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Contents

Editorial

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It is an honor to address our readers on a day marked by the publication of the first issue of *Odontoestomatología* at the beginning of the term of the new Dean of the School of Dentistry of Universidad de la República, Uruguay.

In the proposed "Agenda for debate" submitted to the university community at the meetings where the Dean was elected we set out to: *"Work within an ongoing and open dialog with all the social and institutional actors to have a Dean's term of office that focuses on the training of 'university people', taking into account the multiple dimensions of humans. The main objective is to create the necessary conditions to improve the health of all Uruguayans and honor the mission society has entrusted to us as we run this educational institution".*

Odontoestomatología is a source of pride for our institution, as well as a solid credential when introducing ourselves on account of its high level and academic commitment, in compliance with the international standards of quality scientific journals. Scientific publications in the field of human health are essential to preserve and disseminate scientific advances and to ensure the visibility, accessibility and availability of the knowledge created. In this way they impact the training and updating of students, teachers and professionals, and ultimately, the improvement of health care based on scientific evidence.

Its strength lies in the authors that publish their work, and also, to a large extent, in the scientific editorial committee that assesses and decides on the papers to publish.

Their goals, objectives and targets have evolved and gradually gained prestige and visibility in the national and regional field of publications. Additionally, with its recent online version in Spanish and English, the journal has acquired international status, which has helped open quality communication lines to ensure that the scientific work of these authors fulfills its main objective: reaching and interacting with readers.

It is for this reason that I invite national and regional research colleagues to continue on the path of knowledge generation to ensure that people's quality of life is improved.

Prof. Dr. Raúl Riva Bernasconi
Dean

Influence of cannabis use on periodontal disease: a scoping review

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Abstract

Recent publications suggest the possible association between cannabis (Cannabis sativa) and periodontitis. **Objective**: To analyze the possible influence of cannabis use on periodontal disease, highlighting the available evidence and identifying the associated variables in the studies. **Materials and methods**: We conducted a scoping review applying a structured search method in PubMed/MEDLINE, Science Direct, LILACS, SciELO including publications until May 2017. **Results**: In vivo studies showed greater bone loss in animals exposed to cannabis. Clinical cases show that chronic cannabis use may result in gingival enlargement (with clinical features similar to phenytoin induced enlargement) and localized severe chronic periodontitis. Most of the epidemiological studies confirmed the possible association between cannabis and periodontitis. **Conclusions**: The specific mechanism by which cannabis acts in the gingival tissues is unknown.

Keywords: cannabis, periodontal disease.

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Introduction

Periodontitis is one of the most common chronic diseases, with a high prevalence that varies according to each population group[1,2]. It affects approximately 46% of adults in the United States of America[1]. In turn, a recent *global burden* report showed that over 11% of the world's population presents severe forms of the disease[2]. In Latin America, almost 10% of the young population is affected by periodontal disease, and approximately 35% of children have gingivitis, with the highest prevalence rates in Colombia (77%) and Bolivia (73%)[3]. In adults, periodontitis affects up to 62.6% of individuals[4]. In Uruguay, data from the First National Survey of Oral Health showed that among young people (15 to 24), the percentage of individuals with no signs of bleeding, with absence of tartar and probing depth (PD) ≥ 4mm was close to 30%[5]. Similarly, individuals aged between 35 and 44 had a 16.5% prevalence of moderate and 5.9% prevalence of severe periodontal disease respectively[6]. The highest prevalence was found in individuals aged between 65 and 74, where 34.7% had moderate periodontitis and 17% severe periodontitis[6]. These results emphasize the importance of prevention and treatment in oral health programs[7].

Periodontal disease is mainly characterized by gingival inflammation, formation of periodontal pockets and destruction of the supporting tissues (alveolar bone and periodontal ligament)[8]. It is the result of the interaction of microbial biofilm (necessary etiological factor but insufficient in itself), a susceptible host and modulating factors[9]. Several studies have shown the existence of risk factors, among them tobacco[10], diabetes[11–14], obesity/overweight[15,16] and genetic factors[17,18]. These elements modulate the host's susceptibility or resistance with each microbial challenge[9].

Recent publications suggest a possible association between cannabis (*Cannabis sativa*) and periodontitis. However, the use of cannabis components could have positive effects as they might reduce inflammatory processes[23]. If we consider the increase in the prevalence of cannabis use throughout the world (16% in the United States, 11% in France and 9% in Uruguay)[24], it is essential to research the possible effects of cannabis on the oral cavity and periodontal tissues in order to understand its role in the onset of periodontal disease and to develop and lead appropriate public health policies.

Therefore, the aim of this scoping review is to analyze the possible influence of cannabis use on periodontal disease, surveying the available evidence and identifying the associated variables in the studies.

Methodology

Study design: The scoping review involves a systematic search but does not imply an analysis of the methodological quality of the studies. This review presents a summary of the articles available in the literature by providing an overview of the existing content, setting future research paths and pointing to the gaps in the literature[25,26].

Search strategy: We conducted a scoping review applying a structured search method in PubMed/MEDLINE, Science Direct, LILACS, and SciELO including publications until May 2017. We used keywords and controlled terminology (MeSH terms) based on questions structured according to the PICO Model: "What is the possible influence of cannabis use on periodontal disease?" where we defined:

Population: individuals with periodontal disease.

Intervention: cannabis use

Control: individuals who do not use cannabis

Result: Periodontal disease alteration

In this way, the following search strategy was implemented: (periodontal disease OR periodontitis OR gingivitis OR gingival disease) AND (cannabis OR marijuana). All study designs on humans and animals were included.

Study selection and eligibility criteria: Four researchers participated in the paper search (MM, AF, LC and EA) for the review design, and were advised by a librarian. The records were entered into EndNote (Thomson Reuters, Rochester, New York, NY, USA) to eliminate duplicates and create a virtual library. The researchers read and filtered the titles and abstracts for all records that complied with the predefined criteria. No language or year restriction was applied. All original papers were included. Letters to the editor, *in vitro* articles and reviews were not included in this review. Additionally, the references of each paper were traced in order to broaden the search.

Results

A total of 260 records were obtained from the search, of which 65 articles remained after removing duplicates. After reading titles and abstracts, 10 articles were selected to read the full text. Two of these articles were excluded[27,28]. The reasons for exclusion are presented in figure 1. Finally, eight articles met the inclusion criteria and were included in this review (Figure 1).

***In vivo* studies:** Studies on mice evaluated the influence of the inhalation of cannabis and cannabidiol (CBD) on periodontal disease measuring attachment loss and bone density. Increased bone loss and lower bone density were observed in mice exposed to cannabis or cannabidiol, which shows that these substances can negatively influence periodontal tissues.

Clinical cases: Two clinical cases were included in this review[29,30]. These articles show that long-term cannabis consumption may lead to gingival enlargement with clinical features similar to the ones caused by phenytoin,[30] in addition to chronic localized severe periodontitis[29].

Epidemiological studies: four epidemiological studies were retrieved. Table 1 shows the main features of the studies. The methodological design of most studies was cross-sectional[19,20,31], and there was a prospective study from a cohort of births[27]. Fifty percent of the studies were population-based[19,27], while one was conducted with high school students[20] and another with aboriginal communities from Australia. Three of the four studies were conducted on adults[19,27,31], while only one evaluated adolescents[20]. Similarly, Shariff et al. [19] included patients with diabetes and previous periodontal treatment, while Lopez et al. [20] were the only ones that evaluated the patients' oral hygiene habits.

Although cannabis exposure and the classification of periodontal disease have been categorized differently in the studies reviewed, cannabis use is linked to periodontal disease (Table 2). We observed a significant association between cannabis use and prevalence of periodontitis[21,31], where cannabis users had a prevalence of periodontal disease 44% higher than that of non-users[31]. Similarly, population data from the United States National Health and Nutrition Examination Survey showed that the recreational use of cannabis was associated with advanced probing depth and clinical attachment loss[19]. Additionally, a study conducted on Chilean students found a Necrotizing Ulcerative Gingivitis (NUG) odds ratio that was 53% lower in individuals

who had never used cannabis compared to frequent users[20]. The other associations were not statistically significant[20].

Influence of cannabis on periodontal disease: Table 1 shows the impact of cannabis on periodontal disease according to the action of its chemical components. Authors state that the causes of chronic inflammation in patients that use cannabis are the high temperatures and chemicals released during the consumption, followed by the clinical symptoms of xerostomy (as it has parasympatholytic properties), which would enhance the pathological effect[32]. We must remember that cannabis components that are not cannabinoids (products of combustion) are similar to those in tobacco and can have local and systemic effects [21,22,33]. Only low CBD concentrations can have an anti-inflammatory effect, while high doses would have the opposite effect[34].

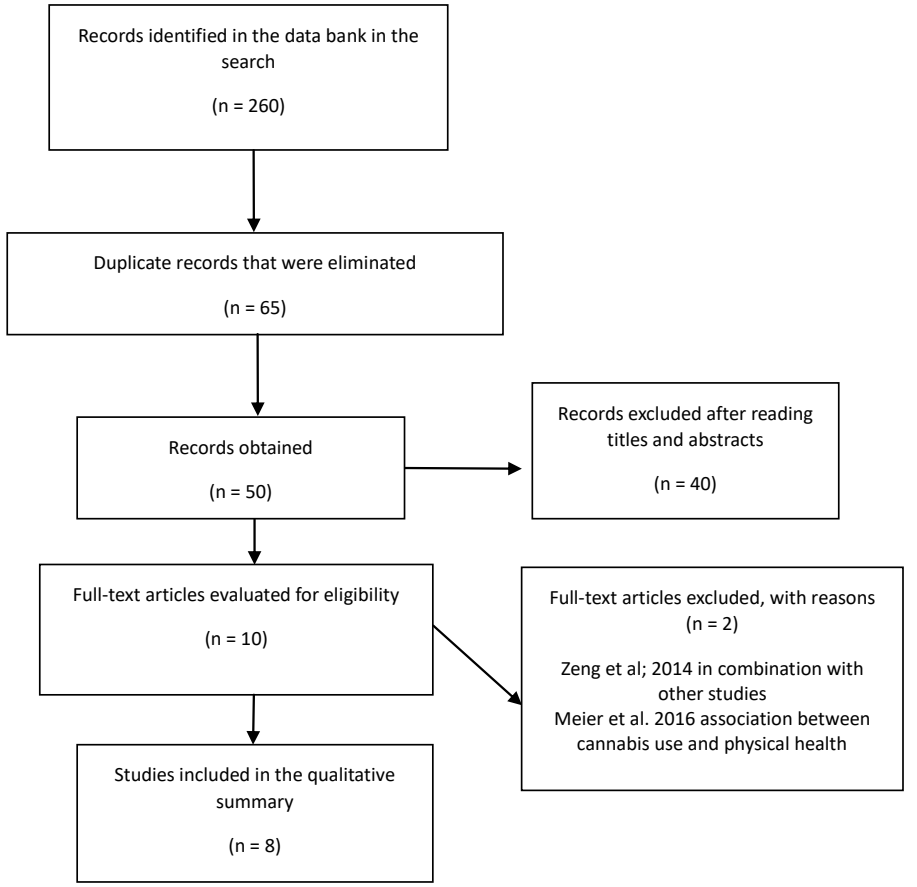

Figure 1: Flow chart showing the articles included.

Table 1. Chart comparing the epidemiological studies conducted to date. Relevant variables are identified to create a standardized research protocol to compare results.

	Thomson et al 2008	López et al 2009	Jamieson et al. 2010	Shariff et al. 2016
Number of patients	903	9203	425	1938
Country	New Zealand	Chile	Australia	United States of America
Study design	Prospective (cohort)	Cross-sectional	Cross-sectional	Cross-sectional
Population	Population-based study	High school students	Aboriginal communities	Population-based study
Age*	Adults aged 26 to 32	Adolescents aged 12-14; 15-17; 18-21	Adults over 18	Adults aged 30 to 49
Probing depth	Si	Si	Si	Si
Gingival recession	Si	Si	Si	Si
Clinical attachment loss	Si	Si	Si	Si
% bleeding	No	No	No	No
Tobacco **	Si	Si	Si	Si
Cannabis **	Si	Si	Si	Si
Diabetes	No	No	No	Si
Previous periodontal treatment	No	No	No	Si
% Plaque	Si	No	No	No
Oral hygiene habits	No	Si	Si	No

***Over 18 years of age. **Frequency, amount, and time elapsed since start of consumption, recreational or medical use.**

Table 2. Summary of clinical studies aiming to prove the association between presence of periodontal disease and cannabis consumption

Author	Cannabis exposure	Periodontal record	Periodontal disease
	Association / Quality of the study		
Thomson 2008	"No exposure", "Moderate exposure", "High exposure"	3 sites / 2 quadrants (26 years old) - Full mouth (32 years old) (except 3rd molars and implants)	CAL ≥ 3mm
	Strong association between cannabis use and prevalence of periodontitis		
López 2009	"Never used cannabis", "Regular use of cannabis"	6 sites/Incisors, 1st and 2nd molars	CAL ≥ 3mm, NUG.
	An association was found only when cannabis had never been used and with a lower NUG prevalence. There was no association for any other case		
Jamieson et al. 2010	"Never used or used only once" "Occasional use" "Frequent use"	2 sites/Full mouth (except 3rd molars and implants)	PD ≥ 5mm and CAL ≥ 4mm (moderate periodontitis)
	Cannabis users had a 44% higher prevalence of periodontal disease that individuals who do not use cannabis.		
Shariff 2016	"Never used" (Non-FRC) "Used every month for a year" (FRC)	6 sites/Full mouth (except 3rd molars and implants)	PD ≥ 4mm and CAL ≥ 3mm (incipient periodontitis)
	A strong association was found for FRC and presence of PD and CAL compared to non-FRC users.		

