growth in the lower lip; B) lesion in the first appointment; C) surgical specimen; D) immediate post-operative aspect; E) two-years follow-up*

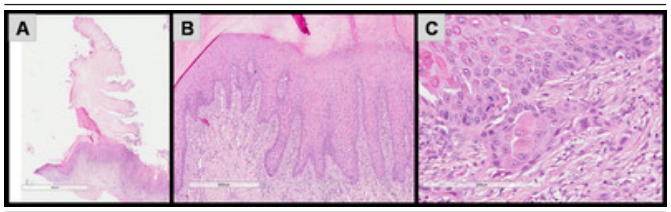

FIGURE 2 – *Histopathological image shows: A) conical hyperkeratosis and parakeratosis consistent with CH; B) at the base of the lesion, the stratified squamous epithelium exhibited hyperkeratosis, spongiosis, acanthosis; C) epithelial area depicting mild-to-moderate epithelial dysplasia*

CH: cutaneous horn.

DISCUSSION

Although CH is a rare and an apparently innocent lesion, we should be aware of the pre-malignant and/or malignant potential of this disease. **Tables 1** and **2** demonstrate the main clinical features of the CH cases affecting oral tissues, published in the Literature[6-26].

On this current review, 23 CH cases arisen in the oral tissues have been retrieved[6-26]. The primary lesions associated with CH arising in oral tissues may be histopathologically benign (discoid lupus

TABLE 1 – Clinical pathological data of retrieved cases from international literature

Number	Gender/age (years)	Skin color	Site	Size (cm)	Underlying pathology	Treatment	Recidive?	Outcome/follow-up (months)
1 [6]	M/56	W	LL	-	Acanthosis and epithelial dysplasia	ST	No	NED/-
2 [7]	M/50	-	LL	-	-	ST	No	NED/-
3 [8]	M/21	NW	LL	1.2 × 0.8	Acanthosis and epithelial dysplasia	ST/isotretinoin	Yes – 3×	AWD/-
4 [9]	M/11	NW	LL	2.4 × 0.8	Pyogenic granuloma	ST	No	NED/19
5 [10]	F/45	-	BM	2 × 0.8	Basal cell carcinoma	ST	No	NED/-
6 [10]	F/67	W	LL	2.5 × 0.6	Actinic keratosis	ST	No	NED/-
7 [11]	F/50	NW	LL	1 × 0.5	Pyogenic granuloma	ST	No	NED/12
8 [12]	M/16	W	LL	3.2 × 0.9	Acanthosis and epithelial dysplasia	ST	No	NED/36
9 [13]	F/72	W	UL	2 × 0.8	Well-differentiated SCC	ST	No	NED/66
10 [14]	M/72	W	LL	3.2 × 2.5	Well-differentiated SCC	ST	No	NED/24
11 [15]	M/45	W	LL	3 × 1.5	Keratoacanthoma/ well-differentiated SCC	ST	No	NED/24
12 [16]	M/9	NW	UL	-	Discoid lupus erythematous	ST	No	NED/-
13 [17]	M/45	NW	OC	3 × 1	Verrucous carcinoma	ST	No	NED/24
14 [18]	F/76	W	LL	6.3 × 1.9	Well-differentiated SCC	ST	No	NED/37
15 [19]	M/66	W	UL	3 × 2	Well-differentiated SCC	ST	No	NED/24
16 [20]	M/77	W	LL	3 × 2	Keratoacantoma	ST	No	NED/24
17 [21]	M/74	-	LL	0.6	Actinic keratosis	ST	No	NED/12
18 [22]	F/63	NW	OC	3 × 1	Well-differentiated SCC	ST	No	-/-
19 [23]	F/82	W	UL	3.5	Well-differentiated SCC	ST	No	NED/6
20 [24]	M/11	NW	LL	2 × 1	Pyogenic granuloma	ST	No	NED/2
21 [25]	M/59	NW	UL	1.5	Verrucous carcinoma	ST	Yes – 2×	-/-
22 [26]	F/52	NW	OC	3 × 1	Verrucous carcinoma	ST	No	NED/-
23*	F/86	W	LL	1.5 × 1	Acanthosis and epithelial dysplasia	ST	No	NED/24

*M: male; F: female; W: white; NW: non-white; LL: lower lip; BM: buccal mucosa; OC: oral commissure; UL: upper lip; SCC: squamous cell carcinoma; ST: surgical treatment; NED: no evidence of disease; AWD: alive with disease; *current study.*

