Carcinoma of the lip five years after bone marrow transplantation

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A 34-year-old man presented to our observation for a persistent exophytic crust on the right lower lip. Five years before he underwent allogenic bone marrow transplantation for an acute myeloid leukaemia. After transplantation, he developed a severe graft versus host disease with cutaneous and hepatic involvement. He was treated with steroids (stopped by 5 months), mycophenolate mofetil (stopped by 15 months) and photopheresis (stopped by 22 months). Crusty lesions on the lower lip appeared about 6 months before our observation and were treated with antiviral drugs assuming an herpetic infection. An incisional biopsy was performed revealing a squamous cell carcinoma (cT1N0M0). The lesion was surgically excised in general anaesthesia and a vermilion reconstruction with buccal musculomucosal flap combined with V-Y plasty was performed after malignant tumor excision (pT1Nx).

Hematopoietic stem cell transplant (HCT) recipients have a well-known substantial risk of developing secondary solid cancers frequently involving the lip and the oral cavity, particularly beyond 5 years after HCT. Several factors as chronic GVHD and prolonged immunosuppressive treatment beyond 24 months are well-documented risk factors for many types of secondary cancers.

Conclusion. Screening and preventive care for secondary cancers are pivotal issues among HCT recipients. The frequent involvement of the lip or the oral cavity highlights the importance of ruling out squamous cell carcinoma in presence of persistent lesions in HCT recipients.

References

Azathioprine-induced oral lesions: a case report

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Introduction. Azathioprine is a purine antimetabolite used for treatment of different autoimmune diseases. In rare situations it can cause life-threatening leukopenia. In the present report we describe a case of azathioprine-associated leukopenia diagnosed for the appearance of widespread lesions of attached gingiva.

Case presentation. In April 2013 a 58-year-old female was referred to the Department of Oral Science, University of Bologna, following the appearance of painful diffuse gingival lesions. Intraoral examination revealed haemorrhagic ulcers diffused to the upper and lower gingiva. The clinical appearance excluded a plaque related periodontal disease or an ulcer-necrotic gingivitis. The medical history revealed that the patient was diagnosed for Adult Still’s disease (ADS) in September 2012. ADS is an inflammatory disorder characterized by quotidien fevers, arthritis and a number of nonspecific hematologic findings, including leukocytosis. The patient was initially treated with systemic corticosteroid and in February 2013 she started therapy with azathioprine; 20 days after, painful oral lesions appeared and rapidly progressed to the all oral cavity.
Blood exams revealed leukopenia (3.66/mmc), and in accordance with Rheumatologist, azathioprine was immediately discontinued and patient started therapy with amoxicillin. Twenty days after azathioprine suspension significant clinical improvement was noted and blood exams showed a mild leucocytosis (10.80/mmc).

**Conclusion.** The gingival crevice is the main source of leukocytes and in leucopenic conditions can stimulate bacterial infections that rapidly progress. Haemorrhagic diffuse gingival ulcers associated with a decrease in the number of circulating leukocytes need for a multidisciplinary approach to prevent potential systemic complications.

**References**

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**Case report**

**Medication-related osteonecrosis of the jaw (MRONJ): a denosumab case report**

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New cancer targeted therapies have been implicated in the development of osteonecrosis of the jaw; several cases associated with antiresorptive (denosumab) and antiangiogenic drugs have been reported. The American Association of Oral and Maxillofacial Surgeons proposed changing the term of biphosphonate (BP)-related (BRONJ) with medication-related osteonecrosis of the jaw (MRONJ)¹ and the American Dental Association recommended the concept of antiresorptive drug-related ONJ².

Denosumab, a RANKL inhibitor monoclonal antibody, has been approved as antiresorptive agent for osteoporosis and to prevent skeletal-related events in patients with bone metastasis from solid tumors. In July 2015 a 47-year-old woman with bone metastasis from breast cancer was referred by her dentist for still pain and persisting not healing socket after tooth extraction performed in April 2015. An area of exposed bone with empty socket was present in the site of the first upper left molar. She had never received biphosphonates but she was treated with denosumab by subcutaneous injection 120 mg every 4 weeks from January 2014 to March 2015, suspended 40 days before extraction. Antibiotic therapy succeeded in relief of symptoms but the exposed bone persisted. Denosumab has a shorter half life (25.4 days) compared with BP and is completely eliminated in a relatively short time; so drug holiday may be effective before bone surgery but there is still lack of data. Nevertheless denosumab seems to be associated with a higher or at least similar risk of ONJ than zoledronic acid³ and due to the better tolerability its use is increasing. Such patients should be referred to a competent dental practitioner to assess and reduce the risk of MRONJ before starting denosumab therapy³.

**References**
Case report

Non-surgical management of stage III medication-related osteonecrosis of the jaw: a case report

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Background. Surgical treatment of MRONJ, especially in presence of exposed necrotic bone or in presence of a fistula that probes to bone (stage 2 and 3) can be quite challenging, and subject of discussion. We describe the non-surgical management of a patient with a Stage 3 MRONJ.

Case details. A 80-year-old woman was treated for 8 years with alendronate for osteoporosis and developed MRONJ after extraction of maxillary teeth. The computed tomographic (CT) imaging were consistent with the clinical diagnosis of MRONJ, showing extensive osteonecrosis of the left side of the hard palate, and the medial and inferolateral walls of the maxillary sinus. The extensive oroantral fistula was infected and, in view of the age and morbidity of the surgical approach was decided conservative management. Initial therapy has predicted amoxicillin + ac. clavulanate (1000 mg orally three times for 10 days) and chlorhexidine rinse 0.12% three times daily for 30 seconds. Adjunctive treatment with subcutaneous teriparatide at a dose of 20 μg per day for 2 months. After 6 weeks of treatment, her symptoms resolved, with improved of the fistula and infection. Our patient clinical condition was assessed as stage 3 according to the American Association of Oral and Maxillofacial Surgeons (AAOMS), which would support an aggressive surgery, but in this case, a primarily non-surgical approach has been successful in the management of symptoms avoiding morbidity related to surgical treatment. The literature highlights that about 71-80% of the MRONJ patient has improved or remained asymptomatic with a non-surgical approach. Teriparatide could successfully be used as adjunct therapy for osteonecrosis in osteoporotic patients, because it has an anabolic effect and potential role in accelerating bone healing.

References

Case report

Hairy leukoplakia: an early sign of HIV infection and evolution after HAART therapy

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The association between oral lesions and HIV infection is well known. Several studies have shown that oral candidiasis, hairy leukoplakia, necrotizing ulcerative periodontitis and the Kaposi's sarcoma often are the primary sign of HIV infection.¹ These lesions reveal a compromised immune system typically with the count of CD4 lymphocytes below 200/mm³.² A 28-year-old man was referred by his doctor to our Department for painless white oral lesions. The patient reported good health status; he had quit smoking since 4 years. The intraoral examination showed white lesions on both the lingual margins. These lesions were not removable, perpendiculars to the lingual margin, slightly elevated and not ulcerated. No local teeth traumatism was detectable.
The clinical hypothesis of hairy leucoplakia was confirmed by histological investigation. The blood tests were positive for HIV1 strain with a concentration of 550,000 copies/ml HIV RNA molecules and a value of CD4 cells of 106/mm³. The patient was referred to the dedicated Department of our Hospital and he started the HAART therapy.

In the following oral cavity follow-up the lesions on the lingual margins progressively reduced up to completely disappear, the CD4 count increased to 514/mm³ and HIV RNA copies turned down to 3/ml³.

References

Case report

Oral ulceration in a patient with agranulocytosis

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Granulocytopenia (an neutropenia) is a significant reduction in the absolute count of circulating neutrophils in the blood (absolute neutrophil count, ANC) and, in particular, the term agranulocytosis is used when ANC drops below 500 cells/µl. Most patients have a secondary neutropenia caused by infections, drugs, tumors or hypersplenism, although several further factors can also contribute to develop this disorder, as decreased bone marrow production and increased destruction cells by immune mechanism. Key clinical features of such condition are bacterial infections of mucosal and skin surfaces usually accompanied by fever.

A 68-year-old male patient suffering from agranulocytosis was referred by his hematologist to our Oral Medicine Service (UO Odontostomatologia II, Clinica Odontoiatrica, University of Milan) because of a large ulcer (3x2 cm), in the mucosa of left upper lip which appeared concomitantly to fever. Patient’s ANC was of 100 cells/µl. The painful lesion was hard at palpation with growing borders, apparently infiltrating. The clinical presentation was suggestive for a plethora of clinical diagnoses, including neoplasia and infection. Therefore, two biopsies of the lesions were performed, and antibiotic therapy with amoxicillin 3 g/die for 6 days was prescribed, together with chlorhexidine 1% gel applications. After 15 days, the patient was almost asymptomatic with a strong improvement of the lesion (resulting in about 8 mm of diameter). The histology report showed a chronic inflammatory tissue with necrotic areas and epithelial hyperplasia. The infiltrate was composed of lymphocytes (predominantly B cell phenotype), plasmacells, histocytes, with the complete absence of granulocytes. After two months, no oral lesion was detectable.

Painful oral ulcerations and severe periodontal lesions are common findings in patients suffering for neutrophil disorders; they may be even the initial or the unique manifestation of cyclic neutropenia.

