## *Proceedings* del XII Congresso Nazionale e II Internazionale (Roma 10 -12 Ottobre 2013) Società Italiana di Patologia e Medicina Orale

Ho il piacere di presentare il volume che pubblica gli *abstract* dei *poster* che verranno esposti al XII Congresso Nazionale e II Internazionale della Società Italiana di Patologia e Medicina Orale (SIPMO).

La SIPMO si è sempre distinta nel panorama nazionale ed europeo per la solida preparazione scientifica e clinica dei suoi soci, per l'operosità e la costante attività di studio e di ricerca. Ho apprezzato molto la scelta dei Presidenti del Congresso e del Comitato Scientifico di far pubblicare in lingua inglese i contributi, e di indirizzare lo sforzo dei giovani odontoiatri italiani, cultori della patologia e medicina orale, su una sessione prettamente clinica, quella dei *case report/series*, con il palese orientamento di rafforzare le basi diagnostiche e terapeutiche nella molteplicità delle malattie odontostomatologiche.

In Italia, i Corsi di studio in Odontoiatria e Protesi Dentaria, in questi ultimi anni, hanno affrontato grandi sfide in termini di riordino didattico, riaffermando, tra l'altro, l'esigenza di un insegnamento unitario e non frammentato della Patologia Odontostomatologica, con obiettivi formativi importanti nell'ambito del sapere e del saper fare. In questa direzione si muove il presente volume, raccogliendo casi clinici rari e particolari delle scuole accademiche italiane, descrivendo protocolli diagnostici e terapeutici, il tutto con uno sguardo attento alle moderne tecniche diagnostiche. Alla SIPMO e, in particolare, ai Giovani appassionati, vanno i miei complimenti e l'augurio di buon e proficuo congresso.

> Antonella Polimeni Presidente del Collegio dei Docenti in Odontoiatria

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## Proceedings del XII Congresso Nazionale e II Internazionale (Roma 10 -12 Ottobre 2013) Società Italiana di Patologia e Medicina Orale (SIPMO)

"Perchè indirizzare le energie dei giovani soci SIPMO verso una sessione unica dedicata a case report/series?"

La scelta, condivisa tra il Consiglio Direttivo SIPMO e il Comitato Scientifico del Congresso, parte dalla convinzione che il giovane cultore della materia si avvicini a essa, in prima battuta, se affascinato dal rebus e relativa soluzione del caso clinico. Il *case report* singolo, o la raccolta di essi, costituisce un *primum movens* nell'approccio non solo al ragionamento, ma principalmente all'essere umano ammalato. Un *training* che il neo-laureato può poi portare con sé nel percorso libero-professionale, allorquando non sarà più, o non solo, frequentatore di un ambulatorio di medicina e patologia orale, quindi come quotidiano valore aggiunto della sua *clinical practice*.

Occuparsi di pazienti oggi significa applicare ad essi (per la diagnosi e la cura) le regole generali di un percorso deduttivo dettate dai testi e dalle ricerche, laddove esistenti, poste al top della gerarchia EBM. Infatti, è molto più a rischio di *bias* il processo opposto (dal particolare al generale), cioè il tentativo anacronistico di dettare regole valide per tutti sulla base solo di un singolo caso andato a buon fine. È questo, i soci anziani e giovani della nostra Società scientifica ben lo sanno e lo professano nella loro notevole e pregevole attività pubblicistica internazionale. È, altresì, irrinunciabile, un percorso ciclico che, dall'EBM e a seguito dell'applicazione delle sue stesse regole ai casi reali, porti, qualora s'incontrino casi non previsti dalle regole o che suscitino interrogativi e curiosità, a successivi step investigativi più metodologicamente corretti fino agli RCT e alla successiva integrazione e modifica dei protocolli.

Allo stesso tempo, sono note alcune critiche mosse all'EBM e sintetizzate in: la fiducia nell'empirismo; la limitata utilità dell'EBM per il singolo paziente; e i rischi di limitare l'autonomia della relazione medico-paziente. Questo porterebbe a considerare l'EBM uno strumento utile, ma con alcuni limiti quando essa è applicata per il *management* del singolo paziente. Il messaggio da portare a casa potrebbe essere quello di stare in equilibrio tra la teoria e la pratica dell'EBM. Dunque, ben vengano in sede congressuale i case *report/series* dei nostri giovani, l'approfondimento sul singolo caso clinico alla luce di quanto già noto.

Un sentito ringraziamento va al Comitato di Revisori che ha lavorato alacremente nei mesi precedenti la pubblicazione e alla Segreteria Tecnica della SIPMO per il costante supporto fornito. Con questo spirito, non resta che augurare agli interessati una buona e proficua lettura degli oltre 60 *abstract*, frutto della solida esperienza clinica dei nostri giovani e dei gruppi clinici italiani.

Palermo, 30/09/2013

Giuseppina Campisi Presidente SIPMO

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# Treatment of post-surgical scars of cheek's mucosa with Er, Cr: YSGG laser

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**Background**. Bulkiness and unsightly scarring are very common complaints after oral cancer surgical treatment. The scar of cheek can give complications such as: reduced mouth opening or interference with mastication. Both of these complications are particularly sources of discomfort for patient and may cause additional trauma to the treated mucosa<sup>1</sup>. There are several ways to reduce these complications: the most widely used is surgical excision of the scar but this treatment often requires more invasive surgery.

**Case report.** A 58-year-old male patient, treated surgically for a carcinoma *in situ* in the right cheek, reported to our oral medicine sector after this excision. The patient manifested the presence of some scars in the buccal side of the cheek with reduced mouth opening, reduced mobility of the tongue and interference of scars with mastication. A laser Er, Cr:YSGG was used to reduce the effects of scars by releasing adhesions<sup>2</sup>. It was used a power of 1.5 watts with a pulse duration of 740 ms, an energy of 50 mJ with a frequency of 30 pulses per second. It took three different interventions, although minor, in order to assess the actual improvement in symptoms. In none of the interventions was necessary to give stitches because haemostasis was achieved with the same laser. No drug treatment is needed, as no post-operative pain symptoms was complaining. The complete resolution of symptoms was achieved at the end of treatment. The mouth opening of the patient varied from 26 mm to 36 mm after the treatment.

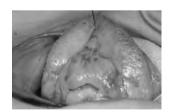
**Conclusion.** The use of laser Er,CR: YSGG may be a valid approach for the excision of scars of cheek's mucosa, as it is minimally invasive and offers many clinical advantages (minimal intra-operative bleeding, haemostasis, reduced times of healing).

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### **Case Report**

### Tongue ulcer in a case of X-linked diskeratosis congenita



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**Introduction.** Dyskeratosis Congenita (DKC) is characterized by immunodeficiency, bone marrow failure, anomalies of the skin, nails and mouth, and cancer predisposition. This syndrome affects predominantly males, with onset in childhood. All individuals with DKC have abnormally short telomeres for their age. The typical oral manifestation is leukoplakia, that may occur in any region of the mucosa.

**Case details.** A male child of 16 months, in apparent good health, was brought by his parents in the emergency room of the dental clinic of AO San Paolo in Milano, for a lesion of the back of the tongue.

The lesion had been present for about 4 months and its appearance was preceded by a condition that the pediatrician had identified as geographic tongue. The kid did not show any discomfort due to the lesion and he was eating regularly.

On examination, an ulcer of about 1 cm in diameter, at the center of the back of the tongue was detected. A cell sam-

ple was collected by brushing for cytology examination. Treatment with topical clobetasol and chlorhexidine gel was recommended, and complete blood count prescribed. The cytological diagnosis was: "sample consisting of granulocyte layer". A biopsy was then performed under general anesthesia, but again the pathological diagnosis was inconclusive. Genetic tests were performed at the Ospedale Pediatrico Gaslini, in Genova, and a diagnosis of X-linked dyskeratosis congenita was eventually proposed. The child under wentallogenic stem cell transplantation.

**Conclusions.** The oral lesion of this young patient was not a typical manifestation of the condition (leukoplakia), but a putative sign of haematological abnormalities.

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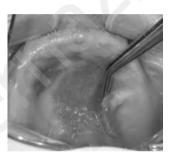
### **Case Report**

### An unusual lesion of the palate

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**Introduction.** Although clinical features are often an useful guide to diagnosis in oral medicine, we cannot rely just on them if we want to avoid frequent misdiagnosis.



**Case details.** A 60-year-old female patient was referred to the department of oral medicine at the University of Milan for a mass of the hard palate. The patient reported that the lesion had been present for about seven months, without causing any symptoms. Clinical examination revealed a subtle pedunculated lesion affecting the right side of the hard palate, with a major axis of about 15 mm, and a warty surface. The central area of the hard palate was affected by inflammatory papillary hyperplasia, probably due to the upper denture (denture stomatitis Newton's type III). The clinical diagnosis of the lesion was of oral papilloma or traumatic lesion. Because of the putative benign nature of the lesion, it was decided to proceed with the excision of the lesion. The histological diagnosis was: "pseudo-fibroma covered by epithelium with diffuse papillomatosis and lichenoid chronic inflammation" and the sample was "negative for HPV DNA".

**Conclusions.** In order to prevent medical errors, it is important to remember that many oral lesions may be present with unusual features, this requires an accurate phase of differential diagnosis. Having a suspected of lesion by HPV has meant that during the histo-pathological analysis have been carried out investigations in this direction. **References** 

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# Management of burning mouth syndrome: a clinical experience

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Burning mouth syndrome represents a puzzling condition both for the clinician and the patient. Professor van der Waal's description of this pathology highlights the application of several and different therapeutic protocols reported in the international literature, which often seem confusing and contradictory.

Salivary substitutes, tongue protectors, acupuncture, psychotherapy, topical benzodiazepines, omeophatic medicine, antidepressants, anticonvulsants, antipsychotics, ormonal substitutes, complementary vitamin diet, even electroconvulsive therapy have been proposed as effective therapeutic protocols, but none of them has proved to offer a satisfactory solution to such a problem.

### **Case Series**

This report explains how we tried to treat burning mouth patients by applying a strict and homogenous protocol: our care pathway ranged from the treatment of peripheral neuropathy to the possible management of the pain central pathways, whose mechanisms are still to be investigated and explained. Pending a better understanding of the underlying pathogenesis of this mysterious illness, the aim of this protocol was to decrease the sensation of burning pain in order to make it more tolerable to patients.

In detail we report the rewarding outcomes of our clinical experience which consisted in applying topical capsaicin through mouth rinses to most burning mouth patients. Chronic application of topical capsaicin regulates the expression of trpv-1 vanilloid receptors, and helps to determine a decrease in oral burning sensations, a process which is known as tachyphylaxis.

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### **Case Report**

# Mithocondrial DNA analysis in a case of multiple oral carcinomas following allogenic

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## hematopoietic stem cell transplantation

### A. Bernardi, D. Gabusi, D. Servidio, F. Diano, D.B. Gissi

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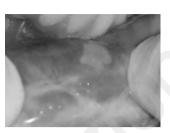
The development of Oral Squamous Cell Carcinoma (OSCC) is a potential long-term complication after hematopoietic stem cell transplantation (HSCT). Chronic Graft versus Host Disease (cGvHD), and prolonged immunosuppressive therapy have been considered as the potential risk factors for the malignant transformation of the oral mucosa. Further, a higher incidence of multifocal OSCCs has been recently demonstrated in HSCT patients as compared to non-HSCT. In the 2011 we have reported a case report of a young female patient treated with HSCT for beta thalassemia major 17 years before, who developed two synchronous OSCC1. At the first presentation, the patient showed bilateral simil-lichenoid lesions histologically compatible with cGvHD and the presence of a tumor mass (T3N0M0) that was surgically removed. A second tumor (T2N0M0), arose few months after the primary and at a distance of more than 4 cm. Considering the spatial/temporal discrepancy between the two tumors, it was quite difficult at that time to distinguish whether or not the second tumor was related to index tumor. In the present report we have retrospectively reconsidered the case, by applying the analysis of mithocondrial DNA D-loop region to both neoplasia in order to evaluate the clonal relationship between the two neoplastic manifestations<sup>2</sup>; mtDNA analysis showed a clonal relationship between two oral carcinomas and permitted to classify the second manifestation as Local Recurrence. The result of the molecular analysis highlighted the aggressive behaviour of the primary tumor and the possibility that a second tumor even arisen more than 2 cm distant can share the same clonal origin of the primary. Thus, mtDNA analysis may be useful to differentiate Local Recurrence from Second Primary Tumor when discrepancies arise between spatial and temporal criteria and to better understand the mechanisms of carcinogenesis in HSCT patients.

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# Behçet's disease in an adolescent patient

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Behcet's disease (BD) is a chronic and multisystematic inflammatory disorder characterized by oral and genital mucous "aphthous-like" recurrent ulcers, uveitis, and skin lesions. Oral ulcerations are present in more than 90% of patients being often the first clinical manifestation. Although the actual etiology is still unclear, BD symptoms are considered to be based on the correlation between the genetic factors (HLA B51) and the triggering extrinsic factors (i.e. oral unhygienic condition). Pediatric onset BD is an uncommon disease that presents before the age of 16 years and has a distinct clinical presentation of recurrent abdominal pain and episodes of fever. We report an interesting case of a 14 years old Caucasian male referring to our Unit for 3-years history of recurrent oral lesions. From a systemic point of view, the patient reported a deficiency of glucose-6-phosphate dehydrogenase, abdominal pain and meteorism. The intra-oral examination revealed "aphthous-like" ulcers on the tip of the tongue and lower labial mucosa and scarring areas in the lower labial mucosa. Furthermore, the lower lip appears swollen and with an increased consistency. Due to the age of the patient and the clinical signs and symptoms we consider as potential diagnosis a gastrointestinal disease as coeliac disease and Chron disease. However, serological screening for celiac disease was negative as well as faecal calprotectin; while, deficiencies of iron and folate and increased VES were showed. After 1 month the patient presented with a genital "aphthous-like" ulcer. So a BD was suspected and the research of HLA B51 was positive. The patient was referred to the ophthalmologist and dermatologist who excluded eyes and skin involvement. This case underlines that the presence of complex oral aphthous episodes also in paediatric patients should prompt the clinician to suspect the presence of an underlying systemic disease, crucial step for its early diagnosis and treatment. References

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# Low level laser therapy (LLLT) as adjuvant in the management of drug induced gingival hyperplasia: a case report



**Case Report** 

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Gingival hyperplasia can be due to various causes; when drug induced it is an adverse result caused by long term anti hypertensive, anticonvulsant or immunosuppressive therapies. Treatment is complicated by the multiple factors involved, like the impossibility of a drug change and the difficulty patients encounter in maintaining an acceptable level of periodontal health due to an insufficient control of bacterial plaque.