CCL: Clinical crown lengthening; PD: Probing depth; CAL: Clinical attachment loss; NUG: Necrotizing Ulcerative Gingivitis; FRC: Frequent recreational cannabis. Non-FRC: Non-frequent recreational cannabis.

Chart 1 Graphic description of the possible impact of cannabis on periodontal disease according to the action of its chemical components.

Tobacco	Cannabis
Nicotine (↑ Inflammation)	CBD (↓ Inflammation)
Combustion compounds (↑ Inflammation)	Combustion compounds (↑ Inflammation)

Discussion

This work is the first scoping review that analyzes the connection between periodontal disease and cannabis consumption as it summarizes the evidence available. Eight articles were included, ranging from animal studies and clinical cases to observational (prospective and cross-cutting) studies, and indicating an influence and possible association of individuals who use cannabis and periodontitis. As this is a very important public health issue, health services should raise awareness of the risk that regular and occasional cannabis users run of having this pathology.

Cannabis sativa L. (hemp), has been known for many years for the use of its fibers in the textile industry, for clothing, material constructions and paper[35,36]. It was only in 1930 that isolated compounds called *cannabinoids* were isolated from cannabis. The most abundant ones within the extract are: Delta-9-tetrahydrocannabinol (THC), which has the strongest psychoactive effect, its precursor, cannabidiol, as well as cannabiol, a catabolic THC product formed spontaneously. These last two lack THC's psychoactive effect, but have been found to have anticonvulsant properties[35,37].

Currently, there are different types of cannabinoids according to their origin: 1) phytocannabinoids, derived from the cannabis plant; 2) endogenous cannabinoids (endocannabinoids), produced by the body itself of humans or other animals and 3) synthetic cannabinoids, with an identical chemical composition, but produced in the laboratory[38]. The characterization of these and other derivatives, as well as the receptors they interact with, have improved our understanding of the Endocannabinoid System[35,39].

THC is the major psychoactive constituent and participates as a partial agonist for cannabinoid receptor type I (CB1) and type II (CB2). CBD is not psychoactive and is an antagonist for CB1 and CB2. It acts in multiple other receptors and may be an agonist for some systems[39]. The pharmacological effects of CBD are mediated by G-protein-coupled receptors, CB1 and CB2. When activated, CB1 receptors inhibit sympathetic transmission by acting on voltage-gated calcium and potassium channels, which are known to modulate epileptiform activity and seizures. CB2 receptors are primarily expressed in the immune system and have limited expression in the central nervous system. The effects of CBD are CB2 receptor-independent[40,41]. Thus, cannabis is usually smoked, vaped or consumed orally in the form of foods, teas or capsules[41].

In this way, approximately 3.8% of the world's population consumed cannabis in 2014 (183 million people): Iceland has the highest figure (18.3% of the population over the last 12 months), followed by the United States (16.2%), Chile in the fifth position (11.83%), France in the sixth (11.1%), and Uruguay coming in eleventh with 9.3%[42]. According to the World Drug Report 2011 (annual United Nations Office publication), in 2006, 14.8% of young people in Uruguay had consumed cannabis in the previous year, while only 6% of adults had used it[43]. The Final Report of the 5th National Household Survey on Drug Use of 2011 published by the National Drug Board

in 2016[44] shows that since 2011 Uruguay has had an increase of three percentage points in marijuana consumption. This is the most widely used substance in the population after alcohol, tobacco and sedatives. Twenty-three percent of the people between the ages of 15 and 65 have used marijuana some time in their lives, and 9.3% say they used it in the last 12 months (161,000 people), and 6.5% in the last 30 days. There are 21,355 daily marijuana users in Uruguay.

Although cannabis is considered a "soft" drug which is only as harmful as coffee or tobacco, cannabis use can cause chronic secondary effects, such as periodontal disease[21,32]. The temperature of the combustion smoke is higher than that of cigarettes[30]. The lesions reported are similar to those of tobacco users, but always less severe[30]. Both acute and chronic consumption lead to nicotine stomatitis and uvulitis[30]. This connection between tobacco and cannabis is even closer: a study reports that not all tobacco users smoke cannabis, but 90% of cannabis consumers smoke tobacco[45]. This makes it difficult to independently diagnose the effects of both types of substances in the studies. This is why some reports conduct statistical analyses also with individuals who do not smoke tobacco, and the association with periodontal pathologies remains[19]. As most individuals who smoke cannabis also use tobacco[31], this may enhance the harmful effects of both drugs.

Multiple studies show the association of cannabis with high levels of oral biofilm, dental caries, candida albicans (but not candidiasis)[41], leukoedema, leukoplakia[46] and stomatitis[37,41]. In addition, excessive exposure to cannabis was associated with an increase in respiratory diseases and in the prevalence of cancer of the oropharynx[39]. Reports indicate that 58.3% of women and 47.4% of men reported pulpitis during cannabis consumption. Pulpitis could be added to the list of adverse vascular effects associated with cannabis use already reported (conjunctivitis, tachycardia, hypotension, angina pectoris)[47,48]. Thus, it is important to note that only low concentrations of CBD can have an anti-inflammatory effect and that high doses are harmful[34]. In the case of oral cancer, proinflammatory and carcinogenic compounds have been found in cannabis smoke, similar to the composition found in tobacco smoke, including carbon monoxide[22,33,49,50].

THC and CBD stimulate the release of prostaglandin E2 (PGE2) from synovial cells and inhibit the *in vitro* synthesis of leukotrienes of human nuclear polymorphic cells[51]. Additionally, it has also been reported that CBDs suppress proinflammatory mediators[52–54] such as IFNγ (gamma interferon), FNT-α (tumor necrosis factor-alpha), IL-1b (interleukin 1 beta)[55] and IL-10 (interleukin 10)[56]. In another study, where CBD doses were systemically injected in rats, researchers concluded that they are an emerging class of mediators that might participate in the control of periodontal pathologies as they help reduce inflammation[57]. Contrary to these findings, other authors conducted the same clinical trial, noting increased bone loss in the furcation area of the teeth with induced periodontitis. However, no effect was noted on periodontally healthy sites, which could be related to an alteration of the immune function during the bone resorption process, or even the activation of specific receptors that could increase bone destruction[58]. In this sense, there has been some speculation regarding the endocannabinoid system, which could play a role in the regulation of bone metabolism[59,60].

In addition, we identified specific receptors for this substance in periodontal tissues, which was followed by an increase in the release of anandamide, an agonist for the cannabinoid receptor derived from arachidonic acid[61]. Other studies confirm these data: the relationship between the endocannabinoid system (receptors and mediators produced by our body) and periodontal disease was studied, and researchers detected a proliferation of gingival fibroblasts when the system was activated, suggesting a

new path for periodontal disease therapy[62,63]. A recent study shows how human gingival mesenchymal stem cells that were pre-treated with CBD before the transplant increase their survival rate in the host as they modulate their immune and inflammatory response[41,64].

In turn, in 2012 Rawal et al.[63] described two clinical pictures in patients and consumers. The features that always appeared were inflammation of papillae and gingival margin, with presence of nodular areas, similar to those in patients that consume dilantin (phenytoin), mainly in anterior teeth. Their analysis is very interesting as they find possible coincidences in these clinical pictures, finding similarities in the chemical components of both substances: 1) cannabidiol (CBD) is also used as an anticonvulsant, 2) CBD may increase the effects of phenytoin and phenobarbital, 3) CBD and phenytoin have a similar structure, displaying rings in the same pattern. The authors conclude that both comply with the stoichiometric requirements suggested for the action of an anticonvulsant, deducing that inflammation may be caused by the same mechanism[63].

In 2016, Momen-Heravi et al. reported the case of a 23-year-old female patient diagnosed with periodontal disease. The patient reported daily use of cannabis for three years. The author recounts inflammation of papillae and gingival margins, mainly at the anterior region of the mandible, where the cannabis cigarette was placed. The X-ray showed loss of alveolar bone in that area[29]. Regarding the treatment, the authors recommend behavior modification and non-surgical and surgical therapy for the successful management of cannabis-related periodontitis[29].

In this context, the first epidemiological study that analyzed the relationship between cannabis use and periodontal disease was conducted in 2008 by Thomson et al.[21] in New Zealand. The population was made up of adults born in a hospital in the country in 1972 and 1973. The periodontal clinical examination was performed on two occasions, at ages 26 and 32. The recording system included two quadrants on three sites per tooth, examining Gingival Recession (GR), Probing Depth (PD) and Clinical Attachment Loss (CAL). Cannabis exposure was assessed applying a self-report methodology. From this information, three groups were obtained: 1) no exposure 2) low exposure (1-40 times in the previous year) and 3) high exposure (over 41 exposures in the previous year). The study also enquired about socioeconomic status, tobacco use, sex, reason for attending the dental clinic, and biofilm accumulation[21].

Of the patients, 32.3% had not consumed cannabis in the previous year, and 47.4% and 20.2% had done so at low and high exposure rates respectively. Most consumers were men, of low socioeconomic status and who rarely sought dental care, with significant levels of bacterial plaque[21]. This confirms the previous findings that show these individuals tend not to worry about their health, which in turn can be linked to oral pathologies[32]. Of the patients, 33% were tobacco smokers and 17% ex-smokers. This is consistent with Fairman[45] in that frequent cannabis consumers were also tobacco users, and as age increased, tobacco consumption increased too[32].

The reported results show that cannabis use was strongly associated with the prevalence of periodontitis, with the greatest differences found in PD $\geq$ 5mm. However, no association was found between the consumption of both substances at the same time and periodontal disease. According to Thomson, the use of cannabis may be a risk factor for periodontal disease when used independently from tobacco, and as age increases so do prevalence and incidence[21].

The association with Necrotizing Ulcerative Gingivitis (NUG) was also researched on a sample of adolescents. In this case, six sites were measured and observed in the incisors and second molars, and the NUG diagnosis was made[20]. Like Thomson et al., they found that it was a limitation not to know the dose and

length of cannabis use, as the questionnaire included "never consumed" or "regular consumption" with 18.9% and 6% respectively. Of the regular consumers, only 16.3% were smokers, contrary to what Thomson et al. and Fairman found. The NUG diagnosis did not consider pain or bleeding, hence its high prevalence compared to other studies; no association with cannabis consumption was found[20,21,45].

In turn, in 2016, Shariff observed the same in adults in the United States, where 26.6% were frequent cannabis users. From that group, 29.2% had PD ≥ 4mm, 24.8% PD ≥ 6 mm and 24.5% PD ≥ 8mm, comparing these data to that of non-frequent users: 22.3%, 19.2% and 18.9%. They concluded that frequent cannabis use is associated with increased probing depth and gingival recession, as well as higher likelihood of severe periodontitis[19].

Although the studies have varying methodologies, *in vivo* studies, clinical cases and epidemiological studies seem to indicate an association between cannabis use and periodontal disease; this may increase bone loss, exacerbating or initiating periodontitis. These authors recommend including the variables there presented (Table 1) to achieve a standardized research protocol, non-existent so far, to be able to compare the results of different studies conducted in the future. The authors recommend that further epidemiological studies, preferably prospective or case-control studies, be conducted, since clinical trials would be ethically unacceptable. In addition, it is important that future research include statistical analyses, checking if individuals are tobacco users in order to reduce any potential bias. The results of this review should be interpreted with caution since they are based on studies that apply very different methodologies.

Conclusions

The specific mechanism by which cannabis acts on gingival tissues is unknown due to the insufficient number of studies conducted so far and the differences in methodologies and in the populations studied. However, cannabis consumption seems to make periodontal disease worse. This is why health services should take action to raise awareness of the strong likelihood of regular cannabis users having this pathology.

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Retrospective clinical study of 656 cast gold inlays/onlays in posterior teeth, in a 5 to 44-year period: Analysis of results

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Abstract

Objective. 1) To assess the clinical performance of 656 cast gold inlay/onlays in a 44-year period; 2) To analyze their indications and distribution regarding the evolution of scientific evidence. ***Materials and Methods***. A total of 656 cast gold inlays/onlays had been placed in 100 patients. Out of 2552 registered patients, 210 fulfilled the inclusion criteria. The statistical representative sample was 136 patients; 140 were randomly selected and 138 were the patients studied. Twelve variables were analyzed. Data processing was done using Epidat 3.1 and SPPS software 13.0. ***Results.*** At the clinical examination, 536 (81.7%) were still in function and 120 (18.3%) had failed. According to Kaplan-Meier's method, the estimated mean survival for the whole sample was 77.4% at 39 years and 10 months. ***Conclusions.*** Knowledge updating is an ethical responsibility of professionals, which will allow them to introduce conceptual and clinical changes that consider new scientific evidence.

Keywords: inlays/onlays, molar, premolar, dental bonding restorations, scientific evidence-based, minimally invasive dentistry.

Disclosure

The authors declare no conflicts of interest related with this study.

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Introduction

Restorations in posterior teeth, mainly molars, withstand the greatest occlusal forces. Moreover, 40% greater forces could be observed in patients with bruxism, increasing their risk of failure[1]. Additionally, restorations covering more surfaces have had greater risk of failure[2]. For over 100 years, cast gold restorations have been widely used, mainly in posterior teeth. Several clinical studies show excellent quality, longevity and functional performance[3-6].

Since the beginning of the nineties there has been a permanent scientific and technological development of adhesive ceramic materials for posterior inlays/onlays. Clinical studies and systematic reviews have shown very good results[7-12]. Moreover, improving biomechanical and optical properties in direct light-activated composite resins[13-18], as well as in the adhesive systems[19-22] has increased their use[23-24], showing high long-term clinical success, performing extended and large restorations in posterior teeth[25-32]. This scientific evidence has provided the basis for using these materials as substitutes of cast gold restorations.