References

Case report

Oral pigmentation in Addison’s disease

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A 23-year-old man presented to our observation for progressive hyperpigmentation of oral mucosa. Three years before he had been diagnosed with Hashimoto’s thyroiditis and he was receiving replacement therapy with levothyroxine.
Since the last 8 months, he has been suffering severe fatigue and weakness, orthostatic hypotension, ongoing weight loss (8kg) and hyperpigmentation of the skin unrelated to sunlight exposure.

Intraoral examination revealed multiple, flat, brown-pigmented lesions involving buccal and alveolar mucosa, gingiva, tongue and hard palate. Skin examination revealed diffuse cutaneous hyperpigmentation. While most of the symptoms and signs were non-specific (fatigue, weight loss), hyperpigmentation is a characteristic sign of chronic primary adrenal failure.

The suspect of primary adrenal insufficiency was confirmed by laboratory findings (simultaneous low levels of morning serum cortisol and high levels of serum corticotropin with Synacten test). Hyponatremia and a tendency to hyperkalaemia were also observed as expression of mineralocorticoid deficiency.

The patient started a three-times daily regimen of hydrocortisone with symptoms/signs reduction in 6 months.

The most prevalent cause of primary adrenal insufficiency is autoimmune Addison’s disease (AAD). AAD can arise as isolated condition or within an autoimmune polyendocrine syndrome (APS). The clinical manifestations of AAD result from deficiency of adrenocortical hormones (aldosterone, cortisol, androgens). Deficiency of cortisol promotes production of corticotropin (ACTH), from a precursor (pro-opiomelacortin) containing other active peptides as melanocyte-stimulating hormones (MSH), resulting in hyperpigmentation.

**Conclusion.** Multiple oral pigmentation are often due to systemic diseases or drug intake. A correct interpretation of clinical features as the timing of the onset or the distribution of pigmentation on mucosa and/or skin represents a pivotal step in the diagnostic process.

**References**


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**Case report**

**Differential diagnosis and management of a recurrent single ulcer: a case report**

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**Background.** Ulcers and erosion of the oral mucosa can be the final common manifestation of several conditions from epithelial damage resulting from an immunological attack, as in pemphigus, pemphigoid, lichen planus, to damage because of an immune defect, as in HIV disease and leukaemia. However it may be caused by chronic infections or neoplasia.

**Case report.** A 52-year-old woman, referred at the Oral Pathology Department of the Second University of Naples, presented with a 1-month history of ulcerated lesion, sore, with uneven edges, on the buccal mucosa of the right lower area along a partially edentulous area, distal to the element 45, in correspondence of the vestibular flange of the prosthesis. The patient was a former smoker with positive anamnesis for type 2 diabetes, hypertension and hypothyroidism. The patient also reports having hypersensitivity to metals (nickel).

The control after two weeks, where the patient does not have worn the prosthesis, showed a lesion which appears clinically in healing phase. After one month, however, the patient returns to our attention with the same ulcerated lesion objectified in first visit. The clinical appearance of the lesion does not allow to make a diagnosis therefore, after informed consent of the patient, we performed an incisional biopsy. Direct immunofluorescence examination was carried out. Moreover the following haematochemical tests were required: research of autoantibodies anti DSG1, DSG3 and anti BP180.

**Conclusions.** A single ulcer persisting for more than 3 weeks without signs of healing must be taken seriously, as it could be a neoplasm. Differential diagnosis should be done between a spectrum of conditions from immunological disease to oral carcinoma. The persistence of the lesion may indicate that the ulcer is caused by neoplasia, chronic trauma, chronic skin disease or chronic infection. Another important feature is that only one ulcer is present, since malignant tumours usually cause a single lesion.

**References**

Paracoccidioidomycosis of the oral cavity: a case report

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Paracoccidioidomycosis (PCM) is an endemic deep infection in the Latin continent, particularly in Brazil, Colombia, Argentina and Venezuela. It is caused by a thermally dimorphic fungus called Paracoccidioides brasiliensis. The contagion occurs by inhalation of the fungal conidium forms which are present in the soil and usually the lungs are the first site affected. In most cases, PCM shows pulmonary involvement, lymphadenopathy and mucocutaneous lesions. We report a case of a 54-year-old man coming from Venezuela, who presented in January 2013 in our Clinic in Florence because of 1-month history of oral pain. No other clinical signs and symptoms were reported. The patient said that he was a car pilot and used to drive a motorcycle exposing himself almost every day to dust. Intraoral examination revealed an exophytic ulcerative mass with a granular aspect located on buccal gingiva of the left lower molars mimicking a squamous cell carcinoma. A similar lesion was also present on the adjacent buccal mucosa. A biopsy of the gingival lesion was performed. Histology revealed a pseudo-epithelioid hyperplasia, granulomatous infiltrate with numerous multinucleated giant cells containing small spherical structures indicating the presence of fungal spores. Immunohistochemical stains were positive for PAS and methenamine silver nitrate (Grocott-Gomori). The clinical features, histopathological examination and immunohistochemistry were characteristic for PCM. Antifungal therapy with itraconazole was administered for 2 months and complete healing of the oral lesions was obtained. Oral PCM, although is an extremely rare lesion, should be included in the differential diagnosis also in non-endemic areas especially when patients use to live in exotic countries.

References


Intraosseous onset of oral low-grade mucoepidermoid carcinoma

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Background. Primary mucoepidermoid carcinoma (MEC) of the jaw is a rare lesion, comprising 2-3% of all MEC reported in literature. The most common site of occurrence is the premolar-molar-angle region of the mandible. The main symptoms are swelling and pain with trismus. Radiographically, MEC usually appears as a well circumscribed unilocular/multilocular radiolucency. It shows variability in clinical behaviour, so it is possible to distinguish a low-grade and a high-grade form. Marginal resection is the gold standard of treatment for low grade MEC.
Case report. A 45-year-old female patient referred to the Complex Operating Unit of Odontostomatology of the Polyclinic of Bari showing a painful swelling in the right posterior mandible of one year duration. The OPT highlighted the presence of a multilocular radiolucency in the right angle region of the mandible. The TC showed an ipodense lesion with reduced thinning of buccal and lingual cortical bone. Under local anaesthesia, FNAB was performed. The specimen was sent for histological examination. Microscopically, the lesion was characterized by squamous cells, mucous cells and ones of intermediate type organised in cystic spaces with Alcian blue and PAS positive content. Thus, a diagnosis of intraosseous parodontal low-grade MEC was made. Under general anaesthesia, the patient underwent a marginal resection of the mandible with piezosurgery of the cavity in order to remove epithelial remnants. One year follow up revealed the complete healing with no evidence of recurrence or nodes metastasis.

Conclusions. MEC of the mandible is a rare tumour of unknown aetiology. Very important is the radiologic differential diagnosis and the pre-operatory FNAB in order to achieve a certain diagnosis and then choose a correct surgical approach.

References

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Case report

Extensive bimaxillary involvement, by giant central cell granuloma, in a patient with Noonan Syndrome

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Noonan Syndrome (NS) is an autosomal dominant disorder characterized by short stature, facial dysmorphism, and a wide spectrum of congenital heart defects. Sometime, patients with NS develop multiple giant cell lesions of the jaw or other bony or soft tissues¹. Here, we report a case of a 27-year-old woman affected by the Noonan Syndrome with a central giant cell granuloma involving extensively the maxilla and the mandible. The patient arrived to our structure with a diagnosis of Noonan Syndrome made by a medical geneticist during the childhood. Clinical examination revealed moderate hemi-facial asymmetry, secondary to an expansive mandibular growth, covered by intact mucosa. In addition, it was possible to see: small jaw, a depressed nasal root, a short nose with broad base, short stature and short neck. The patient had undergone the excision of multiple maxillary lesions from the age of 8 to 22 years. Radiographic examination showed multiple, large, well-defined multilocular radiolucent lesions bilaterally in the posterior region of the mandible. The CT-scan confirmed the bilateral mandibular involvement and revealed the presence of multiple multilocular lesions extending to both maxillary bones, with remodeling of the adjacent bony structures, lytic bone destruction as well as intralesional mineralization and extrusion in the right nasal fossa. The complete resection of the lesions required a total bilateral maxillectomy with preservation of the orbital floors. The reconstruction was performed with a revascularized angular scapular and latissimus dorsi free flap. The bone of the scapular angle reconstructed the palate vault, the latissimus dorsi myocutaneous component of the flap filled the bilateral maxillary bilateral death spaces, to give support to the cheek soft tissues. The histological examination confirmed the diagnosis of giant cell granuloma²-³.

References
An uncommon intra-oral psoriasis case

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Psoriasis is a chronic inflammatory skin disease characterized by complex alterations in epidermal growth and differentiation, leading to a cells turnover eight times greater than normal. Oral lesions are extremely rare\(^1\), so the prevalence of oral manifestations in patients with cutaneous psoriasis is uncertain. This paper describes the case of a patient with mucosal lesions, clinically and histologically studied, classifiable as a peculiar form of intra-oral psoriasis.

A 39 year-old. man was evaluated for two erythematous lesions in hard palatal and right buccal maxillary mucosa, constantly present since 5 months and asymptomatic. His medical history reveals a skin psoriasis with evident cutaneous plaques on the scalp and arms. No allergies and drugs consumption declared, stating he started a vegan diet.

First diagnostic hypothesis was plasma cell stomatitis, but no allergic substance was considered responsible for that. After an incisional biopsy, an histological and direct immunofluorescence (DIF) exam were taken. Negative results of DIF exclude autoimmune diseases, as lupus erythematosus or “atypical” *Pemphigus vulgaris*. The clinical suspect of Psoriasiform mucositis was confirmed by microscopical examination.