This case was treated with SRP and antibiotic therapy, but a high level of tissue inflammation persisted. It was therefore treated with LLLT sessions only, by means of a specifically designed for photobiomodulation diode laser, dual wavelength (904/650nm). A good periodontal health was obtained, and this allowed a correct home oral health care, so as to maintain the result that was obtained.

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### **Case Report**

# Pemphigus vulgaris of the oral cavity in a patient with coeliac disease: coincidence or relationship?



### F. Carroccio<sup>1</sup>, V. Rodolico<sup>2</sup>, A. Padovano Di Leva<sup>3</sup>, G. Campisi<sup>1</sup>, D. Compilato<sup>1</sup>

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**Background.** CD is a lifelong T cell-mediated enteropathy triggered by the ingestion of gluten in genetically susceptible subjects. The disease has a heterogeneous range of clinical presentations. Among atypical aspects of CD some affections interest the oral cavity. Pemphigus vulgaris (PV), can be associated with CD-auto-antibodies and a clinical improvement of the PV after the gluten-free diet has been reported.

**Case report.** A 43-year-old Caucasian female referred to our Unit for a 1-year history of recurrent oral lesions treated with anti-mycosis drugs without any clinical improvement. The patient was in good general health, although she had meteorism and dyspepsia. The intra-oral examination revealed bullous lesions on the lower labial mucosa, on the dorsum of the tongue, on the left buccal mucosa and on the soft palate. These clinical features led us to consider as potential diagnosis an immune-mediated muco-cutaneous bullous diseases. Red blood cells count, iron, folate and vitamin B12 were normal. The auto-antibodies specific for the immune-mediated bullous diseases showed a positivity for anti-skin and anti-desmoglein 3. The incisional biopsy of the lingual bulla confirmed the diagnosis of pemphigus. The patient started the treatment with deflazacort tablets and clobetasol-based ointment for topical application on oral lesions, with poor clinical results. Despite the patient refer us to suffer only of few gastrointestinal symptoms we decided to investigate for CD. Anti-transglut-aminase antibodies were negatives; however, the CD-predisposing haplotype DQ8 was present. A duodenal biopsy confirmed the diagnosis of CD. The patient started the gluten-free diet with clinical improvement of the oral lesions.

**Conclusions.** Although the association between CD and phemphigus has not been widely studied, we think that serological and genetic study for CD should be considered at least in complicated patients with oral and/or systemic immune-mediated diseases.

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### **Case Report**

# Oral cancer and mucosal trauma: a case series

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Oral squamous cell carcinoma (OSCC) is the sixth most common cancer worldwide. OSCC still has high mortality rates due to difficulties in detection at an early stage. The 5-year survival rate is lower than other major cancers. OS-CC etiopathogenesis is mainly related to lifestyle, with major risk factors being tobacco smoking and heavy alcohol consumption. Other risk factors include family history of cancer, dietary habits, HPV infection, poor oral hygiene, chronic local trauma and genetic predisposition. The link between inflammation and cancer was noticed about 150 years ago by Virchow. A retrospective case record review was conducted of all patients who were seen at the Dept. of Oral Diagnosis, University Hospital San Paolo of Milan from January 2007 to June 2013. A total of 14 individuals were seen looking for OSCC and mucosal trauma; the mean age in the cohort was 54,7 years, with 5 individuals younger than 50 years; 6 were men and 8 were women. Only 5 patients were current smokers. The association of chronic oral mucosal trauma with local inflammation induced by fractured and/or malpositioned teeth or prosthetic crowns was easy to detect both through medical history and clinically.

The carcinogenic role of chronic trauma caused by fractured crowns or dental malocclusion is still controversial. In this report the authors describe a series of OSCC cases in which there was an association between younger age and local chronic trauma in non smokers and with no alcohol consumption reported; in this cohort the mucosal trauma could represent a major risk factor for OSCC. Evidence in the literature remains inadequate to allow final conclusions on whether local chronic trauma is a true risk factor for the development of OSCC. However, it could be assumed that general inflammatory mechanisms linked to carcinogenesis play a role in the oral cavity, and dentists have an important function in prevention of OSCC by eliminating chronic trauma of the oral mucosa caused by teeth and prosthetic appliances.

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### Case Series

# Numb chin syndrome

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Introduction. Numb chin syndrome is a sensory neuropathy characterized by

numbness, hypoesthesia or paraesthesia and, rarely, pain in the distribution of the mental nerve. Dental causes, specially iatrogenic ones (e.g., oral surgery), are by far the most common; if not related to those, this complaint is considered a "red flag" symptom of a distant malignant neoplasm. Other possible aetiological factors are: primary tumours, trauma or systemic diseases.

**Case Description**. We describe cases in which the presence of numbness and paraesthesia in the distribution of mental nerve was the most relevant symptom linked to odontogenic infective causes. A 45-year-old woman (# I) reported a continuous not evocated pain in the left molar region and hypoesthesia of mental nerve; ortopantomography revealed a lytic lesion involving the mandibular canal suggesting an odontogenic infection in relation to 3.6. A 59-year-old man (# II) presented a sudden violent pain in the right premolar region; after few days, hypoesthesia of the mental nerve also appeared. The CT imaging showed an osteolytic area extending to the inferior mandibular edge. The clinical and radiologic examinations confirmed the presence of a large osteolytic infective lesion originated from 4.5. A 63-year-old man (# III) was referred to our department for a violent diffuse pain in the right molar and premolar region; hypoesthesia of the mental nerve soon followed. In CT imaging, an irregular osteolytic lesion was present, with focal erosions of mandibular bone, compatible with a sequestrum originated from chronic apical periodontitis of 4.6 and 4.4.

**Conclusion.** Numbness of the area of distribution of the mental nerve could be related to odontogenic infective problems also in case of atypical presentation, with no clear relationship with dental foci at the first clinical examination. A careful attempt to find this relationship should always be made before thinking to neoplastic diseases or systemic causes.

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# Turner syndrome with dental structural abnormalities: histological and morphostructural features by confocal laser microscopy



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Turner syndrome is a genetic disorder characterized by the total or partial absence of the X chromosome. There are different types of karyotypes although the most frequent is the 45X (57%). Mosaicisms such as 46 XX/45X, 45X/46XX/47XXX (29%), the presence of the isochromosome X or the chromosome X-ring (14%) may occur.

The main clinical features are: short stature, gonadal dysfunction, congenital heart disease, renal and skeletal anomalies, endocrine disorders and hypoplasia of the fingernails and toenails. Craniofacial features are due to retarded development of the cranialfacial complex with reduced growth of the skull base, hypertelorism, retrognathic profile, malocclusion Class II (60%), hypoplasia of the maxilla, bilateral cross-bite (9%) with or without cleft palate, anterior open bite (17%). The teeth are altered in shape and structure.

A patient AA 9 years old, suffering from Turner syndrome karyotype 45Xq; the Rx OPT has highlighted the presence of agenesis of 2.2 and a mesiodens in the region between the upper central incisors with the inclusion of 1.1. We proceeded to the extraction of the pre-orthodontic mesiodens and subsequent orthodontic treatment for the correction of Class II malocclusion.

The mesiodens, fixed in formalin, decalcified, and half was embedded in paraffin and stained with hematoxylin-eosin and half including resin-ground sections. Histological examination to the confocal laser scanning microscopy showed:

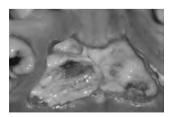
- Alternation of zones of hypo and over calcification;
- Inhomogeneous structure of dentin and abnormalities of the dentinal tubules;
- Reduced enamel thickness and irregular arrangement of the striae of Retzius;
- The pulp and root systems are characterized by the presence of foci of ectomesenchymal dentinogenetic multifocal differentiation that cause irregularities in the shape and structure of the root by an irregular, neoplastical-like growth (odontoma-like).

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### **Case Report**

# A strange "butterly-like" ulcer of the palate. May be it the results of an underlying trombotic event in a predisposed patient?



### D. Compilato<sup>1</sup>, G. Colella<sup>2</sup>, L. Lo Russo<sup>3</sup>, L. Lo Muzio<sup>3</sup>, G. Campisi<sup>1</sup>

<sup>1</sup>Department of Surgical, Oncological and Oral Sciences, Sector of Oral Medicine "V. Margiotta", University of Palermo, Italy; <sup>2</sup>Department of Head and Neck Surgery, II University of Naples, Italy; <sup>3</sup>Department of Surgical Sciences, University of Foggia, Italy A 48-year-old female was referred to us from the emergency ward for a 2 weeks history of erosive/ulcerative lesion of the palate associated with pain, dysphagia, fever and weight loss. Her recent medical history revealed a previous surgical treatment for a breast cancer treated. Although the tumour did not showed positivity to the estrogen receptors, the patient's oncologist recommended to start a pharmacological treatment with Tamoxifen that she keeps on for about 2 months. The oral examination showed a "butterfly-like" lesion symmetric with respect to the midline of the hard palate characterized by two areas of ulceration with a fibrinous yellowish floor and by erosive areas extending anteriorly. The patient underwent oral swabs to research mycotic and/or bacterial infections and was invited to use a chlorhexidine 0.12% without alcohol-base mouthwash and to stop smoking. After 1 week the lesion appeared as a single "butterfly-like" ulcer with central necrosis. Due to the rapid progression and the destroying aspect an incisional biopsy was performed revealing an unspecific chronic inflammatory infiltrate. No microorganisms and lymphomatous infiltrate were seen. All routine hematological, biochemical and microbiological investigations were normal. After another week the lesion worsened appearing deeper. The patient underwent a contrast enhanced CT of the maxilla-facial region that excluded bone perforation. Since the treatment with tamoxifen is associated with an increased risk of tromboembolic event, we hypothesize that the oral lesion may be the result of a ischemic necrosis. A coagulation screening was performed revealing positivity for the lupus anticoagulants antibodies (LAC). The serological tests for autoimmunity (ANA-ENA) were negative. According with the oncologist the patient suspended the treatment with tamoxifen, and the lesions was treated with gentle debridement and topical anti-septic, the wound healed by secondary intention in 4 months.

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**Case Report** 

# An unusual lingual lesion in a little patient

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**Introduction.** The most frequent oral lesions in children have traumatic or infectious origin. Nonetheless, it is important to consider in clinical differential diagnosis also unusual oral conditions, such as lymphangioma and neurofibroma, since early recognition and surgical excision of these tumors are essential for an optimal patient management.

**Case Details.** A 3-year-old female child was referred to our center with an asymptomatic lesion on the dorsum of tongue, increasing in size. A difficult clinical examination allowed to note a well-defined lingual swelling (1 cm of diameter), with numerous whitish papillary and vesicle-like projections. It was soft, nontender and pebbly. No restriction of functions of tongue was observed.

On the basis of history and clinical features, the following diagnostic hypotheses were proposed: lymphangioma, neurofibroma, papilloma, a lesion of salivary gland, although any other diagnosis could not be excluded.

Because of the lack of collaboration, due to the young age, it was decided to obtain a sample of the lesion under general anesthesia, for an extemporaneous histological examination, to elucidate the nature of lesion. When the benign nature of the lesion was confirmed, complete surgical excision of the lesion was performed. The final diagnosis of the lesion was of lymphangioma.

**Conclusions.** Lymphangioma is among the unusual conditions that an Oral Medicine specialist should consider in presence of a localized growing mass in pediatric patient. Lymphangioma is a benign hamartomatous hyperplasia of lymphatic vessels. Seventy-five % of cases are localized at the head and neck area. The most common location in the mouth is the dorsum of tongue, followed by lips, buccal mucosa, soft palate, and floor of the mouth. About half of the lesions are detected at birth and around 90% of them develop by 2 years of age.

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# White tongue and straight hair in a patient with chronic hepatitis C: a case report and review of the literature

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8.0

Introduction. The antiviral treatment for chronic HCV with Interferon alfa or PEG Interferon alfa combined with Ribavirin leads to many skin side effects like alopecia, vasculitis, cutaneous necrosis,

eczema, lichenoid lesions, dry skin, pruritus, psoriasis, vitiligo. During this combination therapy tongue and gums hyperpigmentation both in dark skinned as well as Caucasian have been also reported. There are also two case reports of a complete and acquired straightening of hair due to the administration of the same drugs.

**Case Presentation**. We describe the first case of a patient with a chronic HCV infection who developed a white and spongy tongue and also an acquired straightening of hair, during combination therapy with PEG Interferon alfa and Ribavirin.

**Discussion**. In our case, the pathogenetic mechanisms of this white tongue associated with hair straightening are unclear. The exclusion of the most important diseases as responsible for the phenomenon, the absence of other drugs associated, and mainly the progressive oral lesions remission one month after the end of therapy, and their complete remission three months of the end of the last drug administration, lead us to the association between PEG-INF-2a plus Ribavirin with this abnormalities.

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### Case Report

# The photodynamical treatment of oral verrucous proliferative leukoplakia. Case report at 18 months follow up



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Verrucous Proliferative Leukoplakia is a rare aggressive form of leukoplakia characterized by high tendency of malignant transformation. It is often seen in females over 60yrs often associated to tobacco. Its histological aspect may vary from simple hyperkeratosis to various degrees of dysplasia sometimes mimicking Verrucous Carcinoma. The treatment is surgical but the VPLs show high recurrence rates independently by surgical approach. Recently a non surgical and non invasive treatment of these lesions was suggested, based on photochemical light induced reaction (Photodynamical Therapy/PDT), permitting safe and complete removal of pathologic tissue respecting the nearby healthy one. A case of PDT treatment of a VPL is presented with follow up at 18 months. The patient is a 84yrs non smoker female, presenting a wide asymptomatic white verrucous patch on the right cheek. An incisional scalpel biopsy was early performed to exclude any sign of dysplasia; the diagnosis was of VPL. The PDT was performed by subministration of the photosensitizer, 5-Aminolaevulinic Acid (5-ALA) in 20% oral gel, 90' before laser exposure. The laser adopted was a 635nm diode (Lambda Spa, Vicenza, Italy). The irradiation was performed twice a week, each second day. The protocol previewed a maximum of 6 applications, less in case of clinical disappearance of the lesion. Laser power was 100mW, in CW with scanning no contact movement and total energy of 100mJ. The exposure time was 1000 seconds, in 5 cycles of 3', with intervals of 3' to permit the synthesis of the Protoporphirin IX, and a final cycle of 100 seconds. After 5 applications the lesion totally disappeared, without side effects scarring or pain both in immediate and follow up period. Controls were scheduled each 3 months; at 18 months no recurrence was observed. In this case the PDT resulted very helpful in the management of VPL. More studies and cases are needed to evaluate its real efficacy for the treatment of these lesions.