TABLE 2 – Summary findings from the 23 patients with CH occurring on the oral tissues

Gender	M:F ratio: 1.6:1
Age (years)	Mean: 52.4 ± 24.1 (range: 9-86)
Skin color	W:NW ratio: 1.2:1
Site	Lower lip: 14/23 (60.9%) Upper lip: 5/23 (21.7%) Oral commissure: 3/23 (13.1%) Buccal mucosa: 1/23 (4.3%)
Size (cm)	Mean: 2.5 ± 1.2 (range: 0.6-6.3)
Signs and symptoms	Swelling: 23/23 (100%) Itching and/or soreness: 2/23 (8.7%) Bleeding: 1/23 (4.3%)
Evolution time (months)	Mean: 22.2 ± 17 (range: 1-60)
Underlying pathology	Benign: 4/22 (18.2%) Pre-malignant: 7/22 (31.8%) Malignant: 11/22 (50%)
Treatment modalities	ST: 22/23 (95.7%) ST + isotretinoin: 1/23 (4.3%)
Recidive?	Yes: 2/23 (8.7%)
Outcomes	NED: 20/21 (95.2%) AWD: 1/21 (4.8%)
Follow-up (months)	Mean: 23.9 ± 15.7 (range: 2-66)

CH: cutaneous born; M: male; F: female; W: white; NW: non-white; ST: surgical treatment; NED: no evidence of disease; AWD: alive with disease.

erythematous or pyogenic granuloma), premalignant (acanthosis and epithelial dysplasia or actinic keratosis), or malignant (keratoacantoma or verrucous carcinoma or well-differentiated squamous cell carcinoma or basal cell carcinoma)[6-26].

The reported incidence of CH with premalignant or malignant histological features varies considerably worldwide. In 1979, Schosser *et al.* (1979)[4] reported a series of 230 CH in a USA population, of which 58% showed either premalignant or malignant at their base. In contrast, Yu *et al.*, 1991[1], described 643 CH cases arising in English patients, and revealed that only 38.9% were derived from malignant or premalignant epidermal lesions and 61.1% from benign lesions.

Regarding South America patients, there were three papers reporting the frequency of the CH underlying nature[2, 3, 5]. Festa *et al.* (1995)[2] reported that 25.4% of underling lesions were benign, 49.3% were premalignant lesions and 25.3% were malignant lesions. These findings were similar to Castilho *et al.* (2002)[3] which observed 46% of benign lesions, 41% of premalignant lesions and 13% of malignant. In 2009, Mantese *et al.* (2010)[5] also appointed that the majority of the cases were derived from premalignant and malignant lesions (58.56%). In the oral tissues (Table 2), pre-malignant and malignant lesions prevailed in a very high frequency, when compared with the above sited articles (18/22, 81.8%). Benign lesions were quite unusual at this location and represents only 18.2% (04/22)[6-26].

The literature agrees that risk factors associated with an underlying malignancy include advanced age, male sex, quickly development, previous history of trauma, presence of hardness at the base, large base or height-to-base ratio and location on sun-exposed areas[1-5, 10, 12, 18]. In the current casuistry (**Table 3**), older patients (≥ 45 years), with lesion located in other sites than in the lower lip and with bigger lesions (≥ 3 cm) were statistically associated with malignant-underlying-lesion.

However, CH patients had been described since the 16th century, until today very little is known about it predilections, treatments and outcomes, especially regarding CH arisen in the oral maxillofacial region. The sun exposure is the most important etiological factor in the pathogenesis of the CH, as seen with other skin lesions[1-5]. However, poor oral hygiene and smoking may have an additional role in the occurrence of lesion affecting the oral tissues[10, 12, 18].

In this particular region (Table 2), CH commonly affects white, male patients, in a wide age range[6-26]. Similar data were depicted everywhere[1-5]. Lower lip is the prevalent site in this review, affecting 60.9% of the cases; been followed by upper lip, oral commissure and buccal mucosa[6-26]. Lesions at these sites usually were usually asymptomatic, and for this, patients may delay to seek a medical/dental attention, the mean evolution time was close to 2 years[6-26]. However the long evolution, lesions were usually small (2.5 ± 1.2 × 1.2 ± 0.6 cm)[6-26].