At the same time, cultural, sociological and psychological factors related to self-esteem[33] and new aesthetic parameters[34-36] have increased patients' demand for tooth-colored restorations. All the above can change concepts and clinical procedures, leading to a reduction in the indication and placement of cast gold inlays/onlays. These changes were observed in productivity studies conducted by the operator in his private office in over 25 years. In fact, between 1995 and 2000 there was a 19.8% decrease in the use of gold inlays/onlays (GIOs) (Fig. 1), an increase of 107.6% in the placement of bonded ceramic inlays/onlays (BCIOs) (Fig. 2) as well as a 44.7% increase in posterior direct light-activated composite resins (PCRs) (Fig. 3).

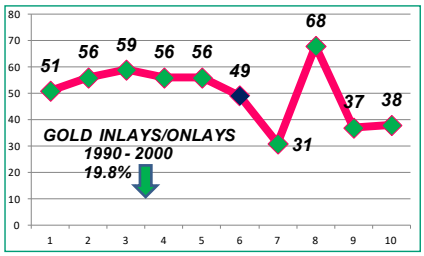

Fig. 1. The chart shows a decrease in the use of GIOs since 1995 (blue dot).

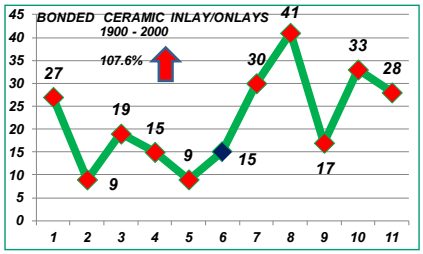

Fig. 2. The chart shows an increase in the use of BCIOs since 1995 (blue dot).

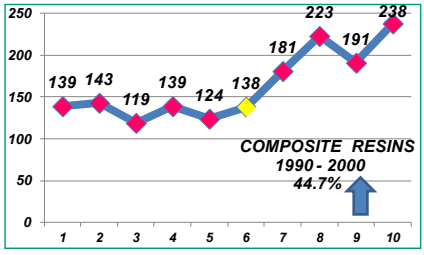

Fig. 3. The chart shows an increase in the use of PCRs since 1995 (yellow dot).

The right selection and the proper handling of the new materials are possible if the professional's knowledge is scientific and evidence-based. In fact, it is an ethical responsibility.

Therefore the aims of this article were: 1) to assess the clinical performance of 656 cast gold inlay/onlays in a 44-year period, and 2) to analyze their indications and distribution in relation to the evolution of scientific evidence.

Materials y Methods

This work was approved by the Ethical Board of the School of Dentistry, Universidad de la República.

From April 2013, a retrospective clinical longitudinal study on six restorative procedures, performed by the first author in his private practice, has been conducted covering a 44-year period. Four articles have been published[12,32,37,38]. This paper presents the results of a new study in the series.

To be included in the study, the patients must have been treated and evaluated in the first author's office for at least seven years and be still in the practice by 2013, with complete dental arches (healthy or restored teeth). The patients with removable dental prosthesis, disabilities, that had moved or passed away were excluded.

The GIOs must have been in function for at least five years to be included in the study. The failure criteria were: loosening, removal of the restoration or tooth lost.

Out of 2552 registered patients, 210 fulfilled the inclusion criteria and 136 patients were a statistically representative sample (at the lowest range, a 95% CI for a proportion, with a length of ± 5%). Of these 210 patients, 140 were selected at random by a dental assistant, who was blinded to the aim of the selection. They were invited to participate in clinical examinations between November 2013 and April 2014: 138 agreed to participate.

Considering the period of the clinical examination (11/2013 - 04/2014), the inclusion criteria for the GIO (> 5 years in function), and the second aim of this research, the restorations to be analyzed were divided into two groups, according to the date of placement: A - between April 1969 and April 1991, and B – between April 1991 and October 2008.

According to the Glossary of Prosthodontics Terms[39], the meaning of inlay and onlay terms were slighty modified in this work for practical reasons: "inlay" is when the GIO covers partially the occlusal surface, including one or more cusps; "onlay" when the GIO covers the whole occlusal surface.

Patient-based data were collected from the clinical personal charts and clinical examinations for each patient, and then recorded on specially designed sheet forms.

All restorations were made by the first author. The co-authors participated as independent evaluators. The calibration between the authors was undertaken previously on 120 restorative procedures from outside the sample. Cohen's Kappa Coefficient on the quality of restorations ranged from 0.78 to 1 (excellent according to Fleiss's guidelines).

To establish the quality of the restorations, marginal adaptation (MA), marginal discoloration (MD), and occlusal surfaces (S) were the variables evaluated according to Ryge's criteria[40] ; however, these variables were modified by the authors (Table 1).

As was previously published[32], four success criteria (SC) were established on the quality of the GIOs:

- ***Excellent*** (Ex): When MA, MD, and S were rated "Alpha" in the same GIO. This group represents the best quality restorations.
- ***Good*** (G): When at least one variable (MA, MD, S) was rated "Bravo" in the same GIO. This group was considered of satisfactory clinical acceptance.
- ***Functional survival*** (FS): When at least one variable (MA, MD, S) was rated "Charlie" in the same GIO. These restorations must be controlled, repaired or retreated.
- ***Clinically successful*** (CS): To establish clinical success, the GIOs in function rated "Excellent" and "Good", were considered jointly.

MARGINAL ADAPTATION
ALPHA: No lack of continuity along the margin observed with the explorer.
BRAVO: Evidence of a crevice along the margin, but the explorer cannot penetrate.
CHARLIE: Evidence of a crevice along the margin, which an explorer can penetrate. Requires control, repair or retreatment.
MARGINAL DISCOLORATION
ALPHA: No pigmentation anywhere on the margin.
BRAVO: Pigmentation present but does not penetrate the margin toward the pulp.
CHARLIE: Severe pigmentation. The restoration must be removed.
SURFACE
ALPHA: The surface of the restoration is unaltered.
BRAVO: The surface of the restoration shows wear and must be controlled.
CHARLIE: The surface of the restoration shows perforations, fractures or significant tear. It must be polished, repaired or retreated.

Table 1. Ryge's criteria[40], modified by the authors, to establish the quality of GIOs.

Patients signed an informed consent before the clinical examination. The clinical exams were blinded among the authors and performed with a mirror and a sharp explorer (Maillefer 6: Maillefer, Ballalgues, Switzerland). In cases of disagreement regarding the assessment of the quality of the restorations, the lowest rating was recorded.

The operator always conducted a clinical and functional study of the Stomatognathic System (SE). These data, as well as the clinical procedures performed, the materials applied and the patient's biological response were thoroughly recorded.

Since its availability in the market, a panoramic radiograph was always indicated. When restorations, carious lesions and/or periodontal disease were present, standard X-rays were requested.

All the patients were enrolled in a comprehensive prevention plan and advised to undergo periodical maintenance therapy. Since the late eighties, if caries disease was present, biochemical and microbiological saliva tests were indicated and a dietary form filled. The aim was to establish the individual caries risk and to implement the specific etiological treatments[41-44]. Furthermore, non-cavitated carious lesions were treated with topical fluorides.

Tooth preparation is considered by the operator as the most important factor in the clinical success of the restorative treatment. It must be based on a conservative approach, according to the extension of the carious lesion, remnant tooth structure, location of occlusal contacts, crown height, occlusal plane, habits of the patient, biomechanical properties of the restorative materials and aesthetic considerations. In deep cavities, a thin layer of self-cured calcium hydroxide was applied in the pulpal wall and covered with glass-ionomer cement.

The variables analyzed were age, gender, type of restoration, number, location, extension, quality and longevity of the restorations, type of cement, secondary caries, parafunctional habits and maintenance therapy.

Data processing was performed using Epidat software (Conselleria de Sanidade de la Xunta

de Galicia, Santiago de Compostela, Spain) v 3.1 developed by the Consellería de Sanidade de la Xunta de Galicia with the support of PAHO-WHO and SPSS software v13.0 (SPSS Inc., Chicago, IL). In all cases, the variables considered corresponded to an identically distributed independent random variable that was generated from a probability sample. A result was considered statistically relevant when, upon the rejection of a null hypothesis, the probability of error was less than 5%. Statistical significance was set at $P < 0.05$. When analyzing the complete and censored values, the mean and median values were used to describe the central tendency. If the number of complete values was too small, a Kaplan-Meier's curve could not be used. Therefore, a Fisher's exact test, Chi-square test, Kruskal-Wallis test, and Mann-Whitney non-parametric test could be indicated to analyze significant differences.

Results

Six hundred and fifty-six GIOs had been placed in 100 patients. The observed mean age was 63 years and 9 months for both sexes. The observed mean time of attendance per patient was 30 years and 5 months.

Carious lesions [557], endodontic treatment [85], functional requirements [7], trauma [4] and periodontal disease [3] were the reasons to perform the GIOs.

The number of GIOs per patient varied from 1 to 20. The average was 6.56 GIOs per patient: 6.22 in women and 7.07 in men. No statistical significant difference was found regarding sex (Mann-Whitney Test, p = 0.92).

GIOs in function and failed

At clinical examination, 536 GIOs (81.7%) were in function. The observed mean survival (OMS) was 27 years and 1 month. According to the date of placement, 343 were in group A- and 193 in Group B-.

One hundred and twenty GIOs failed (18.3%), 61 in 16 men and 59 in 17 women; 119 in Group A- and 1 in Group B-. The OMS for failures was 18 years and 4 months. No statistically significant differences between the IOOs in function (Student T Test, p = 0.65), nor in the failures (Student T Test, p = 0.99) were observed in both sexes.

The annual failure rate was 0.08%. The estimated mean survival time (EMS), according to the Kaplan-Meier's method, was 85% at 25 years and 77.4% at 39 years and 10 months (Fig.4).

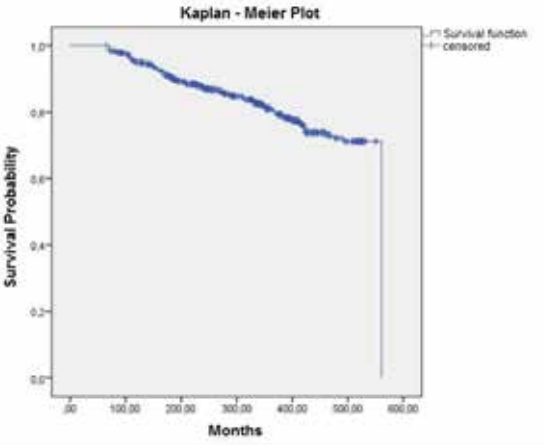

Fig. 4. Overall estimated mean survival according to Kaplan-Meier's curves.

Considering **location**, 331 GIOs in function were in molars and 205 in premolars. There was a statistically significant difference in the number of GIOs in function, restoring molars and premolars, between the two groups (OR = 0.42, p < 0.001) (Fig. 5).

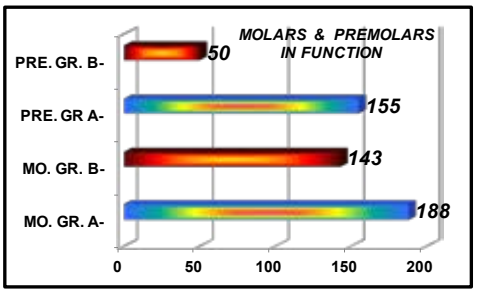

Fig. 5. Distribution and number of GIOs in function in molars and premolars in both groups (MO. =molars; PRE. = premolars; GR. = group).

Out of 120 failures, 89 were in molars and 31 in premolars. Premolar survival was significantly higher (Log Rank Test, $p < 0.0001$). According to Kaplan-Meier's method, EMS was 479.8 months (39 years and 11 months) for premolars and 437.4 months (35 years and 5 months) for molars (Fig. 6).

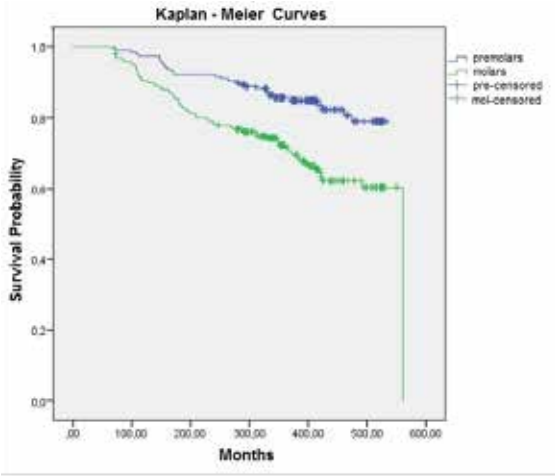

Fig. 6. Survival Kaplan – Meier's curves between premolars and molars.

Regarding **type of restoration**, 424 GIOs were onlays and 112 inlays. A statistically significant difference was observed in the number of onlays and inlays in function between the two groups (OR = 13.9, $p < 0.0001$). Thirty-eight onlays and 82 inlays failed. Onlays presented a higher statistical survival rate (Log Rank Test, $p < 0.0001$). The Kaplan-Meier's curves showed an EMS of 498.4 months (41 years and 6 months) for onlays and 402.2 months (33 years and 6 months) for inlays (Fig.7).

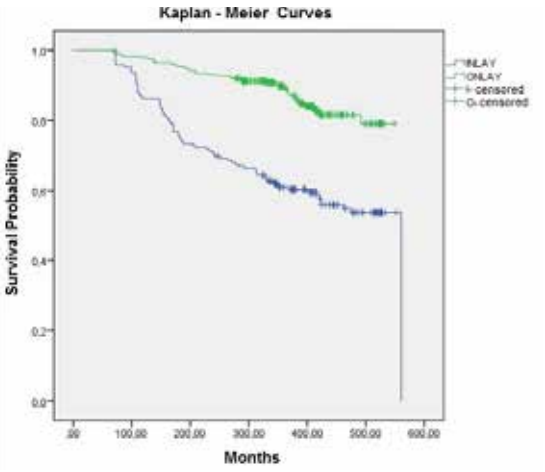

Fig. 7. Survival Kaplan-Meier's curves between onlays and inlays

Considering **extension**, 95% of the all GIOs, covered 3 surfaces: 507 in function and 116 failures; 20 covered 2 surfaces (4 failures) and 13 more than 3 surfaces.