The scientific literature on oral psoriasis is based only on case reports and lacks solid criteria with which to definitively diagnose a lesion as being oral psoriasis\(^2\). Despite the absence of well-defined criteria, the clinicians should first exclude oral inflammatory diseases similar to psoriasis manifestations and secondly consider the case as an oral psoriasis and treat it if necessary.

References

A new auto-fluorescence guided surgical approach for osteonecrosis performed with Er:YAG laser: a clinical and histopathological case

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Background. Identification of necrotic bone margins during osteonecrosis removal is a major difficulty for surgeons. Ristow suggested that also in the absence of tetracycline labelling, vital bone could be highlighted on the basis of its Auto-Fluorescence (AF) whereas necrotic bone lost such an AF appearing very dark. The aim is to describe a new surgical approach for medication related osteonecrosis of the jaw (MRONJ) that couples the advantages of the Er:YAG laser and the usefulness of the AF in highlighting surgical margins.

Case report. A 65-year-old female osteoporotic patient was diagnosed with a Stage II non-exposed mandibular osteonecrosis. Computed tomography scans showed a necrosis involving vestibular and lingual plates. Surgical treatment was performed under local anaesthesia and without preoperative tetracycline labelling. After bone exposure, VELscope™ system was used to induce and visualize bone AF. Necrotic bone showed no or only pale AF. After removal of necrotic bone block AF was used to guide marginal osteoplasty. According to the AF image obtained after osteoplasty, Er:YAG was used for evaporation of necrotic bone. It produces microspots stimulating vasculariza-
tion and attachment of soft tissues. The minimally invasive action of Er:YAG laser acts in the areas where non-or hypo-fluorescence has been displayed. Histopathological evaluation of removed bone was performed and two specimen of hyperfluorescent bone adjacent to necrosis were taken.

**Results.** Histopathological analysis revealed necrotic tissue in the bone block removed and normally structured bone tissue in the hyperfluorescent specimens. After 10 months follow-up no sign of osteonecrosis was detected.

**Conclusion.** AF guided surgical approach performed with Er:YAG laser has not been previously reported. Taking into account advantages associated with the Er:YAG laser and the effectiveness of AF in highlighting surgical margins, this approach would probably allow to achieve excellent outcomes.

**References**


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**Case report**

**Epulis granulomatosa developed in post-extractive alveolar**

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**Introduction.** Epulis granulomatosa is a post-surgical lesion emanating from an extraction socket. It can be misdiagnosed with lesions of similar appearance, for example, foreign body or pyogenic granulomas, or as a herniation of the maxillary sinus. The diagnosis of epulis granulomatosa should be made only after clinical, radiographic, and microscopic studies have been performed to rule out a malignant process. The most common treatment is surgical excision.

**Case report.** A 37-year-old man referred to our institute for an ulcerative lesion in the site of 3.6. The first molar was extracted two years earlier. The patient had no pain and he was treated with antibiotics. An excisional biopsy of the lesion was performed. Histological examination revealed the present of an epulis granulomatosa. The patient was recovered completely.

**Conclusion.** The diagnosis of epulis granulomatosa was made through the history of earlier extraction and histological examination because epulis granulomatosa is similar to other lesion, but it is a post-surgical lesion.

**References**


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**Mandibular torus as known ONJ risk factor:**

**a case report of osteonecrosis of the jaw**

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**Introduction.** Osteonecrosis of the jaw (ONJ) is a rare but serious complication linked to exposure to antiresorptive and antiangenic drugs, especially aminobisphosphonates.
Up to date, many drug-related, systemic and local risk factors are known, even if many researches are in progress in order to identify and define pathogenesis. ONJ is often associated to oncological patients (one of the systemic known risk factors), to intravenous administration (one of the drug-related risk factors) and to dental extraction (the most frequent local risk factor).

**Methods and Results.** In May 2015, a 78-year-old female patient was referred to our department because she could no longer worn her prosthesis for swelling of anterior mandible. She referred to be surgered for a breast cancer in 2003, and no chemotherapy was added. From 2008, she has been treated with weekly alendronate for osteoporosis. No other known local and systemic risk factors for ONJ were reported. During intraoral examination, two mandibular pronounced tori were present. The right one showed bone exposure. No signs of local inflammation were detected and no symptom was referred. After CBCT scans, the bone necrosis was classified as stage II A, according to Bedogni et al.1

Systemic antibiotic (amoxicillin/clavulanic acid orally twice daily for 8 days and metronidazole 250 mg per os twice daily for 8 days), local antiseptics (chlorhexidine 0.2% mouth rinses and 0.5% chlorhexidine gel) were administered. The patient was referred to Oral and Maxillofacial surgery for surgical management.

**Conclusion.** ONJ pathogenesis still remains not fully understood. Although dentoalveolar surgery is the most common local ONJ risk factor, more attention should be given to concomitant presence of other local features (i.e. torus and prosthesis): protruding cortical bone is coverage with thin mucosal favoring a greater risk for necrosis especially if a local additional trauma (e.g. ill-fitting dentures) is detected.

**References**

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**Mucosal leishmaniasis with localized oral involvement**

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**Introduction.** Leishmaniasis is a group of diseases caused by different protozoan species of the genus *Leishmania*. Clinical manifestations can be divided into three forms: Cutaneous, Mucocutaneous and Visceral. Mucocutaneous Leishmaniasis with localized oral involvement is quite uncommon and may represent a challenging diagnostic dilemma.

**Case presentation.** A 45-year-old Caucasian female was referred by her dentist because of the presence of a painful lesion arisen a year before on maxillary gingiva and extended to upper lip mucosa. At oral examination lesions appeared as weakly red and mildly granulated. Medical history was negative for systemic diseases. Incisional biopsy for both microscopic evaluation and Direct Immunofluorescence (DIF) was scheduled. Ziehl Neelsen and Giemsa stained sections revealed non necrotizing granulomatous chronic inflammation; DIF was negative for IgG, IgA, IgM, C3 and fibrinogen. Among several diagnostic hypothesis the Pathologist hinted at a rare but possible Leishmania infection. DNA real-time PCR tests performed on biopsy specimen amplified a segment of the small-subunit rRNA gene of Leishmania confirming the diagnosis of Leishmaniasis (Wortmann et al. 2001). Furthermore, anti-leishmania antibodies specific for *L.infantum* spp. were detected in serum through ELISA. Patient was negatively interrogated for recent travels abroad. Negative results from Chest X-ray and blood samples excluded visceral involvement. Treatment with amphotericin B achieved in a 3 months' time remission in the area of lips and a partial improvement in upper gingiva.

**Conclusion.** Leishmaniasis presenting with non-specific signs of mild redness and fine granulation can be a challenging diagnosis, because a wide spectrum of diseases may mimic similar manifestations. Nevertheless, leishmaniasis should be included in the differential diagnosis of suspicious chronic oral lesions, even in immune competent patients from non-endemic areas.

**References**
Eosinophilic granuloma: a case report

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Introduction. Eosinophilic granuloma (EG) is the most common lesion in the spectrum of disorders under the classification of Langerhans cell histiocytosis. It is the monostotic form of the disease, with the head and neck region representing the most common site of initial presentation. In this report, we describe a case of EG arising from calvarial bones and then involving the jaws.

Case presentation. A 31-year-old man was referred to our Department 3 years ago. He reported a diagnosis of EG in the temporal and parietal bones in 2004 and of insipidus diabetes in 2009. He complains about the loss of several teeth in the last 6-7 months. Clinical examination underlined poor oral hygiene, gingivitis, lack of several teeth. Radiographic examination showed widespread radiolucencies in the right mandibular body and a massive alveolar bone loss on the left one.

Patient underwent Total Body Scintigraphy and only skull involvement was observed. An incisional biopsy of soft and hard tissue of jaw was performed.

Results and Conclusions. Histological analysis revealed proliferation of big cells with irregular margins, abundant eosinophilic cytoplasm and kidney-shaped or oval nuclei arranged in cords and surrounded by other inflammatory cells. The presumptive diagnosis of EG of the jaws was confirmed. Operative procedures consisted of extraction of compromised teeth and curettage of bone lesions. Furthermore, patient underwent chemotherapy with temporary resolution of the disease three years thereafter.

An accurate anamnesis and histological examination after a biopsy are decisive for a correct diagnosis. Clinical and radiological follow up are mandatory to exclude recurrences and to plan a future prosthetic rehabilitation.

References


A diagnostic approach to a particular gingival lesion

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Necrotizing Ulcerative Gingivitis (NUG) is an acute gum infection, characterized by painful ulceration inflammation and flattening of the interdental papillae, that are necrotic and no longer visible, and a more rapid development than other forms of gum diseases. NUG diagnosis is based on three essential symptoms: sore bleeding gums, ulceration and necrosis of the interdental papillae. Patients have generally compromised immune system, malnutrition, poor living conditions, stress, HIV. Fever, malaise, cervical and submandibular lymphadenopathy are commonly detected. A 40-year-old male patient, back from Tanzania, reported pain in the anterior maxilla. The patient had not systemic diseases. During clinical examination, it was noticed a wide swelling, with crater-like depression mixed with purulent necrotic area, painful, on both buccal and palatine gum. An oral swab revealed positivity for Candida albicans. An ultrasonic treatment was immediately carried out. A gel containing sodium hyaluronate (Aminogam gel®, Errekappa, Italy) was administered twice a day for 15 days together with rinses with 0.2% chlorhexidine (Corsodyl®, GSK, Italy).
twice a day for 15 days. After two weeks, a decrease of pain and significant regression of the lesions were recorded.