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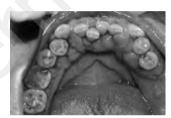
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### Case Report

# Drug-induced gingival hyperplasia, treatment with diode laser

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**Background.** Gingival overgrowth/hyperplasia can be attributed to several causes, but drug-induced gingival overgrowth/hyperplasia arises secondarily to prolonged use of antihypertensive drugs, anticonvulsants and immunosuppressants. A gingivectomy performed with a laser is a short, easy procedure that produces an immediately dramatic effect. Compared to a scalpel gingivectomy, there is excellent hemostasis, which improves visualization, requires less need for periodontal packing, and results in minimal postoperative discomfort<sup>1,2</sup>.

**Case report.** A 70-old-male patient treated with antihypertensive therapy with "Calcium channel blockers" for about 1 year (amlodipine). He had severe gingival overgrowth, hyperemia, easy bleeding, pain and masticatory function compromised. Changing of antihypertensive drug and the execution of a professional treatment of oral hygiene did not allow the resolution of hyperplasia. A diode 810 laser was used to excise the gingival overgrowth. The used parameter was 2W in continuous wave with a activated fiber of 400 mm. This treatment permitted the resolution of the case without any complication.

**Conclusion.** The use of diode laser in the present case proved to be effective in the removal of large amounts of hyperplasic gingival tissue and resulted in fast heeling and mild discomfort.

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### **Case Report**

# Mandibular brown tumor as the first manifestation of primary hyperparathyroidism: a case report



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Background. Brown tumors are erosive bony lesions caused by rapid osteoclastic activity and peritrabecular fibrosis

due to hyperparathyroidism, resulting in a local destructive phenomenon. These lesions are non-neoplastic and they appear as a mass with partly cystic and partly solid areas.

Clinically, they are slow-growing lesions that can be locally destructive resulting in variety of symptoms such as significant bone swelling, pain and pathological fracture; they can be mistaken for a neoplasm.

The classical "brown tumor" is commonly seen in ends of long bones, the pelvis and ribs. Facial involvement is rare and, when present, usually involves the mandible.

Parathyroid adenoma (and most rarely adenocarcinoma) is the commonest cause of primary hyperparathyroidism and usually presents with symptoms/signs of hypercalcaemia.

**Case Report.** We report a case of 71-year-old male with a mandibular brown mass in absence of symptoms. The lesion was expansive and osteolytic, with invasion of the floor of the mouth. Histology revealed the presence of an intrabone giant cell lesion. Blood tests demonstrated elevations in parathyroid hormone (PTH) concentrations. This suggested the diagnosis of hyperparathyroidism initially manifesting as a brown tumor of the mandible. Posterior explorations confirmed the existence of a parathyroid adenocarcinoma as the cause of the condition.

**Conclusion.** The aim is to alert the clinicians to include this entity although extremely rare in the differential diagnosis of red/brown oral masses.

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**Case Report** 

# Loss of p53 protein expression in leukoplakias may reveal the presence of an oral squamous cell carcinoma: a case series analysis



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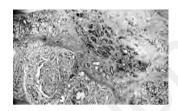
Mutations in the TP53 gene leading to loss of function are the most common genetic damages found in human cancers and Oral Squamous Cell Carcinoma (OSCC). TP53 mutations may result in a over-production of p53 inactive proteins which accumulate in the epithelium either due to blocking by another protein or due to partial degradation, or instead in an abrogation of p53 wild function by epigenetic mechanisms. Immunoistochemical analysis of p53 is a simple method used for the detection of p53 protein, and its evaluation has been widely recommended in oral leukoplakias to a better understanding of the potential risk of malignancy. In the present series report we describe four patients referred to our Department following the appearance of an asymptomatic lesion of the tongue. Patients underwent an incisional biopsy for histology and immunoistochemical analysis of p53 protein. In all cases, the lesions were clinically and histologically classified as oral leucoplakias with sign of mild dysplasia. Immunoistochemical analysis showed in all cases a negative staining of p53 protein. All four lesions were subsequently surgically removed and histological analysis of the whole lesion showed in all cases the presence of a microinvasive carcinomas. Thus, in all four cases the histological conclusions from an incisional biopsy as a "snapshot" of the whole lesion have understimate the true nature of the whole lesion. Predictive value of p53 negative staining is not well described in literature while p53 over-expression has been proposed in numerous studies as a reliable marker associated to oral carcinogenesis, although most of the studies utilized antibodies that cannot discriminate in the single case wild or mutated form<sup>1</sup>. Instead, the presence of p53 negative staining could be considered a specific value representative of an arrest of p53 protein synthesis and seems to be a useful marker for early diagnosis of OSCC also in absence of histological recognizable signs.

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**Case Series** 

# Salivary gland tumors in patients with necrotizing sialometaplasia: a case series



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Necrotizing sialometaplasia (NS) is a benign, reactive, necrotizing lesion that may involve minor salivary glands, and, more rarely, major salivary glands and mucoserous glands of the upper respiratory tract. To date, about 220 patients have been reported in literature, harboring NS in the parotid gland, submandibular gland, incisive canal, lower lip, tongue, floor of mouth, retromolar area, mucoserous glands of the nasal cavity, maxilla, larynx and trachea<sup>1</sup>. The majority of cases of NS, however, involve the hard and/or soft palate. Although benign and self-limiting, NS is significant because it might be clinically and histologically mistaken for malignancy<sup>2</sup>. Furthermore, NS may accompany tumors of minor and major salivary glands<sup>3</sup>. In this series we reported 6 patients affected by NS associated with minor and major salivary gland tumours. 2 patients presented NS of the minor salivary glands of the palate, and had associated neoplasms (pleomorphic adenoma and adenoid cystic carcinoma) at the same site. Four patients had NS of the parotid gland associated with epithelial-myoepithelial carcinoma, adenoid cystic carcinoma, Warthin's tumor and oncocytoma. Necrotizing sialometaplasia is a self-limited disease with spontaneous resolution in most cases; even if regeneration of the affected ducts and acini is usually incomplete, the healing process includes repair of both the ulceration and regeneration of the damaged salivary tissue. During this regenerative process, the presence of ductal and acinar metaplasia, prominent nuclei, frequent mitotic figures and necrosis may lead to an incorrect diagnosis of malignancy. Although infrequently, NS has in fact been reported to obscure an underlying neoplasm, thus resulting in delays in referral or treatment. This further stresses the importance of performing an appropriate biopsy and carefully monitoring patients with NS of the minor salivary glands.

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**Case Report** 

# Familial Tumoral Calcinosis with dentoalveolar anomalies: clinico-pathological findings and Confocal Laser Scanning Microscopy of hard and soft tissues lesions



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**Introduction.** We report on the clinic-pathological features of Familial Tumoral Calcinosis (FTC), a rare disease of early childhood and adulthood, caused by mutations in fibroblast growth factor 23 and GalNAc transferase 3. It is a bone metabolism disorder with abnormal phosphate and calcium (calcinosis) deposits around the joints, in visceral and soft tissues.

**Case Presentation.** A 17-year-old girl complaining for long-standing night leg pain, resistant to FANS therapy, had been diagnosed with osteogenesis imperfecta and was therefore undergoing bisphosphonates therapy. She was referred to our Dental Clinic for diffuse dental anomalies (short and irregular roots, dental chambers anomalies, teeth inclusions) and maxillary hypoplasia and underwent combined surgical and orthodontic treatment. The surgical samples were used for conventional and Confocal Laser Scanning Microscopic (Nikon E-600) (CLSM) examination.

**Results and conclusions.** Microscopically several metaplastic micro and macro-calcificationin soft and periodontal tissue location were detected, along with a typical islands of homogenous, non tubular, dentino-osteoid calcified structures in dentinal tissues. Also, dentinal dysplasia with osteoid-like material, without incremental lines but with strong basophilia, intermingled with remnants of mature mucous connective tissue, was demonstrated. The diagnosis of FTC was confirmed by genetic analysis. CLSM helps to demonstrate distinct odontoblast and osteoblast anomalies in FTC that lead to the accumulation of atypical calcified tissues, responsible for the several clinical signs detected in the patient and formerly attributed to osteogenesis imperfecta.

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### **Case Report**

# Misunderstood oral early syphilis: a meaningful case report

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Syphilis has been increasing in incidence during the last few years and its early presentation may be the oral cavity (40-75%), so it is increasingly becoming a differential diagnosis in routine oral pathology.

A 29-year-old female with no major medical conditions was visited for painful erythematous areas on the cheeks and mandibular vestibules, painless a specific ulceration on right border and dorsum of the tongue and marginal painful gingivitis, recently treated with scaling and root planing, without purulent aspect.

There were neither regional lymphadenopathy nor coutaneous manifestation.

The patient, HIV-negative, reported usual oral sex.

Differential diagnosis were considered: erythema multiforme (caused by 5-day therapy with paracetamol and undefinited FANS after additive mammoplasty), herpetic gingivostomatitis, recurrent aphthous stomatitis, Behçet's syndrome and aspecific ulcerations.

The following investigations were performed: tongue's right border punch biopsy (5mm diameter) for histological examination, cheek's left biopsy for direct immunofluorescency (DIF) and PCR for DNA investigation of bacteria and virus.

Histologic examination revealed endothelial cellular proliferation within small arteries and arterioles and an inflammatory infiltrate with mature plasma cells, in perivascular distribution or as a band-like infiltrate in the lamina propria. In addition, molecular testing (PCR) revealed the presence of numerous intraephitelial treponemes, while DIF proved to be non-diagnostic.

Several clinical tests have ruled out other infected sites, and the patient was treated with a single intramuscular injection of benzathine penicillin G (2.4 million UI). The lesion completely resolved during a 4-week period.

A missed diagnosis can often lead to serious complications for the patient and dental care workers (DCWs) because, unlike other sexually transmitted diseases, syphilis is easily transmissible in early oral presentation.

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# Conservative surgical treatment in the management of BRONJ: a case series of 129 consecutive cases



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Aim. Aim of this case series was to evaluate the out comes of conservative surgical treatment of bisphosphonate-related osteonecrosis of the jaw (BRONJ).

**Materials.** 119 subjects affected by 129 BRONJ and surgically treated in our unit were enrolled. Surgical treatment (sequestrectomy, soft tissue debridement, and bone curettage with no or limited extension) was delivered only to sites that did not respond to medical treatment. Age, gender, underlying disease, tabagism, comorbidity were also analysed. Main outcome was improvement of clinical stage and disease resolution (passage to stage 0). Subjects were followed for at least 6 months.

**Results**. *Sample*: oncologic diagnoses was made in most of the cases (77%). Breast cancer was the primary pathology (36 subjects, 30%), followed by multiple myeloma (26 subjects). Sixty-three percent of the lesions were located in the mandible (81 subjects). The main event leading to BRONJ was tooth extraction (76 subjects, 59%). A reasonable explanation for BRONJ was detected in 82%. The most frequent stage of BRONJ was stage II (77 subject, 60%), whereas stage I (26 subject, 20%) and stage III (26 subject, 20%) were less common.

*Effect of surgical treatment*: during follow-up, one subject died. 84% of subjects showed improvement after surgery, 15% showed no modification, and 1% exhibited a worsening of their clinical condition. Stratification indicated improvement for 100% stage I (and therefore total disease resolution), 87% stage II and 52% stage III. Disease resolution was noted in the 75% of Stage II and 40% of Stage III. Interestingly, a learning surgical curve was effect noted for disease resolution.

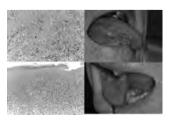
**Conclusion.** Our data suggest that conservative surgery may be the possible treatment of choice in stage I and II. Conversely, stage III subjects might be treated with either a resective surgical approach or clinical monitoring according to the clinical medical scenario of the patient.

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### **Case Report**

# A single biopsy in patients with multiple pre-malignant lesions may lead to an inappropriate treatment: a report of two cases



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Oral lichen planus and leukoplakia both manifest as white/red lesion, but are associated with different rates of malignant transformation. Frequently, patients affected by OLP or leukoplakia show multifocal lesions, and frequently it's a routine practice to perform a single biopsy in the most representative area. In a recent study, patients presenting multiple lesions underwent surgical biopsy of all lesions. It emerged that a single patient may have either lesions with histological features of OLP and lesions clinically and histologically diagnosed as leukoplakia. It was concluded that patients with multiple white/red lesions should undergo multiple biopsies and the prognosis and management should be addressed to each single lesion. In fact, performing a single diagnostic biopsy bears a significant risk of mis-estimating the diagnosis and prognosis. In this study, we report two cases of two patients (one male and one female aged both 78 years) with multiple oral white/red lesions with at least one lesion diagnosed as OLP while at least another as leukoplakia. In the first patient leukoplasic lesion presented moderate dysplasia and was surgically removed. The histological examination of the entire lesion showed an infiltrating squamous carcinoma. In the second patient, the presence of a wide leukoplakia suggested a 3-month regular follow up. Two years after the diagnosis a second biopsy was scheduled due to clinical changing of leukoplakia. Histological evidence of OSCC was found. These two cases can be considered as an example of what can happen when a single biopsy is performed in patients with multiple lesions. If by chance the lesion diagnosed as OLP had been the only one analyzed, longer follow up intervals would have been applied, leading to a late OSCC diagnosis. In conclusion, in patients with multifocal oral white/red lesions, the prognosis and therapy must be differentiated according to each lesion.

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### **Case Series**

# LLLT in combination with nonsurgical periodontal therapy in patients with gingival oral lichen planus: a pilot study



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Introduction. Oral lichen planus (OLP) is a chronic inflammatory disease that can be painful, mainly in the atrophic and erosive forms. Numerous drugs have been studied but most treatments are empirical. However, to date, the most commonly employed and useful agents for the treatment of OLP are topical corticosteroids. The study objective was to detail the clinical effectiveness of low-level laser therapy (LLLT) in combination with a non surgical periodontal protocol for the management of OLP unresponsive to standard topical therapy.

Methods. The authors studied a prospective cohort of 6 patients affected by OLP, who received biostimulation with a 980-nm gallium-aluminum-arsenide (GaAIAs) diode laser in combination with professional oral hygiene therapy. For all patients were evaluated parameters of severity of injury (SEVERITY SCORE) and reported symptoms (VAS) before and after the proposed protocol.

**Results.** Patients showed a decrease in the severity of the clinical appearance in the first two weeks of treatment. The statistical analysis did not show a significant reduction of clinical score; 5 patients reported symptoms' improvement. The statistical comparison of symptoms showed a significant reduction in VAS (P<0.05).

Discussion. To date, few studies have confirmed the efficacy of LLLT to decrease the symptoms of OLP gingival lesions. Our study showed that LLLT is effective in faster reducing pain if associated with non-surgical periodontal therapy. This combined therapy, in the protocols of previous scientific papers, was not practiced and treatment time was extensive and took over a month, while in our work, we obtained significant results within only two weeks.

Conclusion. The LLLT, in association with oral hygiene treatment, might be a feasible alternative in OLP patients with desquamative gingivitis, because it is easy and simple to perform, free of side effects, acting in a short period in reducing reported symptoms.