Although the high frequency of malignant and/or pre-malignant underling lesions at this site, recidivism were not common seen (02/23, 8.7%). Most of the patients achieved cure and remained with no evidence of disease[6-26]. In this Literature-retrieved casuistry, the mean follow-up time was 23.9 ± 15.7

TABLE 3 – Contingency table demonstrating the associations between underlying pathology nature and clinical pathological features

Data	Sub-groups	Benign/pre-malignant	Malignant	*p**
Gender	Male	8	5	NS
	Female	3	6	
Age	≥ 45 years	6	11	0.0175
	< 45 years	5	0	
Skin color	White	5	6	NS
	Non-white	5	4	
Site	Lower lip	10	3	0.0038
	Others	1	8	
Size	≥ 3 cm	2	8	0.0349
	< 3 cm	7	3	
Signs and symptoms	Only swelling	7	10	NS
	Swelling + others	3	0	
Evolution time**	> 21 months	4	4	NS
	≤ 21 months	3	4	
Recidive?	Yes	1	1	NS
	No	10	10	

*NS: statistically not significant; *OneFisher's exact test (one-sided); **segregated accordingly with median.*

months, this follow up time seen to be inadequate, especially when dealing with of malignant and/or pre-malignant conditions[6-26].

Surgical excision, followed by histopathological examination of the base of the lesion were the treatment of choice[10, 12-14, 18]. In addition, follow-up examinations to screen for a recurrence or a new primary lesions are essential for these patients[10, 12-14, 18]. In our case, the excisional biopsy was performed in reason that the lesion depicted clinical benign features such as no induration, erythema or pain at this base; also there is no palpable lymph node in the neck. Since there were a huge percentage of malignant diseases at the CH-base in oral tissues, excisional biopsy must be performed with care.

Cryosurgery is not advocated for the treatment of CH as it does not ensure full thickness excision of the tumor and also it is not appropriate for the treatment of squamous cell carcinomas[10, 12-14, 18]. In cases of squamous cell carcinomas, proper clinical and laboratory staging is recommended, in addition to postoperative radio and/or chemotherapy. Reconstruction of large lower lips defects can be achieved usually by Karapandzic flap, Bernard flap, or modifications of these reconstructions techniques[10, 12-14, 18].

CONCLUSION

CH arising at oral tissues affects mainly the lower lip of white males in a wide age range. Full thickness excision with negative margin should be the treatment of choice to enable detailed pathological examination of the underlying tissue, since at this site there is a huge frequency of pre-malignant and/or malignant lesions.

DECLARATION OF PATIENT CONSENT

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

CONFLICTS OF INTEREST

There are no conflicts of interest.

REFERENCES

1. Yu RC, Pryce DW, Macfarlane AW, Stewart TW. A histopathological study of 643 cutaneous horns. Br J Dermatol. 1991; 124(5): 449-52. doi: 10.1111/j.1365-2133.1991.tb00624.x.

2. Festa CN, Falda S, Rivitti EA. Corno cutâneo: estudo retrospectivo de 514 casos. An Bras Dermatol. 1995; 70(1): 21-5.

3. Castillo D, Zerpa O, Loyo N, López C, Oliver M. Histopatologia del cuerno cutaneo: estudio retrospectivo de 77 casos. Derm Venez. 2002; 40(3): 65-9.

4. Schosser RH, Hodge SJ, Gaba CR, Owen LG. Cutaneous horns: a histopathologic study. South Med J. 1979; 72(9): 1129-31. doi: 10.1097/00007611-197909000-00014.

5. Mantese SA, Diogo PM, Rocha A, Berbert AL, Ferreira AK, Ferreira TC. Cutaneous horn: a retrospective histopathological study of 222 cases. An Bras Dermatol. 2010; 85(2): 157-63. doi: 10.1590/s0365-05962010000200005.

6. Monaghan AM. Cutaneous horn occurring on the vermilion border of the lower lip. Br Dent J. 1993; 175(11-12): 419-20. doi: 10.1038/sj.bdj.4808346.

7. Nahlieli O, Baruchin AM, Shapira Y, Ben-Dor D. Cutaneous horns occurring on the head and neck region: report of four cases and review of the literature. J Oral Maxillofac Surg. 1997; 55(11): 1309-11. doi: 10.1016/s0278-2391(97)90190-7.

8. Baykal C, Savci N, Kavak A, Kurul S. Palmoplantar keratoderma and oral leucoplakia with cutaneous horn of the lips. Br J Dermatol. 2002; 146(4): 680-3. doi: 10.1046/j.1365-2133.2002.04595.x.

9. Souza LN, Martins CR, de Paula AM. Cutaneous horn occurring on the lip of a child. Int J Paediatr Dent. 2003; 13(5): 365-7. doi: 10.1046/j.1365-263x.2003.00481.x.