Regarding **pulp state** at the moment of placement, 86.6% of the GIOs, restored vital teeth (VT): 457 in function and 111 failed. Out of 88 (13.4%) endodontically treated teeth (ETT), 79 remained in function and 9 restored with casts posts failed, but only one was fractured. Furthermore, 9 GIOs in VT had to be removed for endodontic treatment (1.4%).

Three GIOs were fixed with resin cement (Panavia, Kuraray, Japan) and 653 with zinc phosphate cement. One GIO fixed with zinc phosphate cement loosened after 32 years in function.

Secondary caries was the major reason for failures (14.9%): 89 in occlusal surfaces and 9 in gingival location. Of the 120 failures, 15 teeth (14 in group A and 1 in group B) were lost.

Table 2 summarizes the information on failures presented above.

In addition, 105 teeth in group A-, that had lost their GIOs could be retreated: 90 with new GIOs, that were included in this study, and 15 with other restorative procedures. Out of 90 GIOs (89 onlays and 1 inlay), 39 were retreated before April 1991, remaining in group A. Fifty

one performed after April 1991 were included in group B- .

PERIOD	O/C	G/C	End.	Perio.	Fra.	Loos.	R.R.	TOTAL
< 10 y	24	3	2	1	1	0	1	32
10 - 20 y	34	1	3	4	0	0	1	43
20 - 30 y	21	0	4	1	0	0	0	26
30 - 40 y	10	3	0	3	0	1	0	17
> 40 y	0	2	0	0	0	0	0	2
TOTAL	89	9	9	9	1	1	2	120
%	74.2	7.5	7.5	7.5	0.8	0.8	1.7	100

Table 2. Failures of GIOs: causes, number, distribution in decades and percentages (O/C = occlusal caries; G/C = gingival caries; End.= endodontic treatement; Perio.= periodontal disease; Fra.= root fracture; Loos. = loosened; R.R.= rooth resorption; y = year).

Meanwhile, group B- included 194 GIOs **placed after April 1991** (193 in function, 1 failure). The distribution of the 193 GIOs in function was: 48 in 22 patients that had not been in group A-; the above mentioned 51 GIOs retreated, and the 94 new GIOs placed in 62 patients in group A- (Table 3).

PERÍOD	Gr. B	Gr. A/B	RETR.	TOTAL
04/91 - 04/95	9	35	19	63
04/95 - 04/99	15	29	18	62
04/99 - 04/04	12	18	8	38
04/04 - 10/08	12	12	6	30
TOTAL	48	94	51	193

Table 3 . Distribution of GIOs in function in group B-, placed after April 1991, according to the period of placement and their origin (Gr. B- = GIOs installed in new patients; Gr. A/B = new GIOs placed in patients of group A-; RETR. = GIOs retreated in patients of group A-).

Success criteria

According to Ryge's criteria[40] modified by the authors, the results on the quality of the variables analyzed are presented in Table 4. Considering these results, and the previous established criteria[32], the CS of the GIOs in function was 93.6%. The CS for the whole sample was 76.1%.

	MARGINAL ADAPTATION	MARGINAL DISCOLORATION	OCCLUSAL SURFACE
ALPHA	390	533	465
BRAVO	129	2	50
CHARLIE	17	1	21

Table 4. Number, distribution and quality of the variables analyzed, according to Ryge's criteria[40] modified by the authors.

Three GIOs in Figure 8 were qualified Ex, and 2 in Figure 9 were qualified B. The GIOs repaired in Figure 10 were qualified as FS.

Fig. 8. GIOs qualified Excellent (a= year, m = months).

Fig. 9. GIOs in premolars (44 years) qualified Bravo (a = year, m = months).

Occlusal function and maintenance.

Twenty-four patients (24%) had had history of parafunction, and 33 (33%) were using stabilization occlusal splints due to bruxism. Thir-

ty-seven patients (37%), 21 women and 16 men, attended at least once a year for maintenance therapy.

Repairs

Meanwhile, in 21 patients, 27 repairs were performed (23 in occlusal and 4 in gingival areas). The materials applied were Direct Light-activated Composite Resin [20], Glass-Ionomer [1] and Amalgam [6]. These GIOs were considered in function and qualified as functional survival. The OMS of the repairs was 12 years and 8 months (range: 3 years to 22 years and 9 months). Two GIOs repaired are shown in Figure 10.

Fig. 10. Repairs with direct light-activated composite resin, in buccal margins of 46 (19 y) and 45 (8y). Both GIOs have been in function for 44 years (y = year).

Discussion

The high percentage of GIOs in function (81.7%) and their EMS were in line with other clinical studies[3,4,6]. Regarding statistical data previously presented in this paper, the variables age and gender did not influence the results. Secondary caries were 81.7% of the all failures, higher than in other studies: 33.7%[3] and 40%[4].

Occlusal carious lesions could be related to biomechanical factors associated with bruxism (marginal and/or surface wear of GIO, cement dissolution), biological factor (bacterial microfiltration) and the lack of periodic maintenance control. Besides, the operator, according to the minimal invasive criteria, preferred to repair before retreating. This agrees with Fisher and Morgan WW[45]. The fact that 63% of the patients did not attend at least once a year for maintenance therapy could be considered a biological and biomechanical risk factor for failure. Furthermore, parafunction might be a biomechanical risk factor of failure.

Only one GIO loosened (0.15%). This result was very low compared to other clinical studies: 32.7%[3] and 31%[4]. This could be due to the design of the tooth preparation, the quality of the restoration and a thorough fixation protocol by the operator.

The higher survival of premolars is in line with other studies[1,2]. Meanwhile, the higher survival of onlays could be explained because onlays transmit mainly compressive forces, giving better protection to the remnant tooth structure[46,47]. Besides, due to the extension, their margins could be less affected by the greater forces exerted in patients with bruxism. In addition, Studer et al.[4] did not find differences between inlays and onlays. The CS of the GIOs in function was 93.6%, in line with Donovan and Chee[5] and Donovan T et al.[6]. The development above supports the first objective of this work.

The second aim of this study was to analyze the indication and distribution of GIOs, related to the new scientific evidence. Therefore, both groups must be compared to analyze the constitution of Group B- and to assess the possible coexistence of restorations as substitutes of the GIOs.

In fact, the number of GIOs in function in Group B- (193) was 43.7% less than in Group A-. These patients had been using GIOs for a long time, they knew their results and did not refuse to use gold restorations. Nevertheless, when analyzing Table 3, 32.1% of GIOs were placed in the "transitional period" (1995-1999), 19.7% and 15.6% in the two following periods, respectively. This gradual and constant reduction in the use of GIOs might be due to

alternative restorative procedures proposed by the operator.

Using the same global sample, similar inclusion end exclusion criteria than in this work, and relative similar period of placement than restorations in Group B-, in two articles published by the authors[12,32] the following was studied:

- 93 IOCAs in 47 patients, placed between 10/1995 - 10/2008 and assessed in a 5 to 18 - year period. Eighty-seven onlays (93.5%) remained in function[12].
- 105 RCDPs in 61 patients were installed between 10/1993 - 10/2008 and assessed in a 5 to 20-year period; 103 (98%) were in function and 41% were large or extended restorations[32].

Therefore, in the 04/1991 - 10/2008 period, in the same global sample, 50.4% of the partial posterior restorations performed were GIOs and 49.6% RCDPs or IOCAs. Meanwhile, comparing the two groups, in Group B- we observed a reduction of 94.3% and 67.7% in the number of inlays and premolars restored with GIOs, respectively. This could be due to the application of more conservative, aesthetic and efficient restorative materials such as RCDPs.

Besides, the reduction of onlays (21.1%) in Group B-, might be linked to greater use of IOCAs. Whereas in Group B-, only one GIO failed due to periodontal problems. This result could be due to the scientific and clinical changes related to the new preventive actions taken by the operator[41-44].

Considering all the above, besides the very good clinical results, this work also showed a close connection between the clinical and conceptual changes applied by the operator and the updated scientific evidence.

Only one operator can reduce bias in the analytical comparison, but might improve the clinical success[48]. The results could not be reproducible, but the operator believes this could be a relative limitation[49].

Conclusions

In this study, a high clinical performance of GIOs was observed. However, as there are more aesthetic restorative materials, with good longevity and requiring simpler and less invasive teeth preparations, GIOs are less indicated by dentists and less accepted by patients.

Updated knowledge is an ethical responsibility of the professional. It will allow dentists to introduce the clinical and conceptual changes, taking into consideration the new scientific evidence, as was observed in this paper.

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Morphometric characteristics of multirooted teeth and furcation area

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Abstract

Objective. The aim of this study is to describe the morphometric characteristics of multirooted teeth at the furcation area. **Method.** Fifty-four multirooted teeth (maxillary and mandibular teeth) were evaluated using a precision calibrator: root trunk length, root separation, root divergence angle, root length, length of cervical enamel projections and presence of enamel pearls. **Results.** The divergence angle of the maxillary distal furcation (50°) was greater than on the buccal (22°) and mesial (37°) aspects. On the mandible it was 25° on the buccal aspect and 22° lingually. No enamel pearls were found. The cervical enamel projections most commonly found were class I on the oral aspect (60% for maxillary teeth and 31% for mandibular teeth). The length of the lingual root trunk was 2.8 mm, and on the buccal aspect it was 2.2 mm. **Conclusion.** The root trunk of mandibular molars is larger lingually than on the buccal aspect, as is the palatal area of maxillary molars. In maxillary molars the greater divergence angle appeared at the entrance of the distal furcation, which is the one recommended to start using mechanical instruments.

Keywords: furcation defects, periodontal disease, tooth root, periodontal attachment loss, periodontitis.

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Introduction

Multirooted teeth have unique anatomical features that pose a challenge to patients and clinicians because they cannot be properly accessed to effectively control dental biofilm. This condition can affect patients that are susceptible to periodontal disease because insertion loss may progress and involve the furcation area both vertically and horizontally.

Dental furcation is the anatomic area of a multirooted tooth where the roots diverge[1,2]. Anatomically it consists of a roof, the fornix (most coronal surface from the root separation) and the area of root separation[3,4].

It is essential to know the anatomy of multirooted teeth to make the right diagnosis and therapeutic decisions. For example, we know that the distobuccal root of the first mandibular molars and the distal root of maxillary molars have the smallest root surface area; this is why they are more likely to undergo root extraction[4]. Bower[5] found that 81% of furcations have an entrance < 1 mm, and 58% an entrance < 0.75 mm. The width of a traditional curette is 0.75 mm, so its use is not enough in this area. Chiu[6] found that 49% of molars have a < 0.75 mm entrance on average. Additionally, the furcation entrance through the vestibular area is smaller than through the lingual area in mandibular molars[5]; mechanical instruments are harder to use lingually, so furcation involvement is more likely to occur.

Furcation invasion or involvement is the pathological resorption of interradicular bone with the subsequent furcation exposure[1]. This is a condition that increases the risk of bone loss, causing bone defects which are difficult to treat given the complex and irregular anatomy of the area[7]. It is important to know the anatomical features of multirooted teeth such as root trunk length, root divergence angle, root separation, and to observe cervical enamel projections and enamel pearls to control the morphological predisposing factors that lead to furcation involvement. In this study we evaluated the morphometric characteristics of multirooted teeth that are related to dental furcation.

Method

Descriptive *in vitro* observational study that included 54 multirooted teeth (first, second and third mandibular and maxillary molars). The teeth were selected from the Tooth Bank of the Dental Students' Scientific Society, School of Dentistry, Universidad Nacional Mayor de San Marcos, Lima, Perú. The teeth had to be in good morphological conditions from the cementoenamel junction to the root apex. We excluded the teeth with structures that made it difficult to see the root area (hypercementosis, fractures, concavities, etc.). We also excluded teeth with open apices or loss of structure in the tooth roots (root resorption). The teeth were washed with saline solution and scaled at the root surface with 5/6 and 7/8 Gracey curettes to see their anatomical features more clearly.

The characteristics evaluated included: root trunk length (RT, evaluated from the cementoenamel junction to the furcation entrance); root separation (RS, apical distance between the roots at the furcation level); root divergence angle (RD, angle formed by the roots at the furcation entrance); root length (RL, from the cementoenamel junction to the apex of each root) (Fig. 1). We also studied the presence/absence of enamel pearls (EP) as well as of cervical enamel projections (CEP) and classified them following Master's[8] categories: Class I (enamel projection < 1/3 of the root trunk), Class II (enamel projection > 1/3 of the root trunk without contact with the furcation) and Class III (enamel projection > 1/3 of the root trunk that extends to the furcation area).