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### **Case Series**

# Oral hygiene in patients with gingival pemphigus vulgaris: a case series

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**Introduction**. Recent international literature showed impairment of periodontal status in patients with pemphigus vulgaris (PV) compared to healthy patients, this possible contributing to the development and / or progression of periodontal disease<sup>1, 2</sup>.

The intention of this pilot study was to evaluate prospectively the clinical efficiency of an oral hygiene procedure in patients affected by gingival PV.

**Methods**. Patients received oral hygiene instruction followed by non-surgical periodontal therapy including oral hygiene instructions in a 2-week cohort study. Clinical outcome variables were recorded at baseline and 4, 12 and 16 weeks after intervention and included full mouth plaque (FMPS) and bleeding (FMBS) scores, oral PV clinical score (to record the severity of the disease) and patient related outcomes (visual analogue score of pain).

**Results.** A total of 5 patients were recruited. The statistical tests used for data processing were the Wilcoxon Test. During the follow-up period, a statistical significant reduction was observed for FMBS (P<0.05) and clinical severity (P<0.05).

**Discussion**. To date there are no studies designed to show whether the oral hygiene therapy can be accepted as adjuvant treatment in cases of PV with gingival location. A reduction of gingival inflammation, due to the reduction of bacterial aggression, could positively affect gingival response.

**Conclusion.** A protocol of causal therapy does not seem to aggravate the course of bullous-erosive lesions in patients with desquamative gingivitis by PV. The proposed protocol reduces gingival bleeding, but does not affect other periodontal indices over the horizon; it also improves physical signs of autoimmune disease in the gingiva, but does not affect significantly the patients' symptoms.

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### **Case Report**

# Osteonecrosis of the jaw related to everolimus and bisphosphonate: a unique case report?

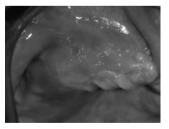
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Osteonecrosis of the jaw (ONJ) is a rare but serious lesion of the jaw, characterized mainly by exposed necrotic bone; it is related to various drugs, usually used for treating patients with advanced malignancies. Drugs implicated in ONJ are: nitrogen-containing bisphosphonates (NBPs), denosumab, anti-angiogenic drugs (e.g bevacizumab, sunitinib, sorafenib) and the selective mammalian target of rapamycin mTOR, everolimus. Previous data regarding the combining of NBPs with antiangiogenic agents conflict with some reports (indicating a similar risk of ONJ compared with the use of NBPs alone<sup>1</sup>); other reports show significantly higher rates (18% vs 1% with NBPs alone) of the incidence of ONJ<sup>2</sup>. The mTOR is a serine/threonine kinase, a component of a complex signaling pathway, involved in cell growth and metabolism, reducing VEGF levels and inhibiting the growth and proliferation of tumor cells, endothelial





cells, fibroblasts and blood vassels. Everolimus has been approved for the treatment of advanced breast cancer, neuroendocrine tumors of pancreatic origin (pNET), and advanced renal cell carcinoma (RCC)<sup>3</sup>.

This case report may help to explain the temporal relationship between therapy and the occurrence of ONJ with the sequential use of NBPs and mTOR.

A 64-year-old male patient underwent a left, radical nephrectomy in 1992 for clear-cell renal carcinoma. In July 2010 he developed a bone metastasis and he was treated with zoledronic acid 4 mg IV every 4 weeks between 7 July 2010 and 17 August 2012. In February 2011 he had another recurrence, a lung metastasis treated with lobectomy and everolimus 10 mg/die for 6 months from 11 April 2011 to 31 October 2012. In 13 October 2012 the patient showed a facial enlargement and oral fistula in the first guadrant with no history of tooth extraction. A bone scan revealed an ill-defined radiolucency and an orosinusal communication. In January 2013 the patient underwent a right and partial left maxillectomy and is currently being followed up to minimize the risk of new adverse reactions. References

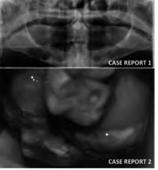
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### **Case Report**

## Unusual solid tumors metastasis to the oral cavity

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Metastatic tumors to the oral cavity account for less than 1.5% of malignant oral neoplasms. They usually occur in patients aged 40-70 years both as bone and soft tissues' lesions, above all in the lower molar area. Metastatic tumors usually derive

from lung, breast, kidney, bone, colorectal and thyroid cancer. Diagnosing a metastatic lesion in the oral cavity is challenging although of great clinical significance since it can be the first manifestation of an unknown neoplasia. We present two case reports of metastasis from solid tumors to the oral cavity.

A 66-year-old male patient, complained about chewing pain in February 2013. He had been operated for prostate (2010), lung (2011) and thyroid cancer (2012). Tumor and swelling in the lower right alveolar ridge and omolateral lymphoadenopathy were found. X-rays revealed a wide osteolytic area in the lower right mandible as well as smaller spread ones controlaterally. Bone biopsy was taken on the right side and revealed metastatic follicular thyroid cancer whereas contralateral lesions were free of disease. Hemimandibulectomy and reconstruction through microvascular fibula flap were performed. Neither mucosal/bone lesions nor symptoms were evidenced after 5-month reevaluation. A 69-year-old patient was referred to the EDU for oral pain of unknown origin, non-responsive to 2-week amoxicillin and NSAIDs therapy. Intraoral examination revealed tooth 1.7 mobility and fistula. After tooth extraction and continuation of medical therapy, no amelioration was obtained. The patient was referred to the Oral Medicine and Pathology Department. Swelling, redness and purulent exudate coming from a large, exophytic and thick mass located in the post-extractive site were present. Immediately, a mucosa and bone biopsy was taken. Pathologist's report revealed metastasis from colon cancer. Afterwards, patient was diagnosed with advanced pleomorphic indifferentiated colon adenocarcinoma. He died one month after palliative hemicolectomy.

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# Rhabdomyosarcoma of the oral cavity in a 24-year-old male patient

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Rhabdomyosarcoma (RMS) is the most common malignant soft tissue tumor. RMS derives from primitive mesenchymal tissues that exhibit a tendency toward myogenic differentiation and probably originate from satellite cells associated with the embryogenesis of skeletal muscle. There are reports of cases arising in oral tissues, which correspond to 10 to 12% of all head and neck RMS. Here we report a case of challenging diagnose of a pleomorphic spindle-cell RMS of the oral cavity.

In June 2012, a 24-year-old male patient was referred to the Department of Oral Medicine and Pathology by his dentist to extract tooth 2.8. On history taking, the patient described a persistent left cheek abscess caused by malposition of 2.8 and non-responsive to 2-month amoxicillin (2 g/die) therapy. He also referred absence of pain, pyretic state and pus/blood discharge during this period. Objective examination revealed slight extraoral asymmetry and intraoral exophityc, hard, immovable cheek proliferation, swelling and redness, associated to ecstasy of Stenone duct. No lymphoadenopathy was recognized. No signs of disease were detected on x-rays. An incisional biopsy was immediately performed. The pathologist diagnosed a "fibromatous lesion" although she stated the small dimension of the specimen could be misleading. An excisional biopsy was scheduled 15 days later and revealed the extension of the lesion deep in the muscular tissue and its pedunculated nature. The pathologist reported a "myofibroblastic inflammatory tumor", positive for desmin, vimentin, CD65 and ki67 but urged on further consultation with another pathologist. Conclusive diagnose after consultation was "spindle-cells/sclerosant multifocal rhabdomyosarcoma". Seven days after, PET, TC and MRI denied the presence of residual tumor cells or of metastasis. Although the patient was recommended to perform adiuvant RT/CT, he refused further interventions. Presently, patient is still free of disease.

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### **Case Report**

# An unusual case of everolimusassociated recurrent oral ulcerations in heart transplant recipient

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Inhibitors of mammalian target of rapamycin that include Everolimus have recently been developed as immunosuppressants, due to their lower nephrotoxicity. Everolimus-induced stomatitis is a poor recognised side effect and very few case reports are present in scientific literature, and none in heart transplant recipients. We report the case of a heart recipient presenting recurrent maior (> 1 cm) ulcerations since heart transplantation, which healed after discontinuing Everolimus therapy.

A 24-year-old female who has undergone heart transplantation in 2010 presented 2 years later to our Department with a history of large (more than 1 cm), recurrent (free periods less than 3 months), long standing (more than 2 months) ulcers in the oral mucosa. The patients assumed Ciclosporin, Everolimus and systemic steroids. The oral

examination revealed a large (> 1 cm), ovoid, well demarcated, with a greyish-white pseudomembrane, bleeding and painful oral ulceration in her right mandibular alveolar ridge behind the second molar. Any traumatic injury or radiographic evidence of impacted or partially erupted third molar was ruled out. Clorexidine, and local corticosteroids were prescribed and the ulcer healed two months later. After an ulcer free period of 3 months the patient developed a further painful large ulcer (> 1 cm) behind the second molar very similar to the last one. It was then hypothesized that Everolimus could be the cause of recurrent ulcerations, and the switch to another class of immunosuppressive agents was suggested<sup>1</sup>. Everolimus was gradually reduced, and one week later, the patient reported a reduction of the pain and evident signs of healing; one month after, the oral ulcer was thoroughly healed. The patient has being free from oral ulcers for 8 months. Everolimus induced recurrent oral ulcers must be take into consideration in organ transplant patients.

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**Case Report** 

## A rare case of (KCOT) keratocyst odontogenic tumor in association with hypodontia

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**Introduction**. Pathogenetic, clinical, radiographic and histopathologic aspects of odontogenic keratocist tumor were analyzed, especially in combination with dental anomalies of number, with the aim of giving useful knowledge to be able to make correct diagnosis, therapy and prognosis in the context of a multidisciplinary approach.

**Clinical case**. The female patient at the age of 14 came to the observation for bilateral agenesis of the upper lateral incisors, diagnosed by Orthopantomografic x-rays. One year after the beginning of the orthodontic treatment, the patient presented for a phlegmonous swelling of the lateral posterior region of the left maxilla. The XR OPT and CT scan showed the presence of a large lesion occupying the left maxillary sinus, radicular resorption of first and second molars and dislocation of the third molar. The treatment of choice is surgical removal by Caldwell-Luc technique. Histological examination confirmed the suspected diagnosis of KCOT. The follow-up at 12 months by OPT and CT control showed the formation of a good trabecular bone in the area affected by the lesion. The follow-up at 24 months showed an optimal healing of the area affected by the lesion, positivity to test pulp vitality of first and second molars and healing of periodontal tissues demonstrated by periodontal probing.

**Conclusions**. The establishment of these two conditions is not due to a common genetic cause, but probably to a random interconnection related to environmental factors. The management of the case required multidisciplinary collaboration between different specialists and careful planning to design a proper treatment protocol and achieve a favorable prognosis.

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# Blandin-nuhn gland mucocele: presentation of a new case and review of the literature



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**Abstract**. Minor salivary glands can be found in the ventral and anterior part of the tongue (Blandin-Nuhn glands). Rarely, these glands can develop a mucocele which appears as an exophytic, sometimes pedunculated, lesion: this feature of the mucocele is due to the absence of a capsule, thus glands are right beneath the mucosa and over the muscle tissue. A 12-year-old healthy Caucasian boy was referred to our Department for the evaluation of an exophitic lesion of the anterior and ventral part of the tongue: the lesion appeared light pink in color, elastic, mobile on the underneath planes, no bleeding and no pain; excisional biopsy of the lesion together with the underneath salivary gland revealed a mucocele of Blandin-Nuhn gland. Anatomy, pathology, clinical features and therapy of Blandin-Nuhn gland mucoceles are discussed: Blandin-Nuhn gland mucoceles, due to uncommon clinical appearance, are often difficult to diagnose. Excisional biopsy is mandatory and, at the same time, diagnostic and therapeutic. A careful removal of the underlining salivary gland is necessary to avoid relapses.

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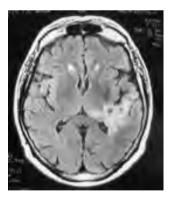
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### **Case Report**

# Brain abscess of odontogenic origin: two cases report and review of the literature

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Brain abscesses are rare, life-threatening infections: odontogenic origin is rarely advocate as the cause of these infections. Neurological manifestation which depend on the involved site (i.e. epilepsy, hemiparesis, hemiparesthesia, aphasia, depressed consciousness) are accompained by systemic symptoms such malaise and fever; mor-

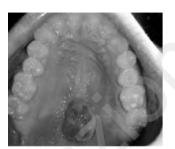


tality rate is high (almost 50%). The identification of causal organism can be difficult and the odontogenic origin is often an exclusion diagnosis. Therapy should be started as soon as possible in order to prevent the rapid diffusion to the rest of the brain: recovery might happen with mild-moderate residual neurological deficit. We present two new cases of brain abscesses; Case 1: a 9-year-old boy with a frontal abscess presented multiple dental foci; a CT scan demonstrated the diffusion to the brain through maxillary and ethmoidal sinuses. Case 2: a 60-year-old female presented with cerebral brain abscess: she was under steroid therapy for reumatoid artritis; a panorex revealed numerous dental foci. No other foci were found in both patients. Due to their rarity, brain abscesses of odontogenic origin are difficult to diagnose: a thorough oro-maxillofacial investigation should always be performed in order to exclude this origin. **References** 

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# Minimally invasive approach to eliminate pyogenic granuloma using Er, Cr: YSGG laser

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**Backgroung.** PG or granuloma pyogenicum is a common tumor like growth of the oral cavity that is considered to be of non-neoplastic nature, arising commonly as a result of constant low grade trauma and poor oral hygiene and in few instances because of hormonal disturbances. Since it is a benign lesion, choice of treatment is surgical excision with removal of underlying cause if any. A newer treatment modality using laser has been reported<sup>1, 2</sup>

**Case report.** A 16-year-old male patient reported to our sector of oral medicine with the chief complaint of intra-oral painless growth since four months. Past history and medical history did not reveal any relevant information. He was taking no medication and had no history of known drug allergy. On intra-oral examination smooth surfaced and lobulated growth of approximately  $1.5 \times 1 \times 0.5$  cm in size was present in the palate. The lesion was pedunculated and was freely movable over peduncle. The surface of the lesion showed non-omogenous red color whereas in certain area was covered by yellowish plaque like material. On palpation, growth was non-tender, nonfluctuant, hard and fibrous in consistency. Treatment plan comprising of excisional biopsy of the lesion using an Er, Cr: YSGG laser. An elliptical incision was made around the peduncle, the lesion was lifted along with the underlying periosteum from the bone surface and removed. Hemostasis was achieved with the same laser and healing was obtained for second intention. Excised specimen was preserved and sent for histopathological examination. At 7 days recall, the gingival tissues were healthy with successful healing.