After one month, mucosal healing was detected. NUG diagnostic hypothesis was reached thanks to anamnestic data, which described an acute onset of the lesions and symptoms. Moreover, the ultrasonic treatment, removing the etiological local factors, allowed us to reach a diagnosis thanks to the good response and rising observed in the gums. Chlorhexidine increased the antibacterial and antiplaque activity and sodium hyaluronate helped the wound healing. NUG is a rare oral disease, that needs an accurate diagnosis and therapy in order to reduce the symptoms restoring quickly the healthy status of patients.

References


Case report

Melanoma of the oral cavity: a case report

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Melanoma is a malignant neoplasm of melanocytic origin. Cutaneous and mucosal melanomas show distinct etiology, behavior and molecular biology. Oral melanoma is an extremely rare tumor and comprises much less than 1% of all melanomas and about 0.05% of malignant oral tumors. Oral melanoma affects male adults in their 5th to 7th decade of life and it is more common in Blacks and Japanese people. The palate and the maxillary gingiva are the most common sites. The prognosis is poor: the 5-year-survival rate ranges between 15 and 38%. We report a 51-year-old female with a 2-month-history of progressively enlarging palatal growth which was a source of pain, burning sensation and bleeding. Clinical examination revealed a grey-brown sessile mass located on the upper right alveolar ridge extending into the palate, measuring about 3 x 5 cm, and a separate brown-dark patch on the adjacent alveolar mucosa. Histology of the sessile lesion revealed a widely ulcerated neoplasm formed by epithelioid cells involving the lamina propria; the immunohistochemical staining were positive for S100 protein, HMB45, MART1, and MIB1 suggesting a diagnosis of nodular melanoma. For the patch lesion, histopathology examination and immunohistochemistry (which was positive for HMB45 and protein S100) revealed a melanoma in situ. The patient underwent to complete surgical resection of both lesions. Unfortunately, the patient died of disseminated disease after 6 months from the initial diagnosis.

References


Case report

Haemorrhagic bone cyst of the mandible: a case report of remineralization

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The haemorrhagic-aneurysmal bone cysts of the jaw are rare not neoplastic bone lesions (2% of all bone cysts)¹. The most frequent clinical sign is a symptomless or painful at touch bone swelling. They are not invasive lesions con-
taining blood; at X-ray imaging they present as a radiolucent cavity with marked irregular perimeter. The pathogene-
sis is not known but the most relevant hypothesis is traumatic, probably causing an hematic extravasation in the
bone tissue\(^2\). The aneurysmal bone cysts are generally classified as pseudocysts because they don’t have an ep-
thelial lining. The clinic-instrumental evaluation associated with a biopsy provides diagnosis. In difficult cases a fine
needle aspiration or an angiographic study can help in getting the correct diagnosis.
A 17-year-old male patient was referred to our Department by his dentist for a radiolucent lesion in the area of
mandibular incisors, revealed by a routine orthopantomography. The medical history was negative for diseases
or therapies. He couldn’t remember any recent trauma and he was completely asymptomatic. No swelling was
detectable; his teeth were all vital and not dislocated. There were no signs of roots alterations or resorption.
In order to get the exact diagnosis we performed a biopsy by surgical bone drilling (5.0 mm). The obtained speci-
men was poor in cells and mainly bloody. The histological examination revealed blood clots and small fibrosis
fragments without epithelial lining, not compatible with keratocyst or ameloblastoma\(^3\). The diagnosis was haem-
orrhagic cyst. The patient was periodically followed-up. After 2 years the X-ray showed the cavity was completely
remineralized.

References

Root resorption caused by osteoma growth

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Introduction. Osteoma is a benign and asymptomatic neoplasm, consisting of well-differentiated mature bone. The
solitary osteoma may be classified as: peripheral when arising from the periosteum, central when arising from the
endosteum and extraskeletal when arising in soft tissue. The etiology of osteoma is still unclear. Some reported cas-
es did have a clear history of trauma; however, this was not the case in the majority of cases. Whereas peripheral
osteomas are fairly easy to diagnose, central osteomas pose a more challenging diagnostic problem and need to be
differentiated from other similar lesions of the jaws, such as central ossifying fibroma, osteoblastoma, cementoblas-
toma and odontoma in cases occurring within toothbearing areas. Osteomas are usually asymptomatic. We reported
a case of the central osteoma associated with partially root resorption of the mandibular first molar.

Case report. A 22-year-old boy referred to our institute with an ortopantomography and CT-scan revealed the pres-
ence of a large and well defined osteosclerotic lesion between elements 4.5 and 4.6. The lesion had caused root re-
sorption of element 4.6. The patient did not report the presence of other systemic pathologies or of trauma in this re-
gion. The surgery conservative approach was preferred in order to avoid permanent injury of the inferior alveolar
nerve and histological examination revealed that the lesion was a benign osteoma of the right mandible.

Conclusion. Osteomas are usually asymptomatic and have a very slow growth rate but we report a rare case of an
extensive osteoma of the right mandible involving the medial roots of the first molar. So corrected diagnosis of this le-
sions is only histological.

References
Unusual histological presence of dysplasia in a case of oral pemphigus vulgaris: a potential diagnostic challenge

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Introduction. Pemphigus Vulgaris (PV) is an immune mediated disorder of the oral mucosa and is not considered a potentially malignant lesion1. In the present report we describe a case of PV with unusual signs of high grade dysplasia in the histology.

Report. A 67-year-old man was referred to the Division of Oral Medicine, University of Bologna, for the appearance of a painful lesion in the soft palate. Intraoral examination revealed a bullous lesion together with a small proliferative area localized in the soft palate. Two incisional biopsies for histology and Direct Immunofluorescence (DIF) were performed, and pharmacological therapy with topical betamethasone (1 mg tablets 3 times/day) and prednisone 50 mg/day was initiated to alleviate symptoms. Histology showed intraepithelial areas of acantholysis associated with high grade dysplasia. DIF revealed intercellular deposits of IgG and C3. Histopathology and DIF were consistent for PV with histological signs of dysplasia. The presence of oral carcinoma or a high risk potentially malignant lesion couldn’t be completely ruled out, so it was decided the surgical removal of the dysplastic area, that was performed one week later the beginning of cortisone therapy. The histological analysis from the excisional biopsy confirmed intraepithelial acantolysis but did not reveal any sign of dysplasia. Pharmacological therapy and regular follow-up were scheduled. Actually the patient had been followed-up for 1 year since the first visit and was still free from malignancies.

Conclusions. Histological signs of high grade dysplasia in oral mucosa are often associated with concurrent or subsequent carcinoma; however histologic dysplasia can be also simulated by the presence of severe inflammation that may induce reactive epithelial cell changes. Pathologic evaluation of dysplasia in an inflammatory disease as PV may be a diagnostic challenge and a careful evaluation is necessary in order to choose the most appropriate treatment.

References

Efficacy of professional oral hygiene procedures and “Fertomcidina-U” in patients with plasma cell gingivitis: a case report

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Introduction. Plasma cell gingivitis (PCG) is a particularly rare condition characterized by diffuse infiltration into the sub-epithelial gingival tissue with a dense, band like plasmacytic infiltrate in the upper dermis. PCG appears as a diffuse and large reddening and oedematous swelling of the attached and free gingiva, with a sharp demarcation along the muco-gingival border. Ulcerations are rare and these lesions are commonly asymptomatic, although some patients could complain of pruritus, burning or pain. Usually of unknown aetiology, even if this condition has been reported as non-plaque induced, effective dentogingival plaque control sometimes could resolve the gingival inflammation. For these reasons, we postulated that efficient plaque control could have been helpful in treating PCG.
Case details. A Caucasian female 70 years old was referred presenting painful and diffuse erythematous gingivitis. A biopsy was performed with PCG evidence. A professional oral hygiene regimen was started to deal with the gingival status. Severity of pain was detailed using a Visual Analogue Scale (VAS). Patient received non-surgical periodontal therapy, including personalized oral hygiene instructions, and thorough supragingival scaling and polishing with the removal of all deposits and staining combined with the use of antimicrobials as Fertomcidina-U. Clinical outcome variables were recorded at baseline and after the intervention and included full-mouth plaque scores (FMPS), full-mouth bleeding scores (FMBS), and the clinical extension of gingival involvement. After finishing the proposed protocol, a significant and statistical reduction was observed for FMPS, FMBS, and clinical gingival involvement.

Conclusion. Professional oral hygiene procedures and non-surgical periodontal therapy, including antimicrobials medication as Fertomcidina-U, were associated with a striking improvement of gingival clinical outcomes in this female with important gingivitis PCG related.

References

Case report

Optical coherence tomography as a simple and non invasive tool for the diagnosis of oral disease: a case report

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Introduction. Optical Coherence Tomography (OCT) is a new biomedical imaging modality that provides high resolution cross-sectional images of tissue. Conceptually, it has been compared with ultrasound scanning but it uses infrared light and records reflections below the surface to produce a cross-sectional architectural image of the tissue. OCT has been applied in ophthalmology and recently in dermatology; several studies have shown the validity of the use of OCT in ex vivo oral lesions but, to date, it does not exist a bank of normative and pathological OCT data of the oral tissues to consent identification of cellular structures of normal and pathological processes. We report a case of fibroma, analyzing in vivo OCT use and comparing data with microscopic evaluation.