10. Copcu E, Sivrioglu N, Culhaci N. Cutaneous horns: are these lesions as innocent as they seem to be? World J Surg Oncol. 2004; 2(1): 18. doi: 10.1186/1477-7819-2-18.

11. Patil K, Mahima VG, Lahari K. Extragingival pyogenic granuloma. Indian J Dent Res. 2006; 17(4): 199-202.

12. Mutaf M. A rare perioral lesion: cutaneous horn of the lower lip. Eur J Plast Surg. 2007; 29(7): 339-41. doi: 10.1007/s00238-006-0101-y.

13. Islam A, Can IH, Dürer P, Demirci M. Cutaneous horn on the upper lip associated with squamous cell carcinoma. Kulak Burun Bogaz Ihtis Derg. 2009; 19(6): 304-6.

14. Skoulakis, C., Theos, E., Chlopsidis, P. Giant cutaneous horn on squamous cell carcinoma of the lower lip. Eur J Plast Surg. 2009; 32(1): 257-9. doi: 10.1007/s00238-009-0338-3.

15. Pinto-Almeida T, Oliveira A, da Cunha Velho G, Alves R, Caetano M, Selores M. Giant cutaneous horn on the lower lip. Dermatol Online J. 2011; 17(12): 10.

16. Chowdhury J, Kumar P, Gharami RC. Multiple cutaneous horns due to discoid lupus erythematosus. Indian J Dermatol Venereol Leprol. 2014; 80(5): 461-2. doi: 10.4103/0378-6323.140315.

17. Kumar S, Bijalwan P, Saini SK. Carcinoma buccal mucosa underlying a giant cutaneous horn: a case report and review of the literature. Case Rep Oncol Med. 2014; 2014(1): 518372. Available at: https://www.hindawi.com/journals/crionm/2014/518372/. [Accessed on: 23 Mar 2020]. doi:10.1155/2014/518372.

18. Popadić M. Squamous cell carcinoma presenting as a giant cutaneous horn of the lower lip. Indian J Dermatol Venereol Leprol. 2014; 80(1): 74-6. doi: 10.4103/0378-6323.125469.

19. Blasco-Morente G, Arias-Santiago S, Pérez-López I, Aneiros-Fernández J. Cutaneous horn on the upper lip. Cuerno cutáneo en labio superior. Actas Dermosifiliogr. 2016; 107(5): 429. doi: 10.1016/j.ad.2015.05.018.

20. Curra M, Martins MAT, Hildebrand LC, Munerato MC, Sant'Ana-Filho M, Martins MD. Keratoacanthoma associated with cutaneous horn manifestation: case report and difficulty of diagnosis. Clin Lab Res Den. 2015; 21(4): 240-4. doi: 10.11606/issn.2357-8041.clrd.2015.123074.

21. Furquim CP, Cavalcanti LG, Piazzetta AP, et al. Cutaneous horn: a rare lesion on the lower lip. Oral Surg Oral Med Oral Pathol Oral Radiol. 2015; 120(2): e53. doi: 10.1016/j.oooo.2015.02.218.

22. Jesija JS, Kumar S, Paul R, Chacko R, Suryawanshi M. Cutaneous horn with underlying squamous cell carcinoma of lower lip. Oral Maxillofac Pathol J. 2016; 7(1): 726-9. doi: 10.5005/jp-journals-10037-1077.

23. Millán-Cayetano JF, García-Montero P, de Troya-Martín M. Squamous cell carcinoma presenting as bird beak-like cutaneous horn. Carcinoma espinocelular manifestado como cuerno cutáneo que simula un pico de pájaro. Med Clin (Barc). 2017; 149(9): e45. doi: 10.1016/j.medcli.2016.11.019.

24. Nair PA, Kota RK, Pilani AP. Pyogenic granuloma underlying cutaneous horn in a young boy. Indian Dermatol Online J. 2016; 7(2): 114-6. doi: 10.4103/2229-5178.178086.

25. Phulari RG, Rathore R, Talegaon TP, Shah A. Cutaneous horn: a mask to underlying malignancy. J Oral Maxillofac Pathol. 2018; 22(Suppl 1): S87-S90. doi:10.4103/jomfp.JOMFP_156_17.

26. Singh P, Nathani D, Ranjan S, Issar R. a giant cutaneous horn projecting from verrucous carcinoma of buccal mucosa: a rare case report. J Clin Diagn Res. 2017; 11(3): ZD04-ZD05. doi:10.7860/JCDR/2017/24657.9361.

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