**Conclusion.** Excisional surgery is the treatment of choice for PG. The use of laser may be a valid approach for the excision of this lesion, as it is minimally invasive and offers many clinical advantages (minimal intra-operative bleeding, hemostasis, reduced times of healing).

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### **Case Report**

# Juvenile Nasopharyngeal Angiofibroma: diagnosis and surgical treatment in I and II stage with dento-alveolar involvement

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**Introduction.** Juvenile Nasopharyngeal Angiofibroma (JNA) is a benign, slowly growing, highly vascular and locally aggressive vasoformative neoplasm that pres-

ents most commonly in adolescent males with a median age of 14 years. The tumour generally originates from the superior margin of the sphenopalatine foramen. Lateral growth can put the tumor in the pterygomaxillary fossa pro-



ducing classic building of the cheek. We report the perioperative management and the different surgical approach of 6 JNA referred to I stage A and B and II stage A and B, according to Radkowski staging ('96).

**Cases presentation.** 6 patients with JNA present epistaxis, nasal obstruction, facial numbness, check swelling, sinusitis and dento-alveolar involvement like wisdom teeth inclusions, confused with odontogenic cysts and tumours and so associated to intra-operative high risk of haemorrhage. They were evaluated by CT and carotid angiography to demonstrate tumour vascular composition and confirm diagnosis. Preoperative biopsy is not recommended due to a risk of profound hemorrhage. Carotid angiography also allows tumour embolization (24 hours before surgery), which reduces intraoperative bleeding.

I stage JNA were removed via trans-palatal approach; II stage lesions were treated by trans-antral approach via Caldwell-Luc incision. Intra- and extra-lesional Diode Laser Photocoagulation (DLP) preceded surgical excision of irregular lump to obtain tumour decreasing.

**Results and conclusions.** Trans-palatal and trans-antral approach were suitable for JNA complete removal and healing. DLP is useful to immediately reduce lesion size and simplifies surgical resection in the same session. At the median follow-up time of 26 months all patients remained free of diseases.

JNA management has changed during the last decades, thanks to technological advances both in radiology (carotid angiography) and surgery (Diode Laser), but it still continues to be a challenge for the multidisciplinary surgical team. **References** 

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### **Case Report**

### **Uncommon gingival swelling**

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**Introduction.** Oral melanocytic nevi are benign tumours of melanocytes. On the basis of histological location of the nevus cells, oral nevi can be divided into 3 categories (junctional, compound and intramucosal). These lesions are usually asymptomatic and commonly involving hard palate and gingiva. Clinically, oral nevi are characterized by high variability in terms of color and morphology. They can occur as a brown or bluishgrav nodule or macule, usually small and well defined.



**Case details.** A healthy, 69-year-old male was referred to our service by his dentist, for evaluation of an asymptomatic gingival lesion. Intraoral examination showed an exophytic mass localized on the attached lingual gingiva of second mandibular molar. The lesion was sessile, painless and hard to palpation. It measured approximately 20x10 mm. The overlying mucosa was normal in color, with a narrow bluish-gray area in the lower portion. Clinical differential diagnosis included: irritation fibroma, peripheral gigant cell granuloma, oral nevus and unknown nature lesion. An incisional biopsy was performed and the histologic description of the sample was "nevomelanocityc proliferation within the submucosalchorion". The final diagnosis was consistent with that of "intramucosal nevus". Consequently, the patient underwent to surgical excision.

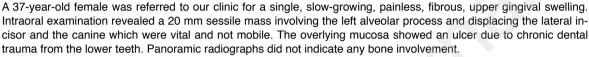
**Conclusion.** Despite melanocityc nevi mainly occur as brown or bluish macules, unusual presentation of these lesions are possible. Indeed, non-pigmented or exophytic nevi have occasionally been reported in the international literature. However, a biopsy to identify the correct nature of these lesions is always recommended.

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# Maxillary peripheral calcifying epithelial odontogenic tumour (Pindborg tumor)

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An incisional biopsy was consistent with peripheral calcifying epithelial odontogenic tumor (CEOT). After a CT assessment showing bone resorption with maintenance of the vestibular cortical bone, the mass was excised under general anesthesia with a conservative surgical approach, comprehending the displaced teeth. The underlying bone resorption was corrected by a fresh frozen homologous bone graft provided by the local Tissue Bank. Healing was uneventful with successful bone graft integration and no recurrence was found after a 2-year follow-up. The histopathological assessment showed sheets of polyhedral epithelial cells with well-defined borders, pleomorphic nuclei and prominent intercellular bridges, amyloid-like material within the sheets of epithelial cells and calcification in the form of Liesegang rings, thus confirming the diagnosis of CEOT.

Peripheral CEOTs are extremely rare, accounting for 6% of all CEOTs and for approximately 0.024-0.18% of all the odontogenic tumours. After the first description by Pindborg in 1966, about 20 cases of peripheral CEOTs have been reported in the literature. They generally occur as single, painless, non-bleeding, gingival masses mainly located in the anterior and premolar regions, that commonly resemble oral hyperplastic or reactive lesions. The significant bone resorption observed in the present case underlines the importance of reconstructive surgery for a proper management and rehabilitation.

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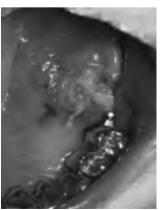
### **Case Report**

# The importance of differential diagnosis between Bronj and hematologic diseases

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**Introduction.** The oral cavity may represent the initial site involved in systemic diseases. A number of systemic diseases have manifestations in the oral cavity, in some cases representing the initial sign<sup>1</sup>. The clinician should recognize these manifestations in order to formulate differential diagnostic hypotheses.

**Case Report.** A 78-year-old woman was referred to our department by her physician for suspected bisphosphonate associated osteonecrosis of the jaw. The patient, affected by rheumatoid arthritis, had been treated with alendronate and per os in the last 5 years and presented an area of exposed bone in the maxilla persisting for more than 8 weeks.



After a careful clinical and radiographic inspection the hypothesis of hematologic disease came out as well<sup>2</sup>. Multiple biopsies of bone tissue and surrounding mucosa were performed. The histopathological examination revealed a large B-cell lymphoma. Thanks to the correct and prompt diagnosis the patient was referred and treated in the department of Hematology. Follow-up carried out in our department showed the progressive and complete healing of the exposed bone.

**Discussion**. The differential diagnosis is a fundamental step in the evaluation of patients, representing the summary of medical history, reported symptoms and objective signs detected during clinical inspection.

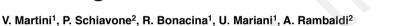
In the clinical practice the formulation of differential diagnostic hypothesis<sup>3</sup> should address the clinician to perform further investigations in order to correctly identify the patient's disease thus ensuring the best treatment.

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### **Case Report**

# NK leukemia: a rare case of oral manifestations representing the initial sign



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**Introduction**. Oral manifestations of leukemia are well documented and may often represent the first sign, as primary lesions (gingival and bone neoplastic infiltrations) or secondary to pancytopenia (gingival bleeding, necrotic ulcerations, petechiae, infections, mucosal pallor)<sup>1</sup>.

**Case Report**. A 52-year-old man presented to the Department of Dentistry in December 2012 complaining painful oral and genital ulcerations; he reported a similar event 2 years before. The oral examination revealed multiple irregular ulcers on the mouth floor, labial mucosae and tongue. Laboratory tests (blood count, inflammatory serum indexes, folic acid-iron-vitamin B12 levels, infectious disease serology) were normal. Topic and systemic steroid treatment succeeded in improving clinical manifestation with total healing of genital ulcers. Histological specimen of relapsed oral ulcers showed only intensive inflammation and the new laboratory tests were still normal. The patient suspended follow-up but in April 2013 he presented in the Emergency Service for acute general malaise; high white cell count was present and the immunophenotyping confirmed an acute mature NK cell leukemia. Wide ulcerations were detectable in the oral cavity.

**Discussion.** Natural killer neoplasms represent a relatively rare group of diseases accounting for <5% of all lymphoid neoplasms<sup>2</sup>; in their pathophysiology widespread necrosis, prominent apoptosis and up-regulation of chemokines involved in these mechanisms are common features<sup>3</sup>. Oral ulcerations of this case report were secondary neither to leucopenia nor to neoplastic infiltration; the importance of a careful evaluation of oral lesions should be pointed out because manifestation in the oral cavity could be sign of systemic diseases.

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# Paraneoplastic Acanthosis Nigricans Maligna

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Acanthosis Nigricans Maligna (ANM) is a paraneoplastic syndrome, defined as a condition that arises in association with a malignancy elsewhere in the body but without malignant nature *per se*<sup>1</sup>. It's a rare dermatopathy that occurs in men and women over age 40, without racial predilection or known familial association. Clinically, ANM showed symmetric, hyperpigmented plaques with variable amounts of epidermal hypertrophy, ranging in color from yellow to brown or black, often with overlaid papillomas. The most common affected sites are body flexures and the posterior neck, but also mucosal surface involvement is frequent and may be the only clinical symptom present<sup>2</sup>. Any mucosal surface can be involved, and as regards the oral cavity, disease affects the lips, tongue, palate, buccal and gingival mucosa.

ANM is primarily associated with adenocarcinomas, mostly of gastric origin. Accordingly, the progression of the tumor will lead to a worsening of the ANM, while regress in case of tumour treatment, and resurface with recurrence and metastases.

A 62-year-old man was referred for evaluation of papillomatous lesions of the oral mucosa, appeared in previous months. The patient was diagnosed a gastric adenocarcinoma at an advanced stage 6 months before. Clinical examination showed diffuse papillomatous or wart-like areas of normal mucosal color and soft consistency involving lips, oral mucosa and hard palate. The tongue and the vestibular mucosa were thickened and furrowed. The lesions are asymptomatic. A biopsy was performed, and histopathological analysis revealed acanthosis and papillomatosis, hyperkeratosis, and lymphohistiocytic infiltrate. The patient was clinically diagnosed with ANM, according to clinical and histologic findings, and the patient's medical history. Isotretinoin 0,05% was prescribed for local therapy. The patient did not return to follow-up due to complications of gastric adenocarcinoma, and died 6 months later.

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**Case Report** 

# Glandular odontogenic cyst: report of a new case with cytokeratin-19 expression

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The glandular odontogenic cyst (GOC) was a rare jawbone cyst described in 1988 by Gardner et al.<sup>1</sup> as a distinct entity. This lesion can involve either jaw, but the anterior region of the mandible was the most commonly affected area. Clinical and radiographic findings were not specific, and the diagnosis of GOC can be extremely difficult due to the rarity of this lesion. The cyst presented a wall constituted by fibrous connective tissue and was lined by a non-keratinized stratified squamous epithelium of variable thickness. Large areas of the lining epithelium presented cylinder cells, sometimes ciliated. A variable amount of mucina was occasionally noted. Due to the strong similarities, this cyst can be easily misdiagnosed as a central mucoepidermoid carcinoma (CMEC). Immunohistochemistry may be an aid in diagnosis; in fact Pires et al.<sup>2</sup> have demonstrated that there were differences in the expression of cytokeratins (CK) in GOC and CMEC. In this study, we reported a new case of GOC in a 38-year-old female patient. The lesion was asymptomatic and was discovered as an incidental finding by a dentist some months before. Oral examination showed normal appearance and color of mucosa. Radiographic examination showed a well-defined, unilocular lesion extending in the left mandible anterior body, in an intraradicular position. The teeth were all vital. The lesion was subjected by surgical enucleation, and the material was sent for histopathological examination. Histopahological analysis of the lesion revealed cyst wall with focally ciliated epithelium lining of variable thickness. The superficial layer of the epithelium showed metaplastic mucous cell with intraepithelial microcystic area. The results of immunohistochemistry revealed strong positive activity for CK 19 in all layer of the epithelium. The postoperative course was uneventful, and there was no recurrence during the 18 months follow-up period.

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## Should jaws positive tracer uptake on scintigraphy be considered a new finding of BRONJ stage 0?

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Stage 0 BRONJ is defined as a non exposed variant of osteonecrosis of the jaws related to bisphosphonates therapy, in patients who present nonspecific clinical and/or radiographic findings. A positive tracer uptake on scintigraphy is not mentioned among the findings of stage 0 disease. We present a case of BRONJ progression from stage 0 to stage 3, in which the earliest documented sign was a maxillar alveolar hyper-uptake found on whole-body scintigrams.

A 64-year-old male was diagnosed a breast cancer in 2003. He developed bone metastatis and the oncologist, before starting intravenous therapy with Zometa, referred him to our Unit for a stomatological evaluation in December 2009. After the extraction of 4.6 considered hopeless, the patient started Zometa infusions in January 2010 and then was visited every 4 months. In December 2010 the bone scintigraphy revealed a positive tracer uptake in posterior maxillar alveolar bone bilaterally, but no evidences, neither clinical nor radiographic (periapical Rx, OPT, CT), of a possible maxillar metastasis were present. A non exposed and asymptomatic bone necrosis was suspected. In December 2011 the patient complained intense pain localized in the left posterior maxillar alveolar bone. Again neither clinical evidences nor radiographic signs of osteonecrosis were present. Antibiotics therapy and laser biostimolation sessions were begun.

In September 2012, during a control visit, a 1 cm<sup>2</sup> necrotic bone exposition distal to 2.5 was noticed. The CT scan revealed bone bilateral alteration in the upper alveolar crest with the thickening of right and left sinus walls. A diagnosis of BRONJ stage III was made.

The patient was treated with antibiotics and then underwent a surgical resection of the left affected tissues with extraction of 2.5 and Er:YAG laser vaporization of necrotic bone (250 mJ, 30 Hz). Weekly session of Nd:YAG laser biostimolation (1,25 W, 15 Hz, 5 min) were scheduled until complete healing was achieved after 1 month.

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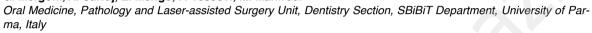
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**Case Report** 

# Oral submucosal hemorrhage as first clinical manifestation of *H.Pylori*-Associated Idiopathic Thrombocytopenic Purpura

### G. Mergoni, A. Sarraj, E. Merigo, P. Vescovi, M. Manfredi



A 35-year-old man was referred for a diffuse bleeding of the oral cavity lasting for some days. Medical history was unremarkable and the patient was not taking any drugs. He did not report any pain, fever or fatigue.

Petechiae on the surface of the soft palate, spontaneous gingival bleeding and hematoma on the left buccal mucosa was present at clinical examination. No head and neck lymphadenopathy was detected.

Several blood test to exclude blood malignancies, coagulopathies or acute viral infections were prescribed. From the blood tests, very low platelets count (12500 plt/ml) was found. No other clinical or laboratory pathological findings were observed. The patient was then referred to the Onco-Hematological Department of the Azienda Ospedaliero-Universitaria of Parma.