Case report. A 44-year-old woman was referred to the Department Surgical, Oncological and Oral Sciences, University of Palermo, with a neoformation on the apex region of the tongue, clinically compatible with the diagnosis of fibroma or papilloma. Firstly, in vivo OCT analysis demonstrated the presence of a white area, due to the presence of hyper-reflective area for collagen storage that is strongly scattering. After, exeresis was performed and histological examination confirmed the diagnosis of fibroma.

Conclusion. Optical coherence tomographic imaging can produce detailed cross-sectional images of tissue of oral cavity and it could be a new non-invasive approach that will help improve the diagnosis and the follow up of oral lesions. The validity of OCT in ex vivo oral lesions is confirmed in literature, while in vivo OCT validity should be supported by comparison of data of several oral disease: further researches are needed.

References
Oral involvement in Paget’s disease as first manifestation: a case report of a diagnosis made by general dentist

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Introduction. Paget’s Disease of Bone (PDB) is a chronic non-inflammatory disease, characterized by an increased bone turnover, leading to trabecular disorganization with a consequent reduction of the strength and quality of the bone.

PDB is the second most common metabolic bone disease after osteoporosis and is more common in the elderly and Caucasian population. The higher prevalence is recorded in the UK, which is around 5.4%. In Turin, the estimated prevalence is between 0.7-1%. Regions of the skeleton involved may be individual (monostotic) or multiple (polyostotic). The jaws are involved in 15% of cases, with a ratio maxilla: mandible = mandible than 2:1.

Case report. A 72-year-old female patient was referred by her general dentist who had noticed considerable lingualized lower incisor and canines and deformation of the mandibular profile, with evident prognathism. Radiographic investigations, in particular the mandibular TC scan, showed increase in size with bone deformation and areas of thickening, coarse and irregular bone rarefaction.

The dentist placed a clinical suspicion of PDB. Subsequent laboratory tests showed increased serum alkaline phosphatase (ALP: 259) and bone alkalin phosphatase (BAP: 49.4), secondary hyperparathyroidism (PTH: 178) and normal hydroxyprolinuria.

The total body bone scintigraphy with 99m Tc showed increased activity of the mandible and the left femur, possible secondary localization of PDB.

Conclusions. To the best of our knowledge, this is the first report, in the literature, of diagnosis of polystotic Paget’s disease of bone by a private general dentist. The modification of dental position and jaws deformity in adulthood may direct the clinician to suspect this disease.

References

An extensive nasopalatine duct cyst: case report

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The Nasopalatine Duct Cyst (NPDC) is a developmental, epithelial, non-odontogenyc cyst arising from degeneration of epithelial remnants of the nasopalatine duct. Radiologic examination demonstrates a well-defined ovoid or round lesion located between the roots of the maxillary central incisors. It is not uncommon to see evidence of endodontic therapy because the NPDC was previously clinically misdiagnosed as a periapical cyst or granuloma.

Aim of this paper is report a particular case of an extensive NPDC of the premaxilla treated with surgical enucleation. A 46-year-old man, with no history of systemic diseases, was referred to our observation complaining of a displacement, on the buccal vestibule, and a dyschromia of the right upper central incisor and to evaluate a midline cystic lesion of the premaxilla detected during a routine clinical and radiographic exam.
Extra-oral examination showed the asymmetry and swelling in the maxillary anterior region and right nasolabial fold. Intra-oral examination showed a tender and painless swelling, covered by normal mucosa, in the midline of anterior buccal vestibule.

Radiologic examination showed a well-defined unilacular radiolucency with corticated margins between the roots of the maxillary central incisors extended on both second premolar teeth, all endodontically treated. The patient revealed that this lesion was previously diagnosticated to be of dental origin by his dentist and the upper right central incisor was treated with endodontic therapy. This is a typical misdiagnosis and mismanagement of NPDC.

Surgical enucleation of the lesion (5x2.5 cm diameter) was performed with no intra-nor post-operative complications. Follow up at 7 days and 1 month showed an increasing primary wound healing and histopathologic examination confirmed the clinical diagnosis of NPDC associated with granulation tissue, due to irritation of periapical area because of inadequate endodontic treatment of teeth included and near to the cyst.

References

Case report

Dental implant during denosumab treatment: a case of medication-related osteonecrosis of the jaw (MRONJ)

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Dentoalveolar surgical procedures are known to be the most important risk factor for developing medication related osteonecrosis of the jaw (MRONJ); current guidelines contraindicate the placement of dental implants in cancer patients treated with intravenous bisphosphonates1. Although there are still uncertain data, dental implantation is not contraindicated in patients who have been taking the less potent oral bisphosphonates for under three years and don’t present other risk factors. In a recent meta-analysis the overall incidence of MRONJ in cancer patients receiving denosumab was 1.7%; factors associated with MRONJ occurrence were dental extraction, chemotherapy, poor oral hygiene, dental appliances and antiangiogenic therapy but no data about implant treatment were available2. In implant treatments not only the surgical procedure itself could be related to an increased risk of MRONJ but also repeated peri-implant inflammation could lead to osteonecrosis3.

In July 2015 a 64-year-old woman with breast cancer was referred by her dentist for necrotic bone area in the site of dental implant rehabilitation persisting since January 2015. She had never received bisphosphonates but her oncological treatment was denosumab by monthly subcutaneous injection 120 mg from July 2013 without any suspension and still on-going. The clinical examination showed an area of exposed necrotic bone surrounding the dental implant that replaced the first right upper molar; a computed tomography confirmed it. The dental implant was placed in June 2014 in the healthy edentulous site of the first molar. The healing cap was put in October and no sign of infection was detectable both clinically and radiographically. The implant was loaded in December 2014.

Beyond the current guidelines on implants placement in patients taking antiresorptive medications dental practitioner should be aware of the importance of long-term oral hygiene and control of implant-prosthetic restorations.

References
Granular cell tumor of mouth’s flor: a case report

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Background. The granular cell tumor, also known as Abrikossoff’s tumor, granular cell schwannoma, is a very uncommon benign neoplasia which affects soft tissues. This lesion was first described in 1926 by Aleski Abrikossoff. It shows a predilection for the oral cavity. The histogenesis is uncertain and debated and recent studies pointed out a derivation from Schwann cell or neuroendocrine cells. Approximately 50% of all lesions arises in the head and neck and over half of these is localized in the tongue. They arise in all age groups, with a peak between the 5th and 6th decades. In about 10-20% of patients, the lesions are multiple. The ratio of incidence M/F is 2:1.

Case summary. A 55-year-old female patient presented to our Department with a chief complaint of a swelling on the right side of mouth floor since 1 month. The swelling was initially small and a gradual increase in size was observed. On examination there was a solitary well-defined, tender and non-fluctuant swelling with smooth borders, localized over the right lingual caruncola, measuring approximately 4 × 3 mm. It was supposed to be a gallstone of the sublingual gland, but ultrasound scan of the salivary glands and radiographic exams were both negative. An incisional biopsy was performed. The hematoxylin and eosin-stained sections revealed cellular proliferation of polygonal cells with abundant granular cytoplasm. No mitotic figures were observed. A diagnosis of granular cell tumor was rendered. A panel of markers were used: S-100, vimentin. Positivity was observed with vimentin, S-100.

Conclusion. Benign mesenchymal neoplasms usually present as a swelling on the tongue. This lesion though uncommon should be considered in the differential diagnosis of benign and malignant swellings of the oral cavity. Considering the aggressiveness of the granular cell lesions, a regular follow up of the patients is mandatory.

References
Case report

Oral focal mucinosis (OFM): a case report

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Aim. Oral focal mucinosis (OFM), classified as the oral variant of dermal focal mucinosis or myxoid skin cyst, is a rare disease characterized by the myxoid degeneration of connective tissue in localized areas of the oral mucosa. The etiology is unknown while the pathogenesis is related to overproduction of hyaluronic acid by the fibroblasts during collagen production, resulting in a focal myxoid degeneration. More common in young women, it appears as a sessile or pedunculated lesion, asymptomatic, covered by normal mucosa. The most common site is on keratinized mucosa overlying the bone (i.e. gum 80% and hard palate 20% of cases respectively); other less frequent sites are alveolar mucosa and tongue.

Case report. In June 2015, a 48-year-old man, strong smoker with poor hygiene conditions, came at observation for the presence of a sessile, firm, well-defined mass measuring 3.0 cm at its greatest dimension sited on the masticatory mucosa of the first sextant, partially covering the crown of the tooth 1.7, 1.6 and 1.5. The lesion appeared, according to the patient, two years before, it was sized approximately 3 cm, covered by a normal mucosa and had a hard-fibrous consistency at palpation. TC scans showed no alterations in the underlying bone or in the adjacent teeth. The neoformation was excised with a quantum resonance scalpel and histological examination showed the presence of myxoid tissue with spindle and stellate cells without atypia and mitosis (Ki67 expression about 2%), immersed in myxoid matrix alcian bleu positive. The histology is compatible with the diagnosis of OFM.

Conclusion. OFM in an uncommon clinico-pathological condition representing a diagnostic challenge: it has no characteristic clinical features and is commonly diagnosed as other more frequent oral tumors (i.e. fibroma, pyogenic granuloma and epulis). The definitive diagnosis is based on histopathological examination, which is always required after exeresis.