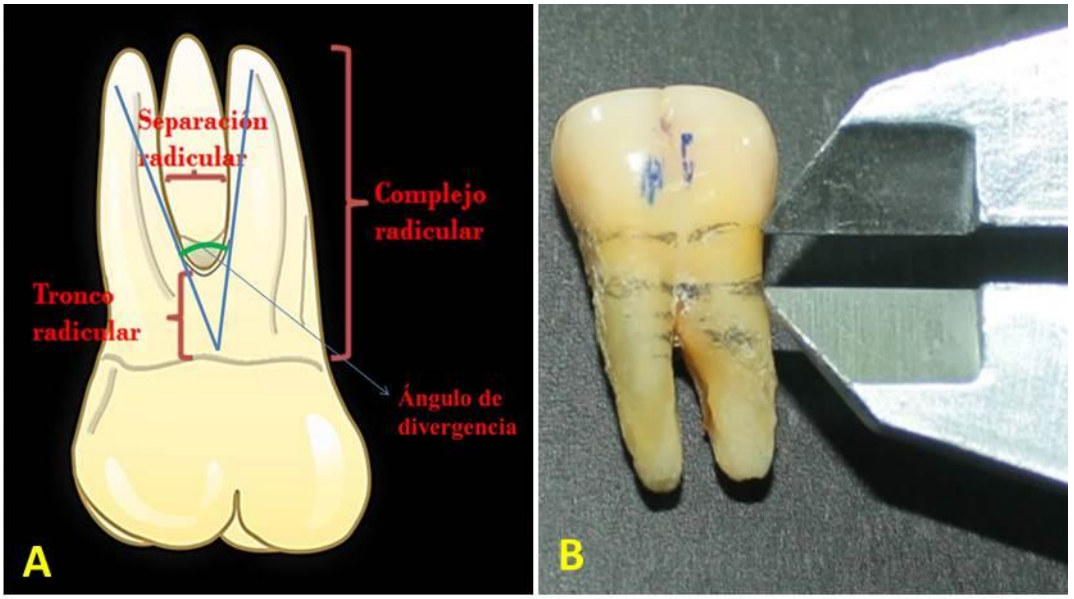

Figure 1. A. Main variables analyzed. B. The measurements were recorded with a precision calibrator.

The measurements were taken with a Mitutoyo© verner caliper and calibrated to an accuracy of 0.01 mm. The measurements were taken on the buccal and lingual/palatal aspects by a rater (YC); the comments on the extent of CEP and EP were made by two raters (YC and KS; interrater reliability Kappa = 0.96).

The data were processed with the SPSS 21 statistical package (IBM, United States). We used descriptive statistics and measures of central tendency and dispersion (mean and standard deviation). The differences between tooth types in the quantitative measurements were set with the ANOVA test for independent groups for mandibular molars; while the Student t test was used for independent groups for maxillary molars. The differences in the qualitative assessments between the groups were set applying the Mann-Whitney U test. A statistical significance of 0.05 was set to disprove the null hypothesis.

Results

Regarding mandibular molars (vestibular aspect), dimensions were greater for the second molar with an average root separation of 2.11 mm ($p < 0.05$), a divergence angle of 27.5° ($p > 0.05$) and a root length of 14 mm ($p < 0.05$). The root trunk of the first molar was greater than the second and third molars with an average of 2.6 mm ($p < 0.05$) (Table 1).

Table 1. Morphometric characteristics of mandibular molars on the buccal surface

Tooth	Buccal characteristics			
	Root trunk	Root separation	Divergence angle	Root length
First Molar	2.59 ± 0.79mm*	1.81 ± 0.27mm	25.77 ± 3.75°	12.68 ± 0.69mm
Second molar	1.63 ± 0.32	2.11 ± 0.72*	27.56 ± 11.31	13.97 ± 1.35*
Third molar	2.5 ± 0.5	1.26 ± 1.15	10 ± 9.16*	10.54 ± 0.43
Total	2.18 ± 0.75 mm	1.86 ± 0.67	24.48 ± 9.9	12.9 ± 1.48

***p < 0.05 ANOVA for independent groups. Tuckey's Post Hoc test.**

On the lingual surface, root trunk, root separation, divergence angle and root length were greater in the first molar, unlike the second and third molars (p > 0.05 (Table 2).

Table 2. Morphometric characteristics of mandibular molars on the lingual surface

Tooth	Lingual characteristics			
	Root trunk	Root separation	Divergence angle	Root length
First Molar	2.9 ± 0.44mm	1.79 ± 0.33mm	23,88 ± 4,51°*	13.69 ± 0.71mm
Second molar	2.7 ± 0.33	1.72 ± 0.86	24.12 ± 9.42	13.03 ± 0.84
Third molar	2.62 ± 0.44	1 ± 0.91	10.4 ± 9.52	9.68 ± 1.58*
Total	2.78 ± 0.41mm	1.66 ± 0.71	22.25 ± 8.65	12.91 ± 1.56

*p < 0.05 ANOVA for independent groups. Tuckey's Post Hoc test.

On the vestibular area of maxillary molars we found a greater root length on the second molar (3.7 mm; p > 0.05) and higher root length (12 mm; p > 0.05). The first molar had greater lengths in the root separation and divergence angle (p > 0.05) (Table 3).

Table 3. Morphometric characteristics of maxillary molars on the buccal surface

Tooth	Buccal characteristics			
	Root trunk	Root separation	Divergence angle	Root length
First Molar	3.42 ± 0.47mm	2.22 ± 0.11mm	23 ± 5.16°	11.83 ± 0.71mm
Second molar	3.72 ± 0.73	1.99 ± 0.58	20.6 ± 7.89	12.15 ± 1.28
Total	3.52 ± 0.57mm	2.15 ± 0.34	22.2 ± 6.02	11.93 ± 0.9

The teeth selected did not include a third maxillary molar. On the palatal aspect we found greater dimensions of root separation at the distal furcation on the first molar, as well as greater divergence angles on the distal aspect on the first and second molars (p > 0.05) (Table 4).

Table 4. Morphometric characteristics of maxillary molars on the palatal surface

Tooth	Palatal characteristics						
	Mesial root trunk	Distal root trunk	Mesial root separation	Distal root separation	Mesial divergence angle	Distal divergence angle	Root length
First Molar	3.92 ± 0.56mm	3.3 ± 0.41mm	3.2 ± 0.65mm	2.92 ± 0.45mm	38.3 ± 7.27°	55 ± 16.2°	13.37 ± 0.6mm
Second molar	3.81 ± 0.52	3.78 ± 0.72	2.9 ± 0.81	2.52 ± 0.65	33.8 ± 10.3	40.2 ± 20.31	13.42 ± 0.71
Total	3.89 ± 0.53	3.46 ± 0.56	3.1 ± 0.7	2.78 ± 0.53	36.8 ± 8.31	50.06 ± 18.4	13.39 ± 0.61

We did not find any enamel pearls on the root surface, neither on the vestibular nor the lingual/palatal areas. Class I cervical enamel projections were predominant: they appeared on 60% of maxillary on the vestibular aspect, and on 31% of mandibular molars. We found no predisposition of cervical enamel projections according to type of tooth and the area assessed ($p > 0.05$) (Table 5).

Table 5. Characteristics of the cervical enamel projections found

Cervical enamel projection	Buccal area			Lingual/palatal area		
	Maxilla	Mandible	Total	Maxilla	Mandible	Total
Absent	6 (40%)	24 (61.5%)	30 (55.6%)	15 (100%)	34 (87.2%)	49 (90.7%)
Class I	9 (60%)	12 (30.8%)	21 (38.9%)	0	5 (12.8%)	5 (9.3%)
Class II	0	3 (7.7%)	3 (5.6%)	0	0	0
Class III	0	0	0	0	0	0
Total	15	39	100	15	39	54

Discussion

Incomplete removal of dental biofilm reduces the success rate of periodontal treatment; multirooted teeth have special anatomical features that prevent the success of the treatment. We know that cervical enamel projections increase biofilm accumulation; additionally, during regenerative therapy they hinder the formation of new connective tissue[9]. Our study found no CEPs on the root surface, and the general prevalence is low; Moskow[10] reports an incidence of 2.6%. If they appear, their removal through odontoplasty is recommended as they are a predisposing factor for furcation involvement.

First maxillary molars have a smaller buccal furcation entrance than the mesial and distal entrances[5]. They have a root trunk ≥ 3 mm[11], with an average of 3.5 mm3.6 mm mesial, 3.5 mm4.2 mm buccal and 4.1 mm4.8 mm distal[12]. In this study the average root trunk for the first molar was 3.4 mm on the buccal aspect, 3.9 mm on the mesial aspect and 3.3 mm on the distal aspect. Knowing these data about the root trunk allows us to predict the development or predisposition to furcation involvement. Molars with short root trunks are more vulnerable to furcation exposure but have a better prognosis after treatment (they provide easier access and use of instruments) when the tooth does not have severe periodontal destruction. These teeth have a smaller area for periodontal adherence[13]. Short trunks are associat-

ed with long roots and thus facilitate resective therapy[13].

In the onset of periodontal disease, molars with long root trunks have a better prognosis as the furcation position is protected; but if the disease progresses, the prognosis is not favorable[13]. Long trunks are associated with short roots and therefore are not candidates for resective therapy, and tend to have greater loss of periodontal support and furcation invasion[2].

Regarding mandibular molars, we found an average buccal root trunk of 2.2 mm, and of 2.8 mm on the lingual aspect. These data are similar to those of Santana et al.[14] (2.8 mm and 3.5 mm on the buccal and lingual aspects respectively). Root separation and divergence angle were higher in lower second molars; this is favorable because it allows for the use of curettes in the area. As the lingual area has greater dimensions, furcation involvement does not progress; however, once present, access through mechanical instrumentation is limited. According to Marcaccini et al.[13], the lingual furcation of mandibular molars has a smaller entrance and a longer root trunk. This suggests a stronger limitation for instrumentation in the area and possibly a worse prognosis for periodontal disease.

Regarding CEPs, they are frequent and difficult to detect in healthy teeth. There is a direct link between them and furcation involvement[15]. Hou[16] found that 63% of molars with furcation involvement had cervical enamel projections, which shows that they are more prevalent in first and second molars. They are considered a cofactor in furcation involvement. Master[8] found a 28.6% incidence on mandibular molars and 17% in maxillary molars, finding a correlation in the development of furcation involvement in mandibular molars. However, Leib[17] did not find such correlation. In this study we found a low CEP frequency. The most common type was class I on the buccal aspect in maxillary and mandibular first molars.

Conclusion

The root trunk of mandibular molars is larger lingually than on the buccal aspect, as is the palatal area of maxillary molars. The divergence angle in the buccal area of mandibular molars was 24.5° and in maxillary molars it was 22°. In maxillary molars, the greatest angle appeared at the entrance of the distal furcation, making this the best entrance to start using mechanical instruments. We found no enamel pearls, and class I cervical enamel projections were the most frequent ones, mainly on first molars.

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Methodological quality of orthodontic clinical trials, 2012-2016

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Abstract

Objective: To determine the methodological quality of orthodontics clinical studies in six journals with the highest impact indexed by the Institute for Scientific Information (ISI), between January 2012 and December 2016.

Methodology: We performed a systematic search in PubMed and we reviewed the abstract and methodology of all the selected studies. We classified as clinical trials the studies that evaluated the effectiveness of a treatment, comparing two or more interventions to a control group.

Results: We reviewed 221 clinical trials with the MinCir scale. Regarding methodological quality average, the European Journal of Orthodontics showed the best average score (15.6 ± 2.83). Regarding continent results, Europe has the highest average score of methodological quality with 14.88 points.

Conclusion: The methodological quality of the clinical trials in orthodontics indexed in ISI journals presented some deficiencies. Therefore, we suggest this information should be considered to analyze its implications for orthodontics practice.

Keywords: clinical trial, orthodontics, malocclusion, dentistry, publication bias.

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Introduction and background

Clinical trials are conducted to learn about the effectiveness of a treatment by testing it on humans. These studies are the basis for good decision-making about treatments since, according to evidence-based health care practice, clinicians should be able to apply the findings to their clinical practice after reviewing these studies. However, due to the methodological flaws in some studies of this and other kinds, the results obtained cannot be replicated or, even worse, cannot be extrapolated to reality[1]. In dentistry, publications do not necessarily have the clinical relevance the discipline demands, and in many cases, errors may occur in its methodology[2]. Therefore, it is essential to know whether studies have been properly conducted, considering the variables which may affect their results and interpretation[1].

One of the main purposes of orthodontic trials is to assess interventions and reach valid conclusions about recommended treatment modalities[3]. Nowadays, however, only a small portion of the clinical trials reported in orthodontic literature are randomized clinical trials (RCTs), while the rest are prospective or retrospective nonrandomized clinical trials, with the latter being of a lesser methodological quality and, often, the basis for the decisions of clinicians[4]. Randomized clinical trials provide valid results if the bias is reduced or absent. It has been reported that the absence of a series of factors such as control, randomization, allocation concealment, blinding, and accounting for loss to follow-up in trials can introduce a bias and, thus, invalid results[5]. In view of this, it is important to conduct a continuous analysis of the scientific production in the field of orthodontics, as in other health science disciplines.

Bibliometric studies are tools used to provide quantitative analyses of the scientific production[6] and also report on current publication trends to expand, compare and improve knowledge[7]. Thus, an analysis of biometric indicators provides objective information about what is being published and the chance of watching the trends followed by each journal. The data obtained and the conclusions reached can help editors improve the management of their journals, by facilitating the decision-making process on things such as article selection. There are instruments designed and validated for analyzing scientific production in dentistry. One of them is the MinCir scale (MS) which evaluates the methodological quality (MQ) of the articles by analyzing research design, sample size and methodology[8], which is an objective assessment of the quality of the methodology used in the clinical trials published on this area.

Malocclusions are the third most prevalent alterations with the third greatest psychosocial impact[9]. According to Bilgic (2014), almost a third of the population evaluated have a very great need of orthodontic treatment[10]. In addition, a study conducted in Chile found that 96.2% of students examined had some kind of malocclusion[9]. Given the global importance of orthodontics issues, we must learn about the reality of this scientific field and form of knowledge dissemination. Therefore, the objective of this study is to describe the MQ of orthodontics clinical studies in six journals with the highest impact indexed by the Institute for Scientific Information (ISI), between January 2012 and December 2016.

Materials and methods

This study evaluated clinical trials published in the six journals with the highest impact factor (IF) between January 2012 and December 2016. All the journals assessed are indexed in the ISI Web of Knowledge, under the Dentistry, Oral Surgery & Medicine category. We selected journals featuring clinical trials published within the period evaluated, written in English and which included the words "orthodontic" and "orthopedics" in their titles. The journals selected were: American Journal of Orthodontics and Dentofacial Orthopedics (AJODO)

IF=2.201, Angle Orthodontics (AO) IF=1.717, Orthodontics Craniofacial Research (OCFR) IF=1.617, European Journal of Orthodontics (EJO) IF=1.627, Journal of Orofacial Orthopedics/Fortschritte der Kieferorthopädie (JOOF) IF=1.073 and Australian Orthodontic Journal (AOJ) IF=0.452.