With the working diagnosis of acute idiopathic thrombocytopenic purpura (ITP), other possible causes of trombocypenia, including leukemia, drugs, lupus erythematosus, cirrhosis, HIV, hepatitis C, congenital causes, and von Willebrand factor deficiency were progressively excluded. According to the recent evidence of a possible role of *H. pylori* in approximately half of the adults with ITP, the patient underwent the 13C-urea breath test, which resulted positive. The *H. pylori* infection was confirmed with a gastroscopy. The standard triple therapy (clarithromycin, amoxicillin, and a proton pump inhibitor) was maintained for 2 weeks in association with corticosteroids. The eradication of *H. pylori* infection was confirmed and a return to a normal platelets count was observed in a month along with the disappearing of the oral signs of hemorrhagic diathesis.

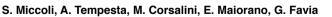
*H. pylori* has a well demonstrated role in several gastroduodenal diseases and recently several studies have focused on its possible causal role in various extragastric disorders, including immune TP. The exact mechanisms of *H. pylori*-induced thrombocytopenia remains unknown, but platelet immunolysis secondary to molecular mimicry has been hypothesized.

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### **Case Report**

# **Oro-Facial Lymphatic Malformation:** management with a new three steps Laser protocol

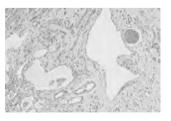


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**Introduction and objectives.** Lymphangioma, or Lymphatic Malformation (LM) according to ISSVA Classification, is a rare benign disorder with unknown etiology. LM may grow slowly over years or develop rapidly over the course of days becoming a bulky lump, infected or bleeding.

Surgical excision is the gold standard treatment for adult LM with 10-15% recurrence rate and 15-30% complication rate. Other treatment modalities have also been proposed: simple aspiration with high risk for recurrence; sclerotherapy (alcohol, steroids, bleomycin or interferon) without results but only complications like fever, pain or lesion enlargement.

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The aim of this study is to show our three steps Laser protocol for LM management, based on its persistent vascular blood component.

**Methods.** We select 11 patients aged between 12 and 60 years with clinical diagnosis of LM of tongue, lower lip and cheek divided in circumscriptum (<3x3cm) and major lesions (>3x3cm). LM are fluctuant and mobile suggesting a differential diagnosis from hemangioma, metastatic lesions and lymphoma. LM laser protocol includes three steps:

- Histological and cytological examination, to evaluate the vascular blood component (10-40%), shows mature lymphocytes with red blood cells and endothelial cells.
- Diode Laser Photocoagulation (DLP) in pulsed mode (on 190-250ms / off 250-450ms) at 14-20W and 800nm, to reduce the lesion.
- Diode Laser surgical excision with histological intraoperative margins control on frozen sections.

**Results and conclusions.** Histological analysis, highlighting the vascular blood component in all LM, validates photocoagulation and surgical combined approach with Diode Laser. Even if it has inconstant results (lesions decreasing rate is 10 to 40% proportionally to vascular blood component), DLP simplifies the last and the most important step. Use of Diode Laser also in surgical excision resets bleeding, infection and lymphorrhea to zero, accelerates healing time and improves aesthetic results.

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### **Case Report**

# Localized granulomatous disease involving mandibular bone and sub-mandibular lymphnodes in a 11-year-old girl



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In August 2011 a 11-year-old girl had the extraction of the first inferior right permanent molar as a result of an odontogenic infection non responsive to antibiotics nor to endodontic treatment. One month later she was referred to our clinic for delay in healing even in absence of any symptom. The extra oral examination revealed right submandibular lymphadenopathy, right mandibular swelling, facial asymmetry; on intra oral examination a pink-strawberry granulating mucosa without any purulent exudate or emission of granular sulfur compounds was observed.

The medical history revealed that the child was suffering from Schönlein-Henoch Purpura.

A CT cone beam scan showed an osteoperiostitis standing from the post-extractive site till the mandibular angle. Both the submandibular US guided FNAB and the incisional biopsy from the alveolus revealed the presence of a granulomatous disorder. Cultural microbiologic tests performed on the biopsy specimen, full blood cell count and chest X-ray were negative. Neither the bone lesion nor the lymphadenopathy improved after a 20-days antibiotic therapy (amoxicillin+metronidazole).

Surgery was performed under general anesthesia aiming to achieve a complete debridement of the mandibular necrotic bone thus removing also the second permanent molar and the germ of the third molar. The lymphadenopathy was removed via an extra-oral approach. All microbiological tests performed were negative and the histopathological assessment confirmed the presence of a non caseating granulomatous disorder with necrosis and epithelioid cells.

The child was hospitalized and underwent a 21-days antibiotic infusion therapy: Tazocin and Clindamycin. No evidence of recurrence was reported in a 20-months follow-up.

In presence of a granulomatous disease both infective and non-infective etiologies have to be ruled out; this a challenging case of granulomatous osteitis and lymphadenitis from unknown cause.

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# Primary intraosseus carcinoma of the jaws arising froma odontogenic cyst - a case report



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Background. Squamous cell carcinoma of the jaw located purely in the bone is extremely rare. Most of these intraosseous carcinomas, also called odontogenic carcinomas are thought to arise from the epithelial lining of an odontogenic cyst. The diagnosis of the development of carcinoma from the cyst lining can only be established by histopatologic examination. Surgeons should appreciate the elevated aggressiveness of this tumor despite adequate surgical treatment. The authors recommend initial aggressive surgical treatment to decrease the local recurrence rate.

Case Report. A primary intraosseous carcinoma arising from an odontogenic cyst in a 58-year-old man is reported. Subjective symptoms were noted by the patient (severe pain and intraoral fistulain the lower jaw). Single-tooth retained with associated presumed cystic lesion was evident in the lower jaw on a routine radiograph. Histology revealed an intraosseous carcinoma after removal of the teeth and "cystectomy". No metastasis was detected. The patient is free of disease after 3 years.

Conclusion. This case report clearly demonstrates the importance of clinician awareness of the malignant potential of apparently innocuous cystic lesion. Although primary intraosseous carcinoma is rare, this case emphasizes the importance of careful histological examination of apparently innocuous odontogenic cysts. In addition, as malignant changes in their epithelial lining are always possible, "cystic" lesions should not only be removed but as completely as possible.

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### **Case Report**

# Uncommon diagnosis of Kaposi's Sarcoma in a HIV-negative adult patient

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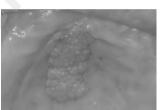
Kaposi's Sarcoma (KS) has been described as a multifocal malignant neoplasm, originating from vascular endothelial cells. Four clinical types has been recognized: classic, endemic, iatrogenic and epidemic<sup>1</sup>. Classic KS has been reported mainly in patients from Mediterranean area and it typically manifests as cutaneous lesions involving the lower extremities and trunk. Classic KS affects only rarely the head and neck region and few cases have been reported as primary oral classic KS. Oral lesions are localized typically in the hard palate (95%), and more rarely they affects the gingiva and the oro-pharynx. Early oral early lesions typically appear as flat pigmented macules of purple or bluish color. After that, the lesions become nodular exophytic and dark and, in certain cases, may ulcerate, resulting

in pain and burning<sup>2</sup>. A 61-year-old man patient was sent at the Sector of Oral Medicine (University of Palermo) for a red oral lesion. The patient reported that the lesion was not healed after tooth extraction (4.3) and 14-day antibiotic therapy. On clinical examination, a flat purple-red macula was observed on the lower labial mucosa with a nodular exophytic lesion of hardened consistency on the midline. Other lesions were not observed in the oral cavity. Two incisional biopsies were performed. For both specimens, the results indicated a proliferation of bizarre shaped vessels, with strands of spindle cells and extravased red blood cells. HHV8 immuno-histochemistry showed a nuclear staining of variable intensity. The patient was HBV and HCV, HIV-1, HIV-2 negative. The patient was then sent to a dermatologist, who described a 0.6 x 0.4 cm red lesion on the plantar surface. The lesion was biopsied, confirming the diagnosis of KS. The patient was admitted to a specialized service for clinical oncology, and is currently under follow-up. In conclusion, clinicians should consider KS as a possibility during the diagnostic work-up of red/bluish oral lesions, especially in the Mediterranean area. Since, albeit very rarely, KS can involve the oral mucosa, when clinicians diagnose classic KS, it is essential to perform a complete dermatological examination, a PET imaging and a gastroscopic examination to rule out the location of the disease in other body regions.

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## Epidermal nevus syndrome: epithelial and cutaneous tumours without systemic disorders: a case report



**Case Report** 

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Epidermal nevus syndrome (ENS) is a rare disease characterized by the association of epidermal nevi with abnormalities in other organs and districts affecting prevalently pediatric patients<sup>1</sup>. The first descriptions of an association of epidermal nevi, neurologic disorders and mental retardation were made by Schimmelpenning in 1957<sup>2</sup> and extensively reviewed in 1975 by Solomon and Esterly. Central nervous system (cortical atrophy, mental retardation, cerebrovascular malformations and neoplasias), skeletal system (bone cysts, scoliosis syndactyly, polydactyly, chinodacctyly) and eyes (choristomas, bilateral cataracts, colobomas) are most commonly involved while endocrine (hypophosphatemic rickets and precocious puberty), cardio-vascular (aneurysms and malformations), urogenital (testicular adenomas, double ureters), oral and skin lesion other than epidermal nevi were also reported<sup>1</sup>. It has been determined that there is not just one category of ENS, distinguished from each other by their clinical features and genetic pattern. In 1995, Happle defined six types of ENS but, for the polymorphism showed by the syndrome, different association of anomalies may be present. Sebaceous nevus syndrome, nevus comedonicus syndrome, Becker nevus syndrome, Proteus syndrome, CHILD syndrome are only some examples of the clinical features showed by ENS<sup>3</sup>. We reported a case of a patients affected by epidermal nevus syndrome with the concomitant presence of oral papillomatosis and cutaneous anomalies including nevi, basal cell carcinoma and siryngocystadenoma without systemic involvement. A concise analysis of the principal epithelial, oral and other lesions associated with ENS and a discussion of the possible relationship between our findings and ENS was performed.

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## Idiopathic fibrous hyperplasia of the palate



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**Introduction.** Idiopathic Fibrous Hyperplasia (IFH) is a rare benign lesion, that involves the connective tissue and is characterized by a slow and gradual increase in gingival volume. It may be generalized or localized. In the first case, it appears during dental eruption, in the localized one, it can appear from the second decade. Generalized form of IFH has generally genetic predisposition and could recur after surgery, in contrast to the localized form. Clinically, IFH appears as a pink swelling of hard consistency while, at histological examination, it is characterized by a proliferation of fibroblasts in a myxomatous stroma.

**Clinical case.** A young female patient of 45 years old without systemic diseases came to our observation since a bilateral swelling of the hard palate, that appeared 5 years ago and had become progressively larger, creating phonation and swallowing problems, referring to IFH. The surgery was performed using a CO<sub>2</sub> superpulsed laser (Smart US20D®, DEKA, Florence, Italy).

**Results.** No bleeding or pain was observed during or after surgery. No sutures were applied and control at 7 and 21 days showed the complete tissutal recovery. Histological examination confirmed the clinical diagnosis of IFH.

**Conclusions.** IFH is a rare benign lesion that, when occurs, needs a surgical approach. In this regard,  $CO_2$  laser could be considered an optimal device since the bloodless field, the relative speed and ease of execution, the absence of suture and the optimal healing for second intention.

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## **Case Report**

## Pyogenic Granuloma: surgical treatment with Diode Laser

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**Introduction.** Pyogenic Granuloma (PG) is a relatively common benign mucocutaneous lesion. The term is a misnomer as the lesion does not contain pus nor it is granulomatous. Etiology of the lesions is unknown, but predisposing factors that have been reported include pregnancy, trauma, vascular malformation and chronic inflammation. PG are usually solitary lesions. The most common intraoral site is marginal gingiva, but lesions have been reported on palate, buccal mucosa, tongue, and lips. Extraoral sites commonly involve the skin of face, neck, upper and lower extremities, and mucous membrane of nose and eyelids. In this report, we seek to highlight the therapeutic advantages achieved with diode laser in intraoral PG treatment compared with surgical excision.

**Material and methods.** We report the cases of 85 patients presenting intraoral dull red, sessile, or pedunculated smooth surfaced nodule that may easily bleed, crust, or ulcerate. 62 were treated with surgical excision and 23 with diode laser treatment. The laser session consisted in diode laser photocoagulation ensued by diode laser excision of the lesion, preceded by treated areas cooling to avoid the tissue demage. According to the literature were used the following specification: wavelength 808nm, 10W power. Histological evaluation showed hyperplastic stratified squamous epithelium with stroma consisted of a large number of budding and dilateted capillaries and a dens chronic inflammatory cell infiltrate. Diagnosis of PG was confirmed.

**Results and Conclusion.** Rapid healing can be observed within a few days of treatment, and as blood vessels are sealed, there is an improvement of haemostasis and coagulation compared with surgical excision. Post-operative

pain discomfort, edema and bleeding are notably reduced. In conclusion, the use of diode laser offers a new tool that can change the way in which existing treatments are performed.

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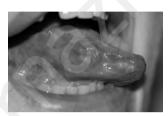
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## **Case Report**

## Oral ulcers and chronic gastrointestinal disorders

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Oral mucosal lesions may be markers of chronic gastrointestinal disorders: Recurrent Aphthous Stomatitis, systematic dental enamel defects and non-specific atrophic glossitis may represent oral manifestations of Coeliac Disease (CD), an autoimmune enteropathy characterized by a malabsorption syndrome<sup>1-3</sup>.

**Case Description**. The authors present a case of CD with no gastrointestinal symptoms and predominance of oral manifestations. A 46-year-old female whit painful with an history of several oral ulcers was referred to the Dentistry Department of Siena University Hospital to undergoing a check-up. Intraoral examination revealed the presence of a large ovoid ulcers with circumscribed marked margins, erythematous haloes and grey floors involving the ventral surface of the tongue; other ulcers were on labial mucosa. Histological examination of the lingual ulceration showed the presence of inflammatory infiltrate and granulation tissue in the deep lamina propria but no neoplastic proliferation was detected. The routine laboratory examinations (biochemical analysis, complete blood counts, and immunological study) resulted normal and no HSV 1-2 antibodies were detected. Serologic screening for CD was performed and elevate levels of anti-endomysial (EMA), anti- transglutaminase(tTG), anti-gliadin peptides (DPG IgA and DPG IgG) antibodies have been found.

**Conclusions**. The diagnosis of CD can be difficult, especially because patients may exhibit a wide spectrum of signs and symptoms clinically heterogeneous, and therefore often unsuspected. Screening tests for CD patients with oral aphthae or idiopathic atrophic glossitis should be considered even in the absence of gastrointestinal symptoms. **References** 

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## **Case Report**

## A case of linear IgA disease limited to the oral mucosa

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Linear IgA disease (LAD), is a rare, chronic, sub-epithelial blistering condition that predominantly affects the skin and mucosal surfaces. Most cases are idiopathic but some drugs, infection, trauma, autoimmune disorders, and malignancies have been documented as potential inducers. In the present report we describe an uncommon case of LAD



in which mucosal involvement represented the sole manifestation of the disease. In May 2013, a 48-year-old man was referred to our Department following the persistence of severe desquamative gingivitis, treated unsuccessfully with conventional periodontal treatment by his dentist. His medical history was significant for hypertension, which was being treated with Athenolol. Intraoral examination showed a localized painful gingival erithema, associated with ulcerations and bleeding. Nikolsky's sign was negative. No skin lesions were detected during clinical examination. The patient underwent two incisional biopsies for histology and direct immunofluorescence (DIF). Histopathology revealed a mixed inflammatory infiltrate. DIF reveled a deposition of IgA at the basement membrane zone; research of IgG, IgM, C3 and fibrinogen was negative. These findings suggested a diagnosis of LAD. The patient was advised to discontinue the use of Athenolol and/or substitute it with other antihypertensive drug. A desquamative gingivitis is more commonly associated with mucous membrane pemphigoid or erosive lichen planus, and although more than half of LAD patients present oral mucosal lesions, there are only a few cases reported of oral lesions as the only manifestation of LAD <sup>1</sup>. DIF plays a very important role in case of vesciculo-bullous diseases to address the correct diagnosis and treatment.

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# Laser-assisted surgery in oral medicine: treatment of fibrous epulis with diode 915 nm

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**Case Report** 

**B.** Polizzi<sup>1</sup>, A. Albanese<sup>1</sup>, G. Giannatempo<sup>2</sup>, G. Colella<sup>3</sup>, G. Campisi<sup>1</sup> <sup>1</sup> Department of Surgical, Oncological and Oral Sciences, Sector of Oral Medicine "V. Margiotta", University of Palermo, Italy; <sup>2</sup> Department of Surgical Sciences, University of Foggia, Italy ,<sup>3</sup> Department of Head and Neck Surgery, II University of Naples, Italy

The purpose of this case is describing the treatment of a fibrous epulis (FE), the most common type of localized gingival overgrowths, with a Diode Laser. FE management involves causal periodontal treatment, surgical excision, usually performed with scalpel, and often the extraction of adjacent teeth. The Laser surgery has been proposed as a less invasive method based on the following advantages: better control of intra/post-operative pain and bleeding by coagulation, resulting in a bloodless surgical field, an excellent intra-operative visibility for the operator and an increase in surgical accuracy.

A male patient of 78 years, anamnestically not reporting systemic diseases, drugs and smoke/alcohol assumption, was observed at Sector of Oral Medicine for a firm, pink and painless mass (> 1 cm) at the interdental papillae between 1.2-1.3. Clinical examination reveals a poor oral hygiene state and severe mobility of 1.2. The treatment plan consisted of extraction of 1.2, excision of the lesion and causal periodontal therapy. After superficial desensitization with application of a lidocaine 2.5% + prilocaine 2.5% cream (EMLA<sup>®</sup>), excision was performed with Diode Laser 915nm (900 LASEmaR, Eufoton, TS, Italy) according to the manufacturer's protocols (C.M. continues, 1.5 W, 300  $\mu$ m). Surgical wound healing occurred by secondary epithelialization. Excised tissue was submitted for histological examination, confirming the diagnosis.

The treatment protocol showed the following advantages: a) reduced administration of anesthetic, despite the considerable lesion size, without any intraoperative pain; b) absence of significant complication (e.g hemorrhage-painedema-swelling-infection) in the post-operative period with complete healing process in 20 days without scaring; c) complete removal of lesion, without any recurrence at 1 year follow-up.

Given the intrinsic features of Diode Laser, it is reasonable assuming that it may be an advantageous device in the surgery of soft oral tissues.

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## Primary intraosseous carcinoma of the mandible arising from epithelial remnants of odontogenic cyst

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Primary intraosseous squamous cell carcinoma (PIOSCC) is a rare malignant

odontogenic tumor arising from odontogenic epithelial remnants within the jawbones. According to the 2005 WHO Classification of Tumors<sup>1</sup>, there are three subcategories of PIOSCC, histopathologically divided into 3 types: solid type carcinoma, carcinoma derived from a keratocystic odontogenic tumor, and carcinoma derived from an odontogenic cyst. We report a case of PIOSCC of the jaw with probable origin from rests of an odontogenic cyst.

A 54-year-old woman complained pain and swelling on the left side of her neck and cheek mucosa three months later a two stages surgery for extraction of mandibular third molar associated with odontogenic cyst, in region of tooth #38. Panoramic radiography (PR) and Nuclear Magnetic Resonance Imaging (NMRI) results suggested an osteolytic mandibular tumor. An incisional biopsy was then performed, which indicated a moderately-differentiated (G2) squamous cell carcinoma, with invasion of sub-mucous tissues. Based on the clinical and pathological findings, a diagnosis of PIOSCC was made. The patient underwent to hemimandibulectomy, left neck dissection, and received chemotherapy and head-neck radiation. Tumor staging was classified as T4,N2,M0.

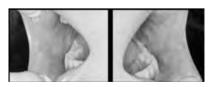
Gardner<sup>2</sup> has argued that the diagnosis of such a tumor arising from a cyst requires the presence of a transitional layer between the normal cyst epithelium and the carcinoma cells. The longstanding chronic inflammation might be the main predisposing factor for malignant transformation in the cyst epithelium.<sup>3</sup> In this case, no transitional layer was visible, as a result of severe inflammation lasting over 3 months.

We concluded that the tumor had probably arisen from the cyst because the two tissue types were so closely aligned in a single specimen characterized by chronic infiltration of lymphocytes, plasma cells, and crystals of cholesterol. **References** 

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## **Case Report**

## A case of Heck's disease treated with Quantum Molecular Resonance Scalpel



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A 13-year-old Hispanic female patient was referred to our Unit by her Dermatologist for multiple and bilateral lesions on the buccal, labial and lingual mucosa. The lesions appeared in clusters and were soft, non-tender, flattened and of the same colour of the oral mucosa.

The patient was otherwise healthy and she did not present other skin lesions.

The clinical findings were consistent with the diagnosis of Heck's disease. This disorder, also called focal epithelial hyperplasia, is a virus-induced, localised proliferation of oral squamous epithelium firstly described in native Americans and Inuits. The lesions, typically seen during childhood, are tough to be caused by papillomavirus type 13 and possibly 32. Spontaneous regression of focal epithelial hyperplasia has been reported after months or years and this spontaneous recovery is presumed to be the cause of the rarity of the disease in adults.

In this patient, a surgical removal of the lesions was planned for aesthetic purposes and because the patient reported frequent masticatory trauma of the lesions. The removal was completed in 3 sessions using the Quantum Molecular Resonance Scalpel (Vesalius<sup>®</sup>, Telea, Sandrigo, Italy). This device is based on a physic theory, the molecular resonance (RQM), which allows the operator to cut and coagulate tissues simultaneously in a precise and controlled way, without application of any manual pressure or burning. It uses electric current to produce a flux of quanta sufficient to break molecular bonds by resonance.

During the interventions no bleeding was recorded and sutures were not necessary.

The histopthological evaluation of the specimen showed considerable acanthosis and superficial keratinocytes with the typical koilocytic changes of papillomavirus infection.

The post-operative period was uneventful and the mucosa healed without scars.

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## **Case Report**

## A case series of 42 non-complaining bad breath patients and the neuropsychology of their disease



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Although halitosis is a common condition that affects approximately 25% of the population in chronic and transient forms, many people imagine that they have halitosis and overstate concerns about having bad breath. Some patients are extremely worried about their breath quality, usually misinterpreting sensory perceptions from their mouth and behaviors of other people. The aim of this interdisciplinary study was to develop a diagnostic assessment tool employing a neuropsychologic rating test. The test may be useful in non-specialistic clinical settings and allow discriminating halitophobic individuals, obsessed by their breath, from patients with real halitosis through the analysis of the neuropsychological components of the disease.

In this study we present preliminary data from a case-series of 42 patients with pathologic values of oral volatile sulfur compounds (VSC). VSC were greater than 100 ppb in spite of the treatments provided. Oral VSC concentration was measured with an Interscan Halimeter<sup>®</sup> device. Patients filled in a 7 point likert rating scale composed by 3 sections: a) questions about anxiety, depression, bad breath, disgust in other people related to proxemic vicinity during interpersonal relationships; b) questions about cognitive ideation regarding bad breath and c) questions about oral hygiene strategies to avoid bad breath. In comparison with a known group of halitophobic patients, the analysis on data from the bad breath cohort showed (i) no demographic differences between patients with and without halitosis; (ii) a greater age of individuals with persistent pathologic VSC (p=.015); (iii) lower educational level in bad breath individuals (p=.036); (iv) no other differences for part b and part c of the questionnaire but bad breath patients judged less frequently disgusted behaviors in other people (6 questions of part a), such as facial expressions of disgust and/or gazing to their mouth (Q3: p=.015; Q8: p = .001; Q9: p = .001; Q10: p < .001; Q11: p = .014). **References** 

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## Severe ulceration of the tongue: first symptom of a psychiatric disorder



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A young male (21 years old) was admitted to Head and Neck Pathology Department of 2nd University of Naples with a wide left tongue margin injury started three months ago; the lesion had been already biopsed at Department of Oral Sciences of the University of Palermo, with the histopathological diagnosis of ulcerated mucous membrane, superficially necrotic, in presence of both acute and chronic inflammation. The patient minimised these tongue bites by keeping a piece of cloth in the mouth: he showed progressive difficulty in chewing and swallowing foods. Additionally, he had multiple motor tics in the form of jerky movements of the trunk, shoulder, neck, limbs and vocal tics. The ulcerated oral lesion get wider quickly, up to a size of 4x5 cm, with the symptom of intense burning. A second incision biopsy was performed, excluding any picture of neoplasia and documenting again the presence of chronic inflammation, with remarkable angiogenesis. In that period the patient uttered often a cry, he was in need to bite the tongue, and he declared to have benefit and gratification during or after the biting. Laboratory investigations, CT, brain MRI and ENMG, did not reveal any abnormality. In the meantime a tongue retainer device was applied to avoid or limit the biting of lesion. The psychopathological history revealed the onset of the disorder one year before when the patient referred that he felt an increasing sense of tension before inflicting injuries to his tongue, besides the attempts to resist, and gratification or relief during or after the injury. After psychiatric evaluations were made a severe obsessive-compulsive disorder (OCD) resulted. Hence, the patient started a pharmacological treatment with citalopram and haloperidol (for the tics). Just one month later, the injury and the tics disappeared and the boy started working and normal social activities.

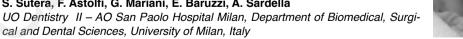
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## **Case Report**

## Multiple melanotic macules of unknown origin

## S. Sutera, F. Astolfi, G. Mariani, E. Baruzzi, A. Sardella



Introduction. Melanotic macules are localized deposits of melanin without an increasing in the number of melanocytes. They are frequent in females between 40 and 50 years of age, with a preferential localization on lips, gingiva, buccal mucosa and palate.

Case details. A 41-year-old woman was sent to the Unità Complessa di Odontostomatologia II of the Ospedale San Paolo di Milano for oral specialist medicine assessment about intraoral multiple pigmentations.

The patient was a no smoker-woman, who only made use of birth control pills.

She reported that the pigmentation of the labial mucosa appeared about 5 years before, while intraoral pigmented were observed, for the first time by her dentist a month before. She also complained discomfort and burning of the upper labial mucosa.



At physical examination, multiple melanotic macules of lips, lingual margins, hard palate and buccal mucosa could be detected.

We carried out an incisional biopsy which allowed the histopathologic diagnosis of "melanotic macule."

During follow-up visits, the patient reported increasing burning and pain at the site of biopsy, and she also showed an increasing number of lesions and intensification in colour of pre-existing labial ones. Blood tests did not show abnormal values.

**Conclusions.** Melanotic macule may appear, together with other signs, in several syndromes or following the use of drugs, but sometimes their origin remains unclear.

In the clinical case here described, multiple pigmented lesions were not associated with other abnormalities of the skin, nails or blood values. Pharmacological anamnesis documented the intake of birth control pills, but only after the appearance of the maculas.

Biopsy allowed the diagnosis of melanotic macules without, however, correlate with any other syndromes. **References** 

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## **Case Report**

## A case of juvenile spongiotic gingivitis

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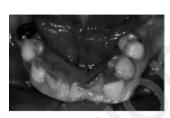
**Introduction.** Juvenile Spongiotic Gingivitis (JSG) is an uncommon clinicopathologic entity, described for the first time in 2007 (Darling et al.). JSG clinical presentation is characterized by bright red patches, with granular, pebbly or velvety surface, involving attached gingiva. The lesions are generally multifocal, painless and sometimes associated with bleeding during brushing in approximately 20% of cases. JSG is most commonly localized at the maxillary and mandibular labial gingiva, not necessary involving marginal gingiva. Histologically, JSG is characterized by the loss of keratinization, that makes the tissue gingiva similar to junctional epithelium with a decrease of defense against oral pathogens and trauma. The age of affected patients ranges from five to twenty years. No specific treatment is at the moment available.

**Case details.** We present the case of a 11-year-old female that came to our attention for a gingival lesion. Intraoral clinical examination showed three bright red lesions, slightly detected, of soft consistency, on the vestibular attached gingiva in the area of 11, 12 and 23. The lesions were asymptomatic. During first examination it was decided to follow up the patient and 1% chlorexidine gel was prescribed.

**Conclusions.** JSG incidence and pathogenesis remain unknown. The lack of response to periodontal treatment and the lack of association with plaque excluded a role of local bacteria in the etiology of this condition. Juvenile Spongiotic Gingivitis treatment is still controversial and the surgical excision showed high recurrence frequency about 25% of cases. **References** 

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# Oral squamous cell carcinoma presenting as a cervical lym-phadenopathy



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Introduction. Oral squamous cell carcinoma originates from epithelium of oral mucosa and it is the most frequent type of cancer in this region. Clinical presentations include white and red lesions, ulcerated areas and/or swelling.

**Case details.** Mr. EF is a no smoker 68-year-old man, affected by hypertension and diabetes, under pharmacological therapy. He was visited in the department of maxilla-facial surgery of our hospital because of a submandibular swelling, appeared about 2 months before on the left side of the neck. A preliminary needle aspiration of the lesion was performed by the surgeon, which showed presence of OSCC cells.

Because of the lack of an evident intraoral lesion of neoplastic nature, the patients was referred to our service for a visit. At physical examination, a large cervical node of hard consistency was present, while intraoral examination showed poor oral hygiene and signs of periodontal disease. It was not present any clinically evident sign of oral cancer, although it was noticed a small lesion of the floor of mouth, close to the left sublingual caruncle, with a consistency harder than the surrounding mucosa.