References


Case report

CO2 laser and soft tissue management in oral cancer patients

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The laser is a very effective device, also for post surgical management of soft tissue in oral cancer patients. CO2 laser, in particular, is characterized by high power, wavelength range between 9.600 and 10.600 nm, high affinity for...
water and achieving its action mainly through the photothermal effect without contact. Due to the undoubted advantages associated with the usage of CO2 laser in oral surgery of soft tissues, this equipment can be used in the management of any complications in patients who had surgical operations for oral cancer. Specifically, we present two cases treated at MoMax division (Department of Oral Sciences and MaxilloFacial Surgery, “Sapienza” University of Rome) by CO2 to excise reactive hyperplastic inflammatory lesions that have been diagnosed histopathologically as a pyogenic granuloma. The first case, the predisposing factor of the pyogenic granuloma was a chronic mechanical trauma of the tooth 2.7 with a medical history of a maxillofacial intervention (4 years before) for removing of a giant cell tumor in the left side of the mandible and contextual reconstruction of it; the second case was a chronic irritation of gingival tissue in the right side of the mandible near to osseo-integrated implants inserted after a maxillofacial surgery for the removal of an ameloblastoma in the same side of the mandible followed by simultaneous reconstruction (4 years before). The CO2 laser, used for treating these clinical cases, showed multiple advantages such as: rapidity in surgical execution, control of hemostasis (precious cutting without suture), intra and postoperative comfort, high quality of healing and low risk of relapse for patients as a consequence of a poor secondary proliferation of collagen and capillaries.

References

Case report

Slow growing cheek swelling resulting in a minor salivary glands pleomorphic adenoma. Report of a case

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Pleomorphic Adenoma (PA) is a benign mixed neoplasm that represents the 3-10% of head and neck tumors; particularly, in parotid and submandibular glands, frequency varies from 44 to 68%. It commonly occurs between the 3rd and 5th decades with M/F ratio 1:1.2.

The minor salivary glands are involved in 39% cases, mostly on palate (10%); buccal mucosa manifestations represents the 5.5%. Clinical aspect is a painless, slow growing submucosal swelling, initially movable, becoming firm on tissues with growing. In July 2014, A 42-year-old. Philippine man was referred to our service to investigate a slow growing and painless swelling of the right cheek of several years’ duration. No history of fever, bleeding, sensory changes, trauma and ulceration was reported. Anamnesis excluded pathological systemic conditions and drugs assumption. Family history excluded granulomatous conditions. Investigation was executed by ultra-sonographic exam, showing hypoechoic, well-circumscribed and distinguishable 5x2x2 cm mass. Therefore, differential diagnosis was against lipoma (20% cases hypoechoic) and other benign conditions, e.g. traumatic fibroma. MRI investigation was performed resulting in low intensity T1 sequences and a rim of decreased signal intensity on T2-weighted images representing the surrounding fibrous capsule. Surgical excision by intraoral approach was performed. No FNA with Us assistance was performed because surgical planning, that consists in radical excision with safe margins, wouldn’t have been affected by findings. Histology was suggestive for PA, characterized by the presence of both epithelial and mesenchymal elements. Epithelial origin cell give rises to ductal structures (stained with H&E), closely intermingled with mesenchymal elements, stromal myxoid change with compressed cord-like myoepithelial elements. Clean and safe margins excision was confirmed. A 3,6-and 12-month- follow-up was set. No clinical and instrumental signs of recurrences were reported.

References
Central giant-cell granuloma involving maxillary sinus. Two years follow-up of surgical management with QMR scalpel and erbium laser

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Central giant-cell granuloma (CGCG) is a benign condition of the jaws affecting twice women and occurring in 20-40-year-old people. The lesions are found more in the anterior part of the maxilla and the mandible. The aim is to report a case of unusual aggressive form of CGCG occurred in a 65-year-old male patient.

A blush 2 cm lesion in the left part of edentulous maxilla was detected by the dentist and firstly diagnosed as prosthetic trauma. After 3 weeks of denture elimination the lesion instead of disappear increased in size. In the oral medicine and laser surgery unit of Parma University an incisional biopsy was performed with Quantic Molecular Resonance (QMR) scalpel. The histopathological diagnosis was giant-cell granuloma (GCG). The CT scan showed a large involvement of bone and maxillary sinus. Surgical excision was performed under local anaesthesia using QMR to cut soft tissues and Er:YAG laser to vaporize bone surfaces. The advantages of these two instruments are the perfect respect of excisional margins for both soft and hard tissues, bleeding control, improving of the healing process. The large oro-antral fistula was closed with a muco-periosteal flap and continuous suture.

At the moment of surgical intervention, 1 month later the biopsy, the lesion size was more than the double (about 5 cm of diameter). The histopathological evaluation of the specimens confirmed the diagnosis of CGCG for both mucosa and bone. At 2 years of follow-up the patient is free of lesion.

The aggressive form of CGCG has rapid growth and resorbs roots and perforates the cortical plate. It also has a high recurrence-rate and can be painful and cause paraesthesia. In this clinical case the lesion showed a rapid progression in few weeks with soft tissues and bone involvement. The surgical excision performed with new technologies (QMR scalpel and erbium laser) achieved intra and post-operative advantages with low invasive but radical procedure.

References

"Morsicatio buccarum, linguarum, labiorum and mucosae oris": a challange for dentistry

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Morsicatio buccarum, linguarum, labiorum and mucosae oris are a group of pathological disorders due to self-induced traumas, known as self-injurious behavior (SIB). They can occur as an isolated action or as a recurrent phenomenon, as is often seen. Several syndromes and congenital conditions have been associated with SIB, including Tourette’s syndrome, XYY syndrome, autism, and sensory neuropathies. Clinically, it is a macerated grey-white patch and plaque of the mucosa caused by external stimuli (self-induced injury) such as habitual biting, chewing, or sucking. It is often confused with other dermatological disorders involving the oral mucosa, which can lead to a mis-diagnosis. Microscopic evaluation reveals marked acanthosis of the mucosal epithelium with an irregular surface, which demonstrates a ragged morphology. The stratum spinosum may show markedly swollen keratinocytes that contain much glycogen. Mild chronic inflammatory infiltration can be seen in the stroma and occasionally reactive
atypia are present if there is ulceration or inflammation. The differential diagnosis of SIB should include other mucosal pathologies of the oral cavity that appear whitish or yellowish, such as oral lichen planus, pemphigus, benign mucosal pemphigoid, candidiasis, leukoplakia, leukoedema, white sponge nevus and chemical burn. Some helpful clinical features are that morsicatio is usually bilateral and located on movable, nonkeratinized mucosa that can be reached by the teeth. Although proliferative leukoplakia may be bilateral and sometimes even symmetric, they will often involve areas that cannot be reached by the teeth (such as the gingiva). If the clinician is not absolutely certain that a white lesion is a morsicatio, a biopsy is always advisable. Authors discuss about two cases: a 82-year-old female presenting a wide bite on the margin of the tongue and a 60-year-old man presenting small ulcerations on the lower lingual surface.

References

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Case report

Laser treatment of a benign oral lesion in a patient affected by vascular parkinsonism: a case report

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Vascular Parkinsonism (VP) is a form of secondary Parkinsonism due to cerebrovascular disease. The principal risk factors involved in VP are diabetes, smoke and in particular hypertension, this latter provoking infarcts of subcortical gray matter nuclei, diffuse white matter ischaemic lesions and large vessel infarcts. Old people are affected with gait difficulties, symmetrical predominant lower-body involvement, poor levodopa responsiveness, postural instability, falls, cognitive impairment, dementia, corticospinal findings, urinary incontinence and pseudobulbar palsy. VP patients are “special needs” individuals and their treatment is often considered problematic. A case of a benign oral lesion in a 76-year-old male affected by VP, has been treated with laser surgery using a 808nm diode laser [SOL®, DenMat Italia, Baronissi (SA)] at 2.5W in CW, with a fluence of 1990.44J/cm² and a 400μm fiber diameter according to the mucosal preservation technique. At clinical exam on the lower right lip it was present a swelling of about 1 cm of diameter. At the end of the treatment the excised sample was sent to the pathologist, in a 10% buffered formalin, for the histological examination. The final diagnosis was lipoma. The patient had no pain after surgery and a complete healing was observed after one month. This case confirmed that the diode laser can be considered an effective and safe device in critical patients thanks to minimal invasion, less intra-operative bleeding, better hemostasis, reduced pain and optimal healing.

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Case report

A metachronous field cancerization of the tongue

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Background. The term “field cancerization” (FC) describes second primary tumors (SPT), independently developed, at multiple sites. SPTs are cancers geographically distinct and separate, not connected by neoplastic epithelial changes from original primary cancer. They can be synchronous (neoplasms that are diagnosed at the same time or within 6 months of primary lesions), or metachronous (lesion develops after 6 months of index tumor).

Case report. A 40-year-old, female patient reported an ulcerated swelling on the right half of the tongue in April 2013; histological examination diagnosed an oral squamous cell carcinoma (G1). After right laterocervical lymph node dissection, radiotherapeutic treatment of right’s lymph node stations of the neck was performed. In January 2015 the patient referred burning on the left half of the tongue and a small area of about 1 cm of desepithelialisation, not bleeding, not ulcerated, surrounded by a small whitish halo was detected. Biopitic examination reported a diagnosis of a well-differentiated squamous cell carcinoma. In May 2015, after Fine Needle Aspiration Biopsy of left laterocervical lymph node, metastasis of well-differentiated squamous carcinoma were diagnosed.