In order to select the articles in each journal, an advanced search was conducted in the Medline database, using the NLM Catalog: Journals referenced in the NCBI Databases. The following search filters were used: 1) Journal, 2) Article types: Clinical Study, Clinical Trial, Clinical Trial, Phase I, Clinical Trial, Phase II, Clinical Trial, Phase III, Clinical Trial, Phase IV, Multicenter Study and 3) Publication dates: 01 Jan 2012 to 31 Dec 2016.We reviewed the abstract and methodology of all the studies found. We classified as clinical trials those that evaluated the effectiveness of a treatment, comparing it to two or more interventions and using a control group. All articles that did not meet these criteria, even if they had been indexed in this manner in the NCBI database, were excluded.

All the studies selected were analyzed using an adaptation of the MS scale[2] (Fig. 1), following the instructions set out by the authors[11]. Of the three domains that comprise the scale, only two were considered for this work: the first one, which evaluates the type of study design and the third one, which consists of four items that assess the methodology used in the study[2]. The Domain 2: Studied population by justification factor was not included. Thus, the final score varies between 10 and 24 points, minimum and maximum respectively.

The methodological quality evaluation of each article was conducted independently by each reviewer. The inter-reviewer kappa value obtained during the calibration process was 91.90%. It was calculated applying the MS evaluation instrument independently to 11 clinical trials selected randomly from the same journals selected for this work. Those articles were not included in this analysis. The articles analyzed were classified according to their geographical origin (continent), subject matter (corrective orthodontics, interceptive orthodontics, orthopedics, preventive orthodontics, orthodontic-surgical and others) and the journals included. The data were tabulated in a Google Docs electronic form (Mountain View, CA, USA). The statistical analysis was conducted using the R program, version 3.3.4, which showed significant differences in scores according to geographical origin. Levene's test was used to check the homogeneity of score variances and, since p value > 0.05, homogeneity of variances is assumed. The Shapiro-Wilk test was then used to check the normality of the data, and p values < 0.05 were obtained, therefore, the Kruskal-Wallis test for non-parametric samples was used. The post hoc pairwise comparison with pooled SD was then used to look at the differences between continents.

	Assigned Score
Domain 1. Study Design	
Multicenter clinical trial	12
Double-masked, randomized controlled clinical trial	9
Clinical trial (simple or without masking; not randomized)	6
Domain 3. Methodology used	
Item 1. Objectives	
The objectives are clearly and specifically presented	3
The objectives are vaguely presented	2
The objectives are not presented	1
Item 2. Design	
The design used is mentioned and justified	3
The design used is mentioned	2
The design used is neither mentioned nor justified	1
Item 3. Sample selection criteria	
Inclusion and exclusion criteria are described	3
Inclusion or exclusion criteria are described	2
Inclusion or exclusion criteria are not described	1
Item 4. Sample size	
Sample used is justified	3
It does not justify the sample used	1

Table 1. MinCir Methodological Quality Scale, modified for clinical trials.

Results

Study selection. A total of 271 articles were found: 89 articles which were defined as clinical trials in the AJODO journal, 49 in the EJO journal, 70 in the AO journal, 38 in the JOOF journal, 10 in the AOJ journal, 16 in the OCFR journal, and 50 articles were excluded because they did not meet the selection criteria to be classified as clinical trials. All of them were obtained as full text (Fig. 1).

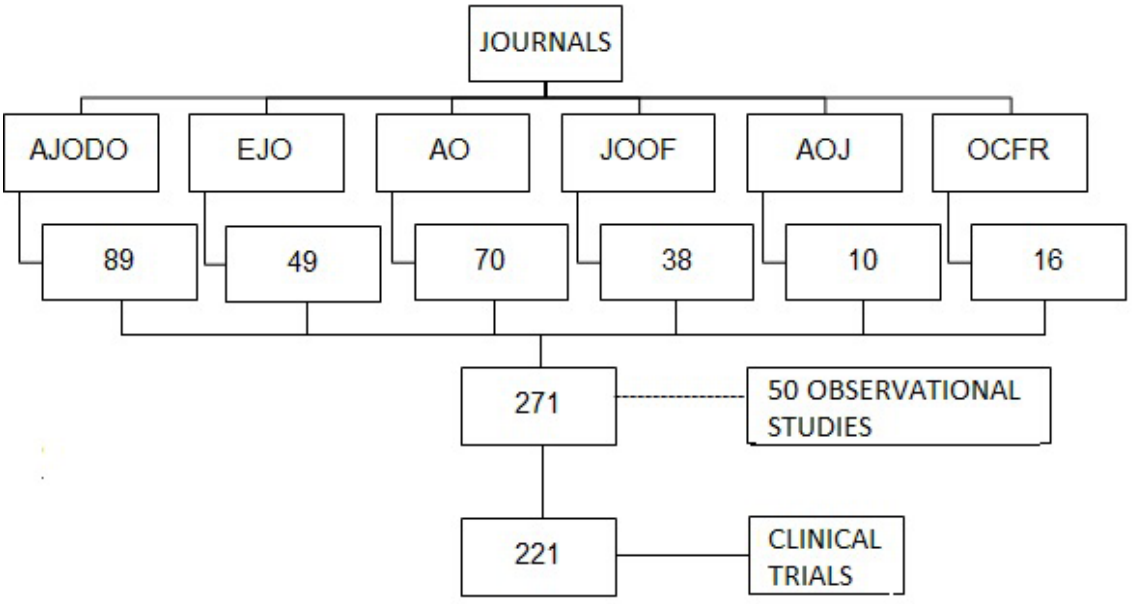

Fig. 1. Flow chart of the number of articles obtained.

Characteristics of the journals. The journal with the largest number of clinical trials published is AJODO, with 83 articles indexed under this category, whereas the journal with the fewest clinical trials published is AOJ, with only 7 articles (Fig. 2).

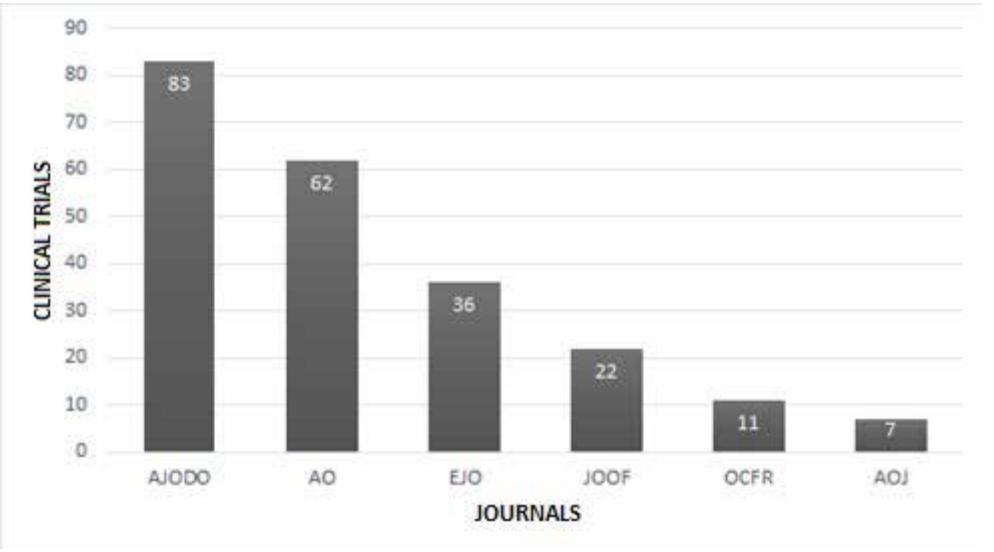

Fig. 2. Number of clinical trials published per journal from January 2012 to December 2016 selected.

As for the geographical origin, the continent with the largest number of clinical trials published was Europe, with 100 articles (45.25%), and the one with the fewest articles published was Africa, with 5 articles (2.26%). The MQ assessment shows the same distribution by continent. Europe has the highest MQ average with 14.88 points in the MS, while the continent that had the lowest MQ score was Africa, with an average of 11.8 points (p = 0.003) (Table 2) (Fig. 3).

Continent	n	(%)	Score	SD
Europe	100	45.25	14.88a	2.8
Asia	64	28.96	14.31b	2.22
Oceania	7	3.17	14c	2
North America	20	9.05	13.55d	1.47
Latin America	25	11.31	13.44e	2.24
Africa	5	2.26	11.8f	0.84
TOTAL	**221**	**100**		

Table 2. Number of clinical trials published per continent and mean Methodological Quality score per continent.
Kruskal-Wallis test post-hoc Pairwise comparison with pooled SD (p = 0.003)
f < a and b; c > d and e

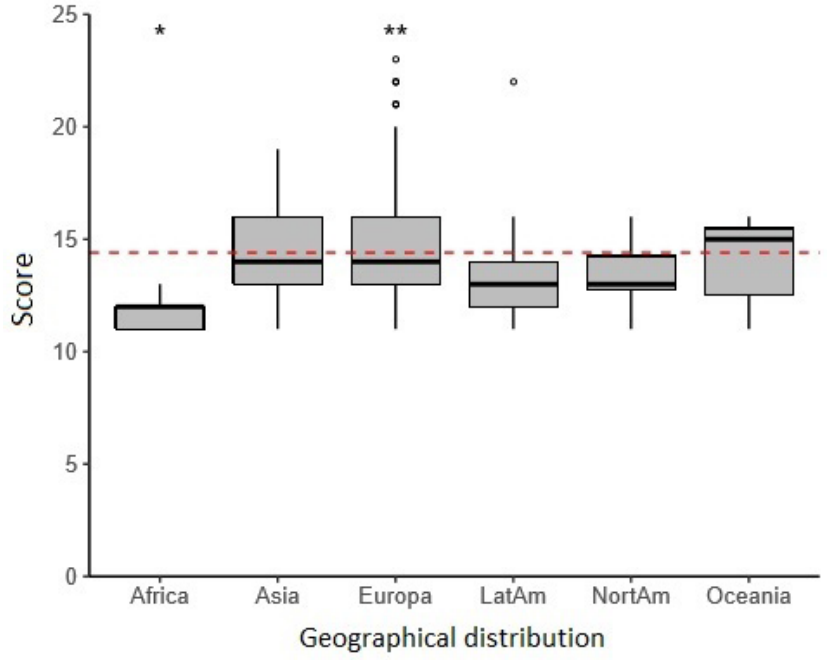

Fig. 3, methodological quality score, according to geographical distribution. The cut-off point is 60% (14.4 points) in the MinCir scale, represented in the chart with the red dashed line.

None of the articles analyzed obtained the maximum or the minimum score in the MS. The minimum score reached was 11 points, and the maximum was 23 points, which was an article in the EJO journal.

As for individual journals, EJO had the highest average score (15.6 ± 2.83), and JOOF had the lowest average score (13.09 ± 1.72) (Table 3).

Journal	N	%	Score	SD	5-y IF
European Journal of Orthodontics	36	16.29	15.6	2.83	1.627
American Journal of Orthodontics and Dentofacial Orthopedics	83	37.56	14.67	2.7	2.201
Orthodontics Craniofacial Research	11	4.98	14.27	2.45	1.617
Australian Orthodontic Journal	7	3.17	14	2.16	0.452
Angle Orthodontics	62	28.05	13.88	2.05	1.717
Journal of Orofacial Orthopedics / Fortschritte der Kieferortho-pädie	22	9.95	13.09	1.72	1.073
Total	**221**	**100**			

Table 3. Clinical trials published per journal and mean Methodological Quality score per journal.
5-y IF: 5-year impact factor
SD: standard deviation
Cut-off point: 14.4 points

The first domain of the MS considers the type of clinical trial conducted, of which the *clinical trial (simple or without masking; not randomized)* was the most common one in orthodontics, with 189 articles (85.52%); and *multicenter clinical trial* was the least frequent type with 15 articles (6.79%) (Table 4).

		n	(%)
Clinical trial	Multicenter clinical trial	15	6.79
	Double-masked, randomized controlled clinical trial	17	7.69
	Clinical trial (simple or without masking; not randomized)	189	85.52
Design	The design used is mentioned and justified	3	1.36
	The design used is mentioned	136	61.54
	The design used is neither mentioned nor justified	82	37.1
Selection	Inclusion and exclusion criteria are described	79	35.75
criteria	Inclusion or exclusion criteria are described	113	51.13
	Inclusion or exclusion criteria are not described	29	13.12

Table 4. Number of articles classified per type of clinical trial, design and selection criterion.

As for design analysis, only 61.54% mentioned the design, while the design used was mentioned and justified in 1.36% of cases. In the remaining percentage, the design used was neither mentioned nor justified.

Regarding the selection criteria item, inclusion or exclusion criteria are described in 52.38% of studies, and they are neither described nor mentioned in 12.86% of articles *(Table 3)*.

When analyzing the objectives set out in the articles using the MinCir scale we found that vague objectives were set out in 96.77% of the clinical studies published in the AO journal. On the other hand, 100% of the works of the of the same kind published in the JOOF, AOJ and OCFR journals reached a score of 2 in this item, that is, their objectives were set out vaguely *(Table 5)*.