We carried out an incisional biopsy of the lesion, which confirmed the diagnosis of squamous cell carcinoma. The patient underwent surgical treatment and neck dissection.

**Conclusions.** What we learnt from this case is that even a small and apparently innocent mucosal lesion can be a cancer of the mouth. And that even a nearly invisible squamous cell carcinoma can spread to cervical lymph nodes. **References** 

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## **Case Report**

## Phaces syndrome with intraoral and perioral hemangiomas: a different approach with Diode Laser



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**Introduction.** PHACES is an acronym which refers to a syndrome charachterized by **P**osterior cranial fossa malformation, Hemangiomas of the head, **A**rterial abnormalities, **C**ardiac abnormalities, **E**ye abnormalities, **S**ternal clefting. The syndrome shows a female to male predilection of 9:1. There is no definitive evidence of a familial tendency although an X chromosome linked dominant single gene etiology has been suggested with lethality in males. The diagnosis of PHACES syndrome requires the presence of a segmental hemangioma >5 cm in diameter of the head plus 1 major criterion or 2 minor criteria. There are many treatment options, one of the mainstay therapies is laser photocoagulation. The aim of this work is to show the differential diode laser treatment of intraoral hemangiomas (IH) and perioral hemangiomas (PH) in patients with PHACES syndrome.

**Methods.** We report the cases of 4 patients (age median 21 years) affected by the syndrome, showing 26 IH and 15 PH. Diagnostic protocol consist on: appropriate clinical exam (highlighting prominent reddish purple plaque-like lesions on lips, tongue, cheek, palate, mouth floor), echocardiogram, ophtalmologic and endocrinologic exams, MRI of the head and MRA of head and neck. Patients were treated with multiple laser sessions. A different approach de-

pending on the site of the lesions was used: repeated diode laser impulses in pulsed mode at the power of 14-20W and the wavelength of 800 nm for IH and lower power for PH, preceded by treated site cooling to avoid the tissue damage.

**Results and Conclusions**. IH totally healed after 1 or 2 sessions. In each session only a limited area of the PH was treated, obtaining a progressive improvement of the lesions. Both in IH and PH slight post-operatory pain, bleeding and edema was proved, with a low incidence of complications such as ulceration and super-infection.

Diode laser has been proved to be very effective as non-invasive treatment for IH and PH in PHACES syndrome patients.

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## Focal epithelial hyperplasia (Heck's disease)

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Focal epithelial hyperplasia (FEH) or Heck's disease is a rare disease of the oral mucosa; it can be mostly found in children or young adults, immunosuppressed and who live in regions with low socioeconomic status. It is characterized by asymptomatic papules on the oral mucosa, gingiva, tongue, and lips<sup>1</sup>. Healing can be spontaneous, and the treatment is indicated if there are aesthetic or functional complications. Human papillomavirus (HPV), especially genotypes 13 and 32, have been associated with FEH and detected in the majority of lesions<sup>2</sup>. Histopathologically FEH is characterized by parakeratosis, epithelial hyperplasia, focal acanthosis, fusion and horizontal outgrowth of epithelial ridges<sup>3</sup>. Here it is described the case of a 40-year-old male patient, HIV-positive, smoker with numerous, asymptomatic oral papules clinically and histologically corresponding to FEH. Labial and buccal mucosa were especially affected by lesions. The treatment was performed using KTP laser 532nm, in continuous mode with a 300mm fiber, power 1,4W (power density 1980.22 W/cm<sup>2</sup>). After anaesthesia without vasoconstrictor the lesions was tractioned with suture or Allis clamp and then completely excised. The lesions were preserved in 10% formalin for histological examination, that confirmed the clinical diagnosis of FEH. In this case the laser has allowed an excellent control of bleeding, without postoperative suture, and optimal wound healing.

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## **Case Report**

# Unusual presentation of a solitary plasmocytoma of the jaw: a case report



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## **Case Report**

**Introduction**. Plasma cell malignancies include two localized forms (e.g. Solitary Plasmocytoma of the Bone-SPBand Extramedullary Plasmocytoma-EMP) and one more severe disseminated form (e.g. Multiple Myeloma-MM). SPB usually affected male during 6th decades, it occurs as a single uni- or multilocular osteolytic lesion, histopathologically characterized by an abnormal proliferation of plasma cell. In some patients SPB may progress to MM, characterized by multiple osteolytic lesion in several bones, presence of abnormal plasma cells in the bone marrow biopsy, detection of amyloid deposit and abnormalities of immunoglobulin production. In both clinical forms, the maxillofacial area is rarely involved, the higher risk sites are the marrow-rich areas (ramus, angle and retromolar trigon of the mandible).

**Case presentation**. A male patient of 60 years, anamnestically reporting diabetes and hypertensions, was observed at the Sector of Oral Medicine for the presence of pain and paraesthesia localized in right emimandibular area arisen from about 2 months after the extraction of 4.7 and 4.6. The patient denied previous therapy with bisphosphonates and/or angiogenesis inhibitory drugs. Clinical examination showed an area of osseous exposition in the site of right emimandibula. Rx exams (OPT and TC) showed a radiolucent unilocular area of osteolysis in the same region (2.1 x 1.2 x 1.8 cm) with infiltration of mandibular canal. Patients showed both local (e.g. pain, swelling, paraesthesia, infection) and systemic symptoms/signs (abnormal protein electrophoresis, anaemia, thrombocytopenia, ESV/ PCR increase, weight loss and renal failure). Histopathological examination of the biopsy confirmed the diagnosis of SPB, complicated by bone exposition and bacterial superinfection after teeth extraction. The patient has been referred to a haematologist for stadiation and therapy.

**Conclusions**. Differential diagnosis of apparent spontaneous osteonecrosis of the jaw should be include haematological disorders, such as SPB and MM.

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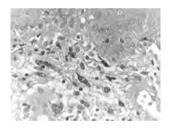
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**Case Report** 

## A rare case of oral anthracosis resembling an oral leukoplakia

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Anthracosis is a non pathologic condition characterized by deposit of carbon particles in the lungs and in the lymph nodes. It is caused by excessive exposure to carbonaceous material including tobacco smoke. Anthracosis is given by deposits of carbon particles phagocytized by macrophages that subsequently move to perivascular connective tissue and lymph nodes. Extrapulmonary anthracosis is very rare, and its etiology is not well established. We report a case of anthracotic pigment deposit in the cheek mucosa of a 56 years old strong smoker woman. The patient presented with a white diffuse oral lesion in the cheek mucosa. The presence of any traumatic factor was excluded and the patients underwent an incisional biopsy. The histological examination showed hyperkeratosis of the epithelium and carbon-laden macrophages located in the superficial chorion. The diagnosis of oral anthracosis associated with tobacco related keratosis was established basing on the histological findings and personal history of tabagism. The patient was convinced to stop smoking. After some time the white diffused lesions disappeared confirming the previously established diagnosis of tobacco related keratosis associated with anthracosis. The pathogenesis of extrapulmonary anthracosis is still unclear. A few case reports described anthracosis of the esophagus 1 but, to the best of our knowledge, there are no reports in the literature describing this condition in the oral cavity. The presence of white plaques diffused to all oral cavity requires differentiation from oral leukoplakia which is a lesion at risk of malignant transformation and that requires a radical surgical approach. Instead, the diagnosis of oral anthracosis in a smoker patient only imposes to convince the patient to stop smoking.

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## Peripheral gigant cell granuloma (giant cell epulis) associated with metabolic diseases: case report and literature review



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Peripheral giant cell granuloma (PGCG) is a relatively frequent benign lesion of the oral cavity, originating from the periosteum following local irritation or metabolic diseases. PGCG is a soft tissue lesion that can affect the underlying bone, which can develop at any age, and manifests as a red-purple nodule located in the region of the gums or edentulous alveolar margins. After complete clinical removal of PGCG recurrence occurs in about 10% of cases, which may result in an esthetic and functional soft tissue defect. In this paper, we report a case of PGCG of about 1 cm, sessile with a painful and bloody appearance with vegetating. The case has bad oral hygiene with a diffused gingivits and affection chronic renal failure stage IV, arterial hypertension with diabetic retinopathy caused by diabetes mellitus type I and dialysis therapy waiting for renal transplantation. The lesion was removed in its entirety with root planing and gingival repositioning and it has been relpased after one year. According to our study and review of the literature, we can assume that among the various causes of etiological PGCG in addition to those already known as trauma and poor oral hygiene, there are also some metabolic disorders such as chronic renal failure, which are often the cause of relapses of PGCG once removed as in this case. Our study illustrates how chronic kidney failure is one of cofactors in the etiology of PGCG, and also causes of recurrence.

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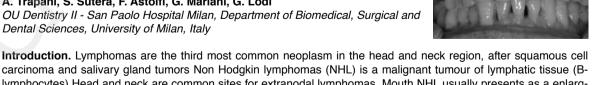
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## **Case Report**

## Lymphoma non-Hodgkin or not: that is the question

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carcinoma and salivary gland tumors Non Hodgkin lymphomas (NHL) is a malignant tumour of lymphatic tissue (Blymphocytes). Head and neck are common sites for extranodal lymphomas. Mouth NHL usually presents as a enlarging mass, sometimes ulcerated, affecting any oral sites. Lesions are frequently asymptomatic, although they may be associated with pain or fever and weight loss.

Case Details. A 65-year-old male patient came to our service referring the development of a lump on the upper gum, noticed about 4 months before and treated by his dentist with antibiotic therapy. Intraoral clinical examination showed a pink and hard swelling, tender at palpation, with the major axes of 3 cm, at level of 12-13.

As the teeth in the area were vital, the clinical diagnosis was of inflammatory process of gingival origin. Because of the unusual presentation and the lack of response to antibiotic treatment, a biopsy was performed. The histological diagnosis was of putative NHL, although the pathologist asked for a larger sample. For this reason a new biopsy was taken and this time the histological examination excluded the NHL diagnosis, although a definitive diagnosis was not provided. One month later, we decided to remove the whole lesion and again it was negative for NHL. During a follow up visit, a new lesion appeared in the same area: immediately a new biopsy was performed and, this time, the histological diagnosis was of NHL.

**Conclusions.** Although histological examination is the mainstay of diagnosis in most of the oral medicine conditions, it is not free from mistakes or limitations. Unfortunately, we have very little evidence on fallacy of histological diagnosis. Final diagnosis always depends on the team work of clinician and pathologist and on their ability to synthesize histological and clinical information.

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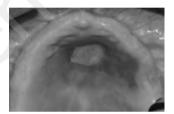
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## Case Report

## An unusual case of Oral Lichen Planus

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**Introduction.** Oral Lichen Planus (OLP) is an immune-mediated chronic inflammatory disease, with a plethora of clinical presentations (from atrophic to ulcerative forms). OLP in its ulcerative form is usually in differential diagnosis with pemphigus vulgaris (PV), pemphigoid of mucous membranes (PMM) and oral lichenoid lesions, including oral lupus erithematosus (OLE).

**Case details.** We present the case of a 67-year-old female, who came to our attention for a palatal lesion. Intraoral clinical examination showed an ulceration of 3 cm of diameter, surrounded by an erythematous area, localized at the anterior third of the hard palate. The clinical diagnosis was OLE mainly because of the palatal collocation (suggestive of lichenoid lesion LE correlated), although OLP and PV were considered in the differential diagnosis. During the first visit, we decided to perform an incisional biopsy for both the histopathological and immunofluorescence exams. The histological exam was suggestive of OLP. The patient was then treated with clobetasol gel 0.05% associated with anti-fungal agents to prevent candidiasis. The treatment was effective, eliminating both symptoms and the palatal lesion.

**Conclusions.** Ulcerative OLP may share clinical features with PMM, PV and OLE. The histopathological exam, together with direct immunofluorescence, can be necessary to confirm the diagnosis, particularly in case of less common clinical features.

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## **Case Report**

## Sebaceous carcinoma of the lip

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Sebaceous carcinoma (SC) is an uncommon neoplasm. To date fewer than 400 cases have been reported in literature. Due to the low incidence and the not universally accepted histopathological classification, it presents diagnostic problems<sup>1</sup>. Generally, the lesions arise in the meibomian glands of the eyelid. However, extra-ocular localisation in the head and neck region have been reported. While several reports have documented sebaceous adenomas arising from sebaceous glands of the oral cavity, oral sebaceous carcinomas are extremely rare. To date only six cases have been described<sup>2</sup>. The salivary glands too are considered an uncommon site, even if some cases arising in the parotid gland were recognised. Herein we reported a case of SC arising in the lateral edge of the lower lip in a 71year-old men. To the best of our knowledge this is the second case described in lips. The clinical differential diagnosis included squamous cell carcinoma, basal cell carcinoma with sebaceous differentiation (BCCSD) and salivary gland neoplasms.

The operation was performed under local anaesthesia. The lesion was removed with 0.5 cm of free margins and a W shaped wedge. The defect was primarily closed. The post-operative course was uneventful. Although SC may be found among the multiple sebaceous neoplasms occurring in association with multiple visceral carcinomas in the Muir-Torre syndrome, in the present case the lip was the only localisation of SC. SC must be distinguished from basal cell carcinoma with sebaceous differentiation (BCCSD). The diagnosis may be facilitated by lipophylic stains on frozen sections or immunostains for EMA and S-100.

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## **Case Report**

## **Recrudescent oral HSV infection**

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A 46-year-old male with chronic lymphocytic leukemia 20 months status-post reduced intensity allogeneic hematopoietic cell transplantation presented with new onset of painful oral ulcers. He had been previously diagnosed with chronic graft-versus-host disease of the skin, mouth, liver and gut, and was being treated with prednisone, my-cophenolate mofetil and extracorporeal photopheresis, valacyclovir (4g/day due to previous oral breakthrough infections) and sulfamethoxazole and trimethoprim prophylaxis, as well as intraoral topical steroid therapy. Clinical examination revealed multiple, extensive, superficial, irregularly shaped ulcers of the lips (Panel A), floor of the mouth (Panel B) and tongue (Panel C, D). Viral culture was positive and direct fluorescent antibody (DFA) test confirmed Herpes Simplex Virus -1 (HSV-1); susceptibility testing demonstrated acyclovir susceptibility. The patient was admitted for intravenous foscarnet therapy with resolution of oral lesions after one week. The patient was discharged on valacyclovir 4g/day, and at two-month follow-up there were no signs of recurrence. After four months the patient presented with new multiple painful oral ulcerations that tested positive by DFA for HSV-1. CMX001 (1-O-Hexadecy-loxypropyl-Cidofovir) was initiated and the ulcers healed completely with resolution of symptoms after 2 months. The patient remains on long-term antiviral therapy at the same dose.

### References

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