Conclusions. This clinical picture shows all the typical characteristic of a FC. To date it is still not easy to identify this condition until the appearance of the SPT, but recent studies have highlighted that the presence of a field with genetically altered cells is a risk factor which is likely to increase dramatically for the development of a SPT. Moreover, has been demonstrated the value of mtDNA D-loop analysis to better define differential diagnosis between SPT and local recurrences, which is essential to establish a right prognosis and the appropriate treatment. A short follow up and early detection of cancer remain still essential to ensure patient survival and quality of life, and moreover the secondary prevention may warrant a life-long surveillance.

References


Case report

Oral manifestations in severe GVHD: a case report

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Graft versus host disease (GVHD) is a complication of hematopoietic stem cell transplantation (HSCT) and the most common cause of morbidity and mortality unrelated to the malignant primary disease. Immunocompetent T lymphocytes of the donor recognize as foreign the antigens expressed by the recipient cells following transplantation. GVHD affects multiple organs and tissues such as the skin, gastrointestinal tract, liver, in acute or chronic form (cGVHD). The oral cavity may be involved only in cGVHD. Oral manifestations of cGVHD are lichenoid lesions, hyperkeratotic plaques and oral opening limitation which is secondary to tissue sclerosis. The diagnosis is clinical, whereas in the presence only of the distinctive clinical features without disease involvement in other organs, or if malignancy is suspected, an oral biopsy would be indicated. Local corticosteroids are the treatment of choice. Extracoroporeal phoopheresis and systemic corticosteroids constitute the second line treatment. The overall survival rate is about 76% three years after the diagnosis of cGVHD, and 52% after 5 years1, 67% after 6 years in pediatric patients2.

The Authors report a severe cGVHD (overlap syndrome with extensive skin involvement at the time of diagnosis, as well as bowel, liver and oral cavity, Lerner grade II GVHD confirmed by rectal biopsy) in a 13-year-old patient with pre-B-cell acute lymphoblastic leukemia (diagnosis in June 2009) who has received HSCT from a HLA haploidentical donor (mother). BCR/ABL translocation was positive and therapies with imatinib, rituximab, prednisone, cyclosporine and extracorporeal photopheresis were administrated. The latest follow-up exams demonstrated inverted CD4/CD8 T-cell ratio (T-CD4 n/ul 240; T-CD8 n/ul 400) and normal platelet count (245,000 platelets/mcL) with positive prognostic significance3.

Early diagnosis and a multidisciplinary treatment plan are pivotal factors to improve the quality of life in these patients.

References


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**Case report**

### Coexistence of early primary biliary cirrhosis and mucous membranous pemphigoid: fortuitous case or new autoimmune syndrome?

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**Introduction.** Primary Biliary Cirrhosis (PBC) (ICD-10; K74.3) is a chronic liver disease characterized by an immune-mediated inflammatory destruction of small intrahepatic bile ducts, which leads to a progressive cirrhosis and eventually to liver failure. A high-titer of anti-mitochondrial antibodies (AMA) is the serological hallmark of PBC (sensitivity>95%). Mucous Membrane Pemphigoid (MMP) (ICD-10; L12.1) is a rare autoimmune blistering disease, which mainly target different proteins in epithelial basement membranes. We report the simultaneous occurrence of PBC and MMP in a patient with previous thyropathy.

**Case report.** L.M., a 56-year-old woman comes to our attention referring a widespread burning of bilateral buccal mucosa, tongue and gums since 2010. She complains also itching, fatigue, dry eyes, abdominal pain, arthralgia, burning sensation of vagina and rectum, which are typical symptoms of PBC; she reported weight loss of 25 kg in 8 months and recurrent episodes of stools emission associated with mucous without blood. Skin hyperpigmentation (typical of PBC) and itching, painful and erythematous areas were found. Hypertension, osteoporosis, erosive gastritis, aortic insufficiency, chronic asthma, thyropathy, are the most relevant comorbidities. Colonoscopy reveals a non-specific inflammation and ultrasound examination of the abdomen show hepatomegaly.

Clinical features, Nicolsky’s positive sign, direct immunofluorescence and histology on gum biopsy confirmed the diagnosis of MMP. A persistent high-titer of AMA-M2, 1:320 (normal value < 1:40), was found.

**Conclusion.** Although to our knowledge there have been few reports of the coexistence of PBC and pemphigoid, they have both an autoimmune pathophysiological mechanism and are often associated to the same autoimmune disease; this suggests that they may have been interrelated. Reporting this association is relevant in order to investigate immunopathogenetic mechanisms to improve diagnosis, prognosis and treatment.

**References**

Healing of MRONJ after discontinuation of bevacizumab in a patient with sacrum metastasis of colorectal cancer: a case report

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Introduction. Bisphosphonate-Related Osteonecrosis of the Jaws (BRONJ) is a well-known severe complication of the use of bisphosphonates that occurs in the oral and maxillofacial regions. This nomenclature was changed in Medication-Related Osteonecrosis of the Jaws (MRONJ) by the American Association of Oral and Maxillofacial Surgeons position paper in 2014 as there is a growing number of cases of ONJ associated with antiresorptive and antiangiogenic drugs other than BPs such as bevacizumab, denosumab, sunitinib, sorafenib and sirolimus.

Case description. A 54-year-old man diagnosed with colorectal carcinoma with sacrum metastasis referred to Biomedical and Neuromotor Sciences Department, University of Bologna. He was undergoing treatment with Zometa and intravenous bevacizumab when he developed spontaneous osteonecrosis of the mandible, comparable to SICMF-SIPMO stage 2B BRONJ, with moderate pain, pus and mobility of the tooth involved in the ONJ (37). The necrosis was treated with therapies such as oral hygiene, use of topical chlorhexidine, bone debridement, antibiotics (1g amoxicillin every 12 hours in combination with metronidazole 500 mg every 12 hours for 10 days) and extraction of 37, which was completely surrounded by necrotic bone. Such therapies did not improve the patient’s clinical conditions, as the necrosis expanded from 37 region to mesial 36 region and mandibular ramus. 18 months later, the patient discontinued bevacizumab with his oncologist’s permission replacing it with bortezomib and the lesion immediately stopped its spreading, then completely healed after 6 months with a spontaneous bone sequestrum and complete mucosal recovering.

Conclusions. Bevacizumab is a recombinant humanized monoclonal antibody that inhibits VEGF-A, its antiangiogenic properties are associated with an increased risk of developing MRONJ. This case report suggests the possibility that the discontinuation of Bevacizumab, unlike zometa, may have a role in the arrest of ONJ.

References

Micro-histology in the detection of oral cancer: case report

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Oral cancer is a disease of high impact globally. It ranks as the sixth more frequent one among all types of cancer. The scalpel biopsy conventionally confirms diagnosis, but the micro-biopsy is a non-invasive tissue sampling technique first described in 2008, which is able to provide representative samples from wide areas of the oral mucosa. Its use has been suggested in the follow-up of potentially malignant disorders, above all in the presence of multiple lesions, such as in proliferative verrucous leukoplakia, leukoplakia, erythroplakia or oral lichen planus, which may require repeated histological assessment to detect any dysplastic/malignant evolution as early as possible.
The report illustrates the important diagnostic contribution provided by micro-biopsy sampling in the early detection of squamous carcinoma. A 78-year-old female, non-smoker was referred to the Department of Dentistry and Oral and Maxillofacial Surgery of San Rocco Hospital Brescia for the presence of white and red spots of the left buccal mucosa. She had an history of squamous carcinoma of the left buccal mucosa. A micro-biopsy was performed. The sampling allowed collecting small tissue fragments from the whole surface of the lesion. These fragments were processed as described in the previously cited paper, which first described the technique, resulting in a diagnosis of squamous cell carcinoma. The patient underwent surgery to remove the lesion with a definitive pathological diagnosis of squamous carcinoma. Micro-biopsy could be performed as a first-level diagnostic test in presence of oral potentially malignant disorders showing lesions with a low suspicion index in order to reduce the need to perform more invasive incisional biopsies and consequently in order to reduce the patient stress. Therefore it could represent a potential diagnostic aid for practitioners involved in the management of oral potentially malignant disorders.

References

Case report

Triple simultaneous oral squamous cell carcinoma in a heavy smoker patient: a case report

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Introduction. The presence of multiple malignancies in the aerodigestive tract is a rare disorder defined as multiple primary carcinoma (MPC)1, which lesions must be at different sites separated by normal mucosa and histologically confirmed, and should not be metastatic disease from the index carcinoma2.

The reported incidence of MPC in the oral cavity has been reported to be 1.4%. MPC can be simultaneous (diagnosed at the same time as the index tumour), synchronous (additional primaries diagnosed within 6 months of the index tumour), or metachronous (primaries that develop more than 6 months after the index tumour)2.

Case report. A 79-year-old edentulous male patient presented 3 different oral lesions: a) an erythro/leukoplasic lesion on the soft palate and uvula (2,5x1cm), b) a verrucous/ulcerative lesion on the floor of the mouth (1x1cm), c) an exophytic/ulcerative lesion on the edentulius ridge of the 4th sextant (3,5x1cm). Lesions were not associated to any symptoms; the lesions b and c were bleeding and fixed to the underlying tissue. The patient reported no health concerns; he referred a 10-pack-year of cigarettes history followed by 50-pack-year of cigars. All the lesions presented a dark royal blue aspect after the toluidine blue staining3.