Journals	The objectives are clearly and specifically presented		The objectives are vaguely presented		The objectives are not presented		Total	
	n	%	N	%	n	%	n	%
American Journal of Orthodontics and Dentofacial Orthopedics	0	0	80	96.39	3	3.61	83	37.56
European Journal of Orthodontics	1	2.78	34	94.44	1	2.78	36	16.29
Angle Orthodontics	0	0	60	96.77	2	3.23	62	28.05
Journal of Orofacial Orthopedics / Fortschritte der Kieferorthopädie	0	0	22	100	0	0	22	9.95
Australian Orthodontic Journal	0	0	7	100	0	0	7	3.17
Orthodontics Craniofacial Research	0	0	11	100	0	0	11	4.98
Total	**1**	**0.45**	**214**	**96.83**	**6**	**2.71**	**221**	**100**

Table 5. Number of clinical trials published per journal per presentation of the study objective.

Finally, in the sample size item, the EJO journal stood out because the sample used was justified in 63.89% of the articles (Table 6).

Journals	Sample is justified		Sample is not justified		Total	
	N	%	n	%	n	%
American Journal of Orthodontics and Dentofacial Orthopedics	38	45.78	45	54.22	83	37.56
European Journal of Orthodontics	23	63.89	13	36.11	36	16.29
Angle Orthodontics	24	38.71	38	61.29	62	28.05
Journal of Orofacial Orthopedics / Fortschritte der Kieferorthopädie	3	13.64	19	86.36	22	9.95
Australian Orthodontic Journal	2	28.57	5	71.43	7	3.17
Orthodontics Craniofacial Research	4	36.36	7	63.64	11	4.98
Total	**94**	**42,53**	**127**	**57,47**	**221**	**100**

Table 6. Number of clinical trials published per journal per justification of the sample size.

Discussion

The subject matter was defined as the area of knowledge related to orthodontics the paper was dedicated to. Articles were then grouped as follows: preventive, corrective, interceptive orthodontics, orthopedics, surgical orthodontics and other topics, only for the purpose of organizing the information.

The instrument used was the MS, which has been shown to have adequate psychometric properties, which makes it a valid option for assessing the MQ in dental therapy research articles[8]. In this study we used an adaptation of the MS and assessed two of the three domains of the scale. Domain 2, which looks at the sample size, was excluded because it assigns a high score to studies with a larger number of participants, and a lower score to those with a smaller number of participants, although we did not prove the existence of a proportional relation between the number of subjects included in a sample and the quality of the study. Thus, according to this domain of the MS, a double-blind, randomized, multicenter clinical trial comparing two interventions can require 100 patients (6 points) or less to obtain valid results, while a study of a series of cases can include 500 patients (12 points) and still produce low-quality results. Cartes et al. propose, as a solution to this issue, giving more weight to the justification of the sample size, than to the number itself[13].

A geographical analysis of the scientific production has shown that the best indicators for medical and dental research can be found in Europe, North America, Australia and Oceania, versus Asia, Africa and South America[12]. Nevertheless, our results showed that research in orthodontics is more developed in Europe and Asia, since these two continents stand out both regarding the number of studies published and their quality.

The MS adaptation provided a score ranging from 10 to 24 points, and the average MS score for the clinical trials in the six journals analyzed was 14.33. The cut-off point used for determining the MS was 14.4 points. A score above 14.4 is considered a good MQ, therefore, Europe and Asia were the continents that stood out the most with the largest number of studies over the cut-off point.

It is also worth noting that the IF for each journal is not directly related to the average MQ

of its clinical trials, as the AJODO journal has the highest IF of 2.201, but did not reach the highest score, with only 14.67 points. This can be interpreted in many ways, the first of which is that perhaps the authors who are citing the papers in that journal are not aware of the fact that the methodological designs used to conduct the clinical trials are not optimal and/or they prioritize other aspects that they believe to be more important at the time of citing research. It could also be concluded that authors are aware of the poor methodological quality of these trials and are citing them to emphasize the mistakes in those publications, or that the authors know that the methodological quality of the clinical trials is deficient, but they are citing other kinds of studies published in those journals whose methodological quality is good. In view of this, we suggest conducting systematic reviews of the different therapies used currently in orthodontics.

Another interesting finding in this analysis is that in the MS objectives, those that were presented were, mostly, vague (96.83%). This is explained by the fact that the MS requires for an objective to be considered clear and specific to include what will be measured, by what means, by whom, where and when the measurement will take place. In the case of a therapeutic procedure or intervention, the objective must state what it is, whether it has a comparator, who applies it, to whom it is applied, when and how it is applied[11]. It is also worth noting that if any of the criteria detailed above are not met, the objective is classified as vague. Only one article complied with all of these items, the others usually only mention what will be measured, when it will be measured, the therapeutic intervention and the comparison.

Among the different study designs, the RCT is considered to potentially provide the best quality evidence, but there is substantial evidence in the biomedical literature that the quality of the RCTs published is suboptimal[1]. In this study we found mostly (85.52%) RCTs in which the study design was not explicitly mentioned, leading the reader to infer this information, which can cause interpretation problems with less experienced readers. In addition, multicenter studies were the least, which could be due to the complexity of their execution: taking into account all the parameters that should be considered for the procedure standardization, planning, execution and funding[2].

Masking is considered one of the most important procedures to obtain impartial results. We must also remember that it is not always possible to blind participants and/or reviewers; it is often possible to blind the reviewers of results and data analysts[1]. This is what happened in most cases.

According to the parameters set by evidence-based dentistry, clinical decision-making must be based on the best and most current evidence available, yet there are few studies focused on evaluating this evidence, which leaves unexperienced readers with the dilemma of whether to accept or not the data published. Therefore, we recommend conducting further studies to assess other areas of dental sciences, to provide data that will guide the decision-making process of readers to determine how reliable and applicable to other cases the results found in a scientific paper can be.

Conclusions

Implications for clinical practice. After analyzing the methodological quality of the clinical trials in orthodontics indexed in ISI journals between 2012 and 2016, and concluding that it presented some deficiencies when the MinCir scale is applied, we suggest this information should be considered to analyze its implications for orthodontics practice.

Implications for research. We suggest researchers conduct controlled trials taking into account the methodological quality, including, for example, masked examiners to reduce biases.

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Prevalence of signs and symptoms of temporomandibular disorders in a young population at the beginning of treatment for drug dependence

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Abstract

Introduction:
Temporomandibular disorders (TMD) is a collective term that includes several clinical problems affecting the muscles of mastication, temporomandibular joints and other related structures. Consumption of licit and illicit drugs may be an etiological agent of TMD which should be considered when diagnosing a patient and developing a treatment plan.
Objective:
To estimate the prevalence of TMD signs and symptoms in a drug-dependent population at the beginning of their admission, and to analyze possible associated factors.
Materials and methods:
This was a descriptive cross-sectional study. Inclusion criteria: a) age range: from 15 to 35; b) beginning of treatment for problematic drug use; exclusion criteria: a) non-lucid patients: b) non-collaborative patients. We studied a sample of 135 subjects admitted to Portal Amarillo, Uruguay, under a residential system. The patients were examined by a calibrated researcher. The Uruguayan Survey Sheet for Prevalence of Bruxism and TMD was used. Informed consent was requested from each participant and/or their legal representative.
The prevalence of subjects with one or more current TMD signs was 29.7% (19.3% showing one sign, 8.9% two, and 1.5% three); those with one or more current TMD symptoms accounted for 68.8% (45.9% with one, 14.8% with two, and 8.1% with three) of the patients. The prevalence of current TMD symptoms showed a significant association with self-perceived stress (p=.03), mate consumption (p=.03) and alcohol consumption (p=.03).
Conclusions: The prevalence of current TMD symptoms in the population under treatment for drug dependence is high compared to the general population. This should be taken into account when developing prevention and therapeutic strategies.

Keywords: prevalence, temporomandibular disorders, drug dependence.

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Introduction

The American Academy of Orofacial Pain defines Temporomandibular Disorders (TMD) as a collective term that includes several clinical problems affecting the muscles of mastication, temporomandibular joints and other related structures[1]. TMD are a subgroup of musculoskeletal disorders suffered by the body, with a multifactorial etiology[2]. Occlusal, structural, psychological, traumatic factors, and general health conditions are risk factors which may be considered to predispose to, trigger, and perpetuate TMD[3]. The diagnosis and therapeutic issues which arise from TMD should not be limited to the dental-occlusal component, as it was suggested a few years ago[4]. Recent studies have shown that static and dynamic occlusal conditions may not be the most relevant aspect in TMD development[5].

Oral parafunctional habits such as bruxism, biting of the lips, cheeks, and objects, nail biting, thumb sucking, and gum chewing could be associated to signs and symptoms of TMD in children and teenagers[6]. A nationwide study conducted in Uruguay analyzed signs, symptoms, and potential factors linked to bruxism and TMD. It showed a high prevalence of TMD and bruxism in the populations under study, both in Montevideo and in the rest of the country: there was 55% of prevalence of at least one symptom of TMD, and 44% prevalence of at least one clinical sign[7]. In a similar study, the most frequent findings were limitation of mouth opening, asymmetrical mandibular movements, and TMJ noises[1]. A comparative study showed that pain on palpation of the muscles of mastication, the muscles adjacent to the neck, and the TMJ is more frequent in the case group (in treatment for TMD) than in the control group[8]. Other epidemiological reports have shown a high prevalence of recurrent tension headaches (episodic or chronic) in individuals suffering from TMD in relation to the control group without TMD, 95% versus 34%[9]. Surveys conducted among children and teenagers link TMD symptoms to orofacial and cervical pain[10]. Sleep is an important factor in the restoration of functions, and is also related to hormonal changes. Sleep disruptions such as bruxism, insomnia, and apnea could be indicators of the risk of TMD[11].

According to the World Health Organization (WHO) "a drug is any natural or manmade substance which, when taken into the body, alters one or more of its functions". Psychotropic or psychoactive drugs are those which act mainly on the central nervous system (SNC), altering mood, behaviors, consciousness, and perceptions[12]. These substances have been present in every culture in the history of humankind, and they are used for religious, cultural, recreational, and medicinal purposes. Any use of substances, whether licit or illicit, poses risks that vary depending on age, sex, forms and frequency of use, personal context and circumstances[13].

The term "abuse" refers to misuse or use for non-medical purposes, regardless of the consequences. Substance abuse generally refers to problems related to the use of psychoactive substances. The WHO has recommended replacing "abuse" with "dependence"[14].

Although initial and exploratory use may be voluntary, once the addiction has developed, this control disappears abruptly[15]. When under the influence, addicts undergo a series of changes that affect their physical, mental, and social state. When the dependent individual stops using one or more drugs, they experience the withdrawal syndrome, whose effects and duration vary depending on the substance used[16]. The withdrawal period during the drug-addiction treatment is particularly critical for dental health, mainly due to poor hygiene habits[17]. After a period of intense cocaine use, withdrawal is evidenced in a strong desire to use the drug, an extreme need to sleep, suicidal thoughts, and paranoia. The recovery process for individuals who use drugs usually involves

psychiatric treatment for anxiety, restlessness, and hallucinations[18].

A study conducted in a care center for drug-dependent people in Israel, which compared an addicted group to a non-addicted paired sample, showed a high prevalence of TMD signs and symptoms, sleep bruxism and daytime teeth clenching in the addicted population under treatment, as compared to the control group[19].

In this study we used the working hypothesis that the population under treatment for drug dependency would register a higher prevalence of symptoms and clinical signs of TMD as compared to the general population. The intended aim was to estimate the prevalence of TMD signs and symptoms among teenagers and adults at the start of the treatment for drug abuse, as well as to analyze possible associations between different factors and current TMD signs/symptoms.

Materials and methods

The study included 135 patients (25 women, 108 men, and 2 transgender people, averaging 23 years of age) on a residential basis, being treated at Portal Amarillo (Uruguayan Information and Reference Center of the Drug Network, which provides treatment for young users of the State Healthcare Administration in the Republic of Uruguay)[20]. This was a descriptive cross-sectional study. The inclusion criteria used were: a) age range between 15 and 35, b) being at the inception stage of their treatment for problem drug use. The exclusion criteria were: a) nonlucid patients, b) noncooperative patients. The size calculation made based on the difference in proportions was 135 subjects.

The study used the Uruguayan Survey Sheet for Prevalence of Bruxism and TMD[7]. The variables considered in the examination for current TMD symptoms were: pain when opening the mouth, blockage of the jaw when opening the mouth, noise in the joints, and headaches. The clinical variables considered for signs of current TMD were: mouth opening less than 40 mm, pain on palpation of the muscles and pain on palpation of the TMJ.

Bias control

The operator was trained and calibrated (Kappa index 0.90 inter-operator and 0.95 intra-operator). Each participant was given a code to avoid using their names.

Ethical considerations

The thesis project for the MSc in Dental Sciences, with a focus on Craneal-Mandibular Function and Orofacial Pain was approved by the Research Ethics Committee of the School of Dentistry of Universidad de la República Oriental del Uruguay, file number 206/15. Each individual involved in the study or their legal representatives were requested to give their authorization to take part in the survey by signing an informed consent.

Statistical analysis

A descriptive analysis was conducted to detect the presence of current TMD signs and symptoms using statistical tables of absolute and relative frequency. The association between qualitative variables was analyzed by preparing contingency tables and applying the chi-square distribution test, with a 0.5 significance level. The potential association between response variables was assessed: current TMD signs and symptoms with explanatory variables such as head, neck or jaw trauma, self-perceived stress, regular consumption of mate, coffee, or tea, alcohol and cigarettes. Each binary variable was recorded as present or absent.

Results

The sample showed characteristics that are typical of the drug-using population, as compared to another study published in 2009 which focused on the same care center[20]. The age range under study was 15 to 35 years of age (80% males, 18.5% females, 1.5% transgender), the individuals were likely to have a criminal record (50.4%), to be victims or perpetrators of gender, sexual, or domestic violence (25.9%), to engage in prostitution with the risk of contracting or transmitting STDs, to have teenage pregnancies, affecting the health of both mother and child, to live on the streets (19.3%) or with few belongings, to experience social exclusion, to have dropped out of formal education at an early age (93.3%), to be recently unemployed (76.3%), to have attempted suicide (22.2%), to be aggressive towards themselves and others, to cause traffic, domestic, and workplace accidents, to neglect their own health and their general dental-oral hygiene.

Poly-drug use was frequent in the population under study. They used predominantly legal drugs and some illegal ones, as well as psychiatric drugs without medical prescription, and other substances in a smaller proportion (see Table 1).

Table 1 - Description of consumption variables

	frequency	%		frequency	%
Alcohol			Cocaine base paste		
Yes	111	82.2%	Yes	23	17.0%
No	24	17.8%	No	112	83.0%
Tobacco			Heroin		
Yes	119	88.1%	Yes	1	0.7%
No	16	11.9%	No	134	99.3%
Marihuana			Solvents		
Yes	114	84.4%	Yes	30	22.2%
No	21	15.6%	No	105	77.8%
Cocaine			Hallucinogenic mushrooms		
Yes	100	74.1%	Yes	12	8.9%
No	35	25.9%	No	123	91.1%
Cocaine paste			Floripon		
Yes	109	80.7%	Yes	7	5.2%
No	26	19.3%	No	128	94.8%
Crack			Psychiatric drugs		
Yes	7	5.2%	Yes	37	27.4%
No	128	94.8%	No	98	72.6%

The prevalence of current signs of TMD and the presence of one or more signs are described in Table 2. The prevalence of current symptoms of TMD and the presence of one or more symptoms are described in Table 3.

Table 2- Prevalence of current signs of TMD

Frequency		%
Mouth opening lower than 40 mm		
Yes	19	14.1%
No	116	85.9%
Pain on muscle palpation		
Yes	19	14.1%
No	116	85.9%
Pain on TMJ palpation		
Yes	18	13.3%
No	117	86.7
One or more signs		
None	95	70.4%
One	26	19.3%
Two	12	8.9%
three	2	1.5%

Table 3- Prevalence of current symptoms of TMD

Frequency		%
Difficulty to open mouth		
Yes	10	7.4%
No	125	92.6%
Jaw blockage		
Yes	9	6.7%
No	126	93.3%
Functional difficulties		
Yes	18	13.3%
No	117	86.7
Joint noises		
Yes	33	24.4%
No	102	75.6%
Headache		
Yes	65	48.1%
No	70	51.9%
One or more symptoms		
None	42	31.1%
One	62	45.9%
Two	20	14.8%
three	11	8.1%

Table 4 summarizes some of the associations studied. There was a significant association between TMD symptoms and self-perceived stress and the consumption of mate and alcohol ($p < 0.05$).

Table 4- Association between the prevalence of TMD symptoms and some factors

	TMD Symptoms		
	No	**Yes**	**p-value**
Trauma			
Yes	24.1%	75.9%	0.14
No	37.7%	62.3%	
Self-perceived stress			
Yes	43.8%	56.2%	0.03
No	24.1%	75.9%	
Consumption of mate			
Yes	22.2%	77.8%	0.03
No	8.9%	91.1%	
Consumption of coffee			
Yes	28.2%	71.8%	0.51
No	35.1%	64.9%	
Consumption of alcohol			
Yes	38.0%	62.0%	0.03
No	19.7%	80.3%	
Smoking cigarettes			
Yes	21.2%	78.8%	0.28
No	46.7%	53.3%	

Discussion

The American Academy of Pediatric Dentistry (AAPD) recognizes substance abuse in teenagers as a significant social, family, and public health issue in the United States[21]. Latin America has seen a higher prevalence of psychoactive substance abuse in recent years, with the corresponding increase in the number of medical consultations related to this behavior[22]. Substance abuse and dependence in Uruguay has seen qualitative and quantitative changes over the last decades, which have

positioned it as a serious public health issue. Problematic use of drugs, acts of violence, crime, the tendency to be involved in risky situations, suicide attempts, eating disorders, pathological gambling, internet addiction, etc., are symptoms of our contemporary society[20]

The pilot study conducted in Portal Amarillo in 2013[23], with the same Sheet as the "Uruguayan Survey for Prevalence of Bruxism and TMD"[7], found a 67.6% prevalence of current TMD symptoms in the population aged 15 to 35 in treatment for drug dependence (difficulty or pain when opening the mouth wide, blockage of the jaw when opening the mouth, functional difficulties, noise in the joints, headache); and a 42.3% prevalence of current signs of TMD (mouth opening less than 40 mm, pain on palpation of the muscles and pain on palpation of the TMJ)[23]. The survey conducted by the same team of researchers with the national sample of Uruguayan population in Montevideo and the rest of the country showed a 61.3% prevalence for current symptoms of TMD and 37.3% for current signs of TMD[7]. These preliminary results showed a higher prevalence of TMD signs and symptoms in dependent population. The results of this study, which used an enlarged sample, confirm this tendency for current symptoms of TMD.

These findings are in line with previous studies that show harmful overall health and oral health consequences for drug-addicted population[24]. Trauma is one of the main reasons why drug users are admitted to hospital emergency rooms[22]. Furthermore, cocaine users have been proven to have a higher incidence of bruxism as a secondary manifestation of the use of the drug[25] which causes pain in the TMJ and the muscles of mastication[26]. It has also been observed that illegal drug use has a negative impact both on hygiene habits and in the prevalence of decay, gingivitis, and periodontitis as compared to the general population[27,28].

Conclusions

At the beginning of the treatment for addiction to legal and illegal drugs on a residential basis at Portal Amarillo there is a higher prevalence of TMD symptoms as compared to the general population. Furthermore, individuals who perceive themselves as being under stress or consume mate or alcohol show a significant increase in the symptoms of TMD. These results suggest the need to incorporate measures to identify, prevent, and treat TMD in this vulnerable population.

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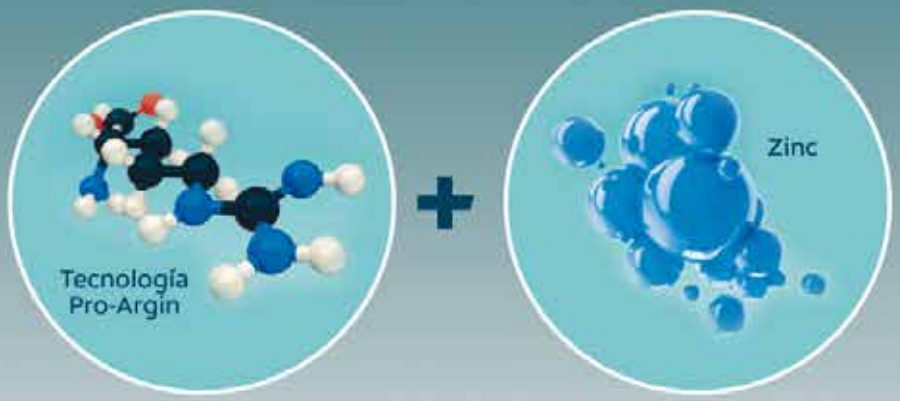

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Multidisciplinary rehabilitation with velopalatal obturator prosthesis: a case report

Carlos M. Cabrera Díaz[1], Angélica Joris Forker[2], Flavia Sartorio Freire[3]

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Abstract

A rehabilitation case is presented where the patient suffered a loss of velopalatal tissue following the surgical resection of a soft palate tumor—adenocarcinoma—performed by the multidisciplinary team of the Oral and Maxillofacial Prosthesis Service at the School of Dentistry, Universidad de la República.

The care protocol includes the prosthetic rehabilitation and the participation of a speech therapist and a psychologist to provide the comprehensive rehabilitation treatment which is essential in these cases.

Different procedures are described, such as clinical stages, laboratory processing, speech therapy records and psychological interviews.

Keywords: velopalatal prosthesis, shutter bulb, rehabilitation, multidisciplinary team.

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Introduction

The rehabilitation of a loss of substance in the tissues of the soft palate poses a very specific challenge for the prosthesist and the speech therapist, for they must use a static prosthesis to restore mobile tissues.

This complex problem can be solved in a satisfactory manner with a multidisciplinary approach; it is thus imperative to work with a team of specialized professionals. The Oral and Maxillofacial Prosthesis Service has worked in this way since its creation under the direction of Dr. Isabel Jankielewicz.

This case presents the complete rehabilitation of an extensive loss of velopalatal tissue as a consequence of the surgical resection of the soft palate to treat a tumor in the salivary gland (polymorphic low-grade adenocarcinoma) in a 61-year-old female patient, a working teacher, who is very self-conscious about her appearance[1].

As the patient is an educator, she is considered to be a professional who works with her voice.

Definition

The main aim of the shutter prosthesis (from the Latin verb *obturare*, shutting, plugging) is to close the existing connection between the oral cavity and the nasal cavity.

Furthermore, the procedure seeks to restore the oral functions which were lost or altered by the lack of palatal tissue, both hard and soft. Said functions include: eating, speech quality, quality of the voice, breathing and hearing, all of which are detailed below.

The velopalatal prosthesis, a variant of the shutter prosthesis, is an anatomical and functional restoration using man-made, alloplastic, inert media, of the loss of tissue involving the soft palate area (velopalatal defects), which may have a congenital or acquired etiology, the latter including trauma and oncological surgery.

Background

Michel Benoist describes the work of Ambroise Paré (1517-1590), the first author to describe a palatal shutter, as well as that of Pierre Fauchard (1678-1761), who developed an ingenious shutter design, which was placed in the palatal defects, opening and closing with a butterfly-style device made of silver and gold.

Around 1820, Goodyear vulcanizes rubber, and vulcanite rubber is introduced as a dental material and used to make this kind of shutters. At the same time, there is growing concern over how to combine the prosthesis with speech therapy.

Benoist also notes the work of Suersen (1877), who studied these aspects in depth, describing in his work not only the velopalatal restoration but also the functional nasopharyngeal obturation; as well as that of Claude Martin (1889), who designed velopharyngeal shutters with a double water pocket, a higher and a lower pocket which allow water to circulate from one to the other, accompanying the movement of the soft palate. The appearance of acrylic resin in 1950 made it the material of choice for dental prostheses[2].

Regarding the design of velopalatal shutters, Ponroy and Psaume's articulated shutter has a hinge that articulates the hard palate and the soft palate parts of the prosthesis. The Schiltsky shutter uses a metal guide between the base and the shutter, and the Froschel and Schalit shutter is projected behind and on top of the upper airways (meato-shutter)[3].

Classification

According to Alfredo Álvarez Rivero et al. (CIMEQ-CUBA) velopharyngeal shutters can be classified into two groups:

a- Soft palate shutter, velopharyngeal shutter or pharyngeal shutter: designed to follow the curve of the palate, drawing near it and seeking contact to try to shut the velopharyngeal sphyncter.

b- Meatus shutter, differs from the above for it is projected backwards and up, partially

shutting the air intake through the nasal cavities (whether perforated or not) improving the voice and preventing the reflux of solid food and liquids[4].

Despite the advances in reconstructive surgery, in the cases of extensive loss of tissue in the soft palate, it is not always possible to use the patient's own tissue for the reconstruction. It is therefore necessary to proceed to an anatomical and functional restoration using man-made inert media, alloplastics, prostheses, supplementing surgery and prosthetics, which applies especially to oncological patients.

Functional alterations

One of the consequences of the loss of velopalatal tissue is that, as the mouth, nose, and sinuses communicate, it hinders all the functions of the mouth.

- Eating: mainly due to the alteration of the oral and pharyngeal swallowing times[5]. The patient must make remedial movements in the mastication process and when containing the alimentary bolus. In other cases it is necessary to change both the consistency and the viscosity of the food, and the way in which the food is ingested (using a nasogastric tube or a gastrostomy tube in more severe cases).
- Speech: it is affected because the patient loses the intraoral points of reference necessary to utter the different phonemes that make up the words. Points of articulation where the tongue makes contact with the palate are lost or altered. This makes the articulation of words blurry, which may lead to unintelligible speech[6].
- Quality of the voice: (resonance, intensity, duration): the oral and nasal cavities are part of the vocal resonation system, together with the larynx, the pharynx, and the paranasal sinuses. The connection between the oral and nasal cavities gives the voice a hypernasal resonance, also known as rhinolalia aperta. The air and the voice come out through the nasal cavity because the cavity cannot be fully closed, and there is a reduction in intraoral pressure due to the lack of palatal or soft palate tissue[7]. The intensity of the voice is low and the sounds produced are short.
- Breathing: due to the nasal air escape, the patient must inhale more frequently, which modifies the breathing rhythm, so the patient gets more tired when speaking.
- Hearing: it is indirectly compromised, since the lowered intraoral pressure does not allow for proper ventilation of the middle ear. After a resection of the soft palate, hearing declines severely because the internal and external stylopharyngeus muscles of the soft palate have been sectioned and removed during surgery. These muscles open and close the pharyngotympanic tube, which balances the air pressure between the middle ear and the outside. This creates mild to moderate conductive hearing loss. Some patients who undergo radiotherapy or chemotherapy lose their sense of hearing as a consequence.

These functional alterations also have psychological, social, family, and workrelated impacts on the patient. (Chart 1)

Functional alterations	Consequences
Eating	Psychological
Speech	Social
Quality of the voice	Family
Breathing	Work
Hearing	

Chart 1: Functional alterations and consequences of the loss of velopalatal tissue.

For the velopalatal prosthesis to achieve its aims, it must relate well with the remaining velopalatal or oropharyngeal tissues. The velopalatal defect is not static, on the contrary, the velopalatal region is largely mobile, and it is a juncture between the digestive tract and the respiratory system.

It is therefore essential to record the movements of the surrounding tissues, which are caused by

the different functions. This requires special impression techniques which allow the patient to reproduce said movements[8].

The rehabilitation of patients who receive this kind of prostheses must be conducted by a multidisciplinary team made up of an oral and maxillofacial prosthesist, a speech therapist, a psychologist, a dental lab