Incisional biopsies were made and the histological examination reported a diagnosis of "oral squamous cell carcinoma" in each of the tissue samples. Patient was referred to the Department of Oral Maxillofacial and Plastic Surgery for the TNM staging and the management.

Conclusion. The MPC pathogenesis is uncertain involving genetic susceptibility, tumor immunity and environmental factors. Moreover, multiple cancers have a poor prognosis, 3-year survival rate of 50%, underlining that secondary prevention should be promoted and supported in adult heavy smokers, as the early diagnosis of oral carcinoma arises the probability of successful treatment.

References
Case report

Dental device closing a maxillary sinus communication in facial region

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Objective. This case shows a simple, economic and quick way to close a severe physical damage: a communication of maxillary sinus in facial region.

Case report. A patient with metastatic Melanoma underwent local radiotherapy in maxillary region and developed a deep necrosis of facial tissue which led the maxillary sinus opening outside near the nose; the pit which was temporarily closed with Formic-Iodized gauze. The hole determined an important aesthetic and functional impairment for the patient (difficulties in speech, chewing and swallowing) and he demanded its closure. So he was referred to our Department in order to fit out something to cover the injury up.

To eliminate the problem we established to build-up a basic resin prosthesis. We took a primary impression of the face with "Alginate" and cast a hard gipsy model; here we pointed the boundary-line on the skin and we prepare a wax model which was checked directly on the patient. At least we build up a soft-resin prosthesis (Vertex® Soft). Before its application the edges were corrected using a silicon paste (Fit-Cheker®). Patient was instructed to use a cream with Hyaluronic Acid and collagen precursor-Amino acids (Aminogam®) on the edge of the skin hole and, once a week he comes back to the clinic to perform an antibiotic washing of the sinus (Rifocin®).

Results and conclusions. Extended maxillary tissue loss may produce serious impairment which, not always, may be surgically corrected both for unique complexity and for patient’s general condition. In the reported case the basic resin prosthesis was able to replace effectively the lost tissue, restoring a suitable appearance and comfortable speech and chewing.

References

Workshop

Position papers in the medical literature: lights and shadows for ONJ treatment

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Management of osteonecrosis of the jaws (ONJ) associated with bisphosphonates (BPs) remains mainly empirical, despite ten years already passed since the first Marx’s case series publication dated 2003 (Marx R, et al., 2003). Several treatment protocols have been proposed in the medical literature to cure bisphosphonates-related osteonecrosis of the jaws (BRONJ), which lack robust evidence and add much confusion to the clinical decision-making of the specialists involved.

With the identification of a growing number of prescriptions other than BPs that are able to promote/sustain osteonecrosis of the jaws (e.g. denosumab, TKIs, anti-VEGF monoclonal antibodies, Inhibitors of mTOR), a new “disease-container” has been recently coined to include all of these drugs, namely medication-related osteonecrosis of the jaws (MRONJ). Starting from 2014, position papers from the most recognized Medical Associations/Societies implicated with BRONJ are being updated to include these new informations (Ruggiero S., et al., 2014; Khan AA, et al., 2015). However, little is known on the main features of non BP-ONJ.
As the treatment strategies included in the updated version of such recommendations have been originally developed for use in BRONJ exclusively (Ruggiero SL et al., 2014; Khan AA et al., 2015) their implementation non BP-ONJ is rather empirical and may be hazardous, as different drugs could promote osteonecrosis of the jaws through different pathogenic mechanisms and may deserve distinct treatments (Otto S et al., 2015).

Looking at the latest American Association of Oral and Maxillofacial Surgeons (AAOMS) recommendation (Ruggiero SL et al., 2014) and the systematic review and consensus of the American Society for Bone and Mineral Research (ASBMR) (Khan AA et al., 2015) indication to the use of conservative management strategies for MRONJ still visibly prevails over surgical treatment. Nonsurgical management of MRONJ is aimed at the elimination of pain, infection control and minimization of bone disease progression. It includes the use of antimicrobial mouth rinses (0.2% chlorhexidine digluconate), local disinfection/cleaning of exposed bone and fistula, pain control, the administration of antibiotics and nutritional support when required (Campisi G et al., 2014).

In other words, AAOMS and ASBMR keep on favouring a more prudent approach for BRONJ and MRONJ in general, and consider surgical treatment only when non-operative strategies have failed, without a documented curative purpose (Ruggiero SL et al., 2014).

Although there is certainly a lack of consensus in the literature about the correct management of MRONJ, both AAOMS and ASBMR fail to provide a fair and comprehensive summary of current knowledge and available evidence to support their statements (Otto S et al., 2015).

In fact, their observations are in contrast with the growing body of literature (Graziani et al., 2013; Rupe1 K et al., 2014; Kuhl S et al., 2012; Fielert R et al., 2019) which shows that surgery performs better than medical therapy both in terms of clinical healing and resolution of pain. Notably, surgical treatment of BRONJ has been shown to be curative rather than symptomatic (Bedogni A et al., 2011; Wilde F et al., 2012; Carlson ER et al., 2014; Schubert M et al., 2012). This information has not been reported by AAOMS and ASBMR, despite the former opens up to the potential role of resective surgery in restoring form and/or function (Ruggiero SL et al., 2014).

Initial management of BRONJ was based on stage-specific treatment strategies that have been currently up-to-dated to include all new forms of MRONJ. The AAOMS expert panel has substantially modified the disease-stage classification of BRONJ, as compared with the 2009 recommendations (Ruggiero SL et al., 2009) by shifting patients with mucosal fistulas and without bone exposure from stage 0 to stages 1, 2, 3. In other words, AAOMS recognised the idea that the previous stage classification was underestimating the disease itself, as recently confirmed by a large multicenter study (Fedele S et al., 2015). The anticipated result is that previously neglected patients will have now access to formal treatments. On the contrary, ASBMR consensus has not modified the disease stage classification accordingly, despite the fact that AAOMS is one of the most recognised societies contributing to their statements. AAOMS and ASBMR disease-stage classifications are defined almost exclusively by clinical criteria. It has been recently shown that a clinically driven staging system of BRONJ does not correctly address the extent of bony disease and is very likely to assign patients to inappropriate treatments (Bedogni A et al., 2014). The limited use of the more efficient surgical therapies and the presence of patients with various degree of bone disease within the same disease-stage are likely to be the reason for the poor and unpredictable treatment results so far obtained adopting classification systems as such. Future therapeutic implications may be even more significant, as the new AAOMS staging system will allocate more patients to treatments than in the past. We should therefore expect, to some extent, a worse scenario where a bigger number of patients will receive ineffective treatments. The recently published Recommendations of the Italian Societies of Maxillofacial Surgery (SICMF) and Oral Medicine and Pathology (SIPMO) introduced an alternative stage-disease classification, which assign patients to treatments based on the clinical and radiological signs of ONJ (Bedogni A et al., 2012). It is likely that the adoption of a classification system as such will be more predictable in terms of stage-specific results, regardless the type of treatment given (non-surgical vs surgical).

Since the assumption that novel antiresorptive and antiangiogenic drugs can promote osteonecrosis, the debate has now been reopened on the potential benefit of medication interruption. While there is general agreement that BPs should not be waived for the risk of precipitating the disease for whom BPs were originally given, it is likely that temporary interruption will play a role in the treatment of MRONJ caused by medications with a much shorter half-life then BPs (i.e. denosumab, bevacizumab, sunitinib, everolimus).

In conclusion, management of MRONJ remains controversial; non-operative treatment is still advocated as the leading therapeutic option, despite its poor healing potentials. In BRONJ patients, superior results can be achieved with surgery that can be curative in the long-term. It has to be ascertained whether non-surgical therapies may play a major role in the management of MRONJ caused by denosumab and antiangiogenic drugs. Temporary suspension of denosumab and antiangiogenic drugs may contribute to the efficiency of non-surgical therapies.

References

Workshop

EU/IT Multicenter Studies (ON/OFF) and the “DoctOral” System for ONJ

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The aim of the presentation is to describe two new technological projects applied in the field of medicine and pathology of the oral cavity.

On Off (Osteonecrosis Oral Findings & Future), supported by AIFA national project “Farmaci anti-angiogenetici e rischio di osteonecrosi dei mascellari - progetto multicentrico su dati retrospettivi, ottimizzazione della farmacovigilanza e della prevenzione secondaria, studi genetici”, is a web portal, aimed for the collection of medical history, clinical and radiological findings related to patients with drug-related osteonecrosis of the jaw. The coordinating and validating center of cases is represented by the Sector of Oral Medicine “V. Margiotta” in Palermo, and all data may be carried out following recognition of credentials in different national centers that will join free of charge. The creation of this information system will break down the power bias resulting from imprecise set of data on this new and serious emerging disease, in order to have more control in the incidence value and to give a fast service of second opinion for management. Primary goal is to widen our network, in order to design multicentric study and confirm the need of a standardization of the diagnostic and therapeutic protocols.

Doctoral® is an application (currently free of charge), which will be accessible on iOS and Android platforms, and will provide, exclusively for healthcare professionals and students of the School of Medicine, a tool for achieving the diagnosis of oral lesions: it is a matter of an easy pattern way, by means of questions and answers, to find the possible diagnosis. In addition, everyone, at the end, can take advantage of the second opinion service by mail.

These projects are under the patronage of SIPMO.

Workshop

Effect of prevention on the incidence of BRONJ: three years clinical follow-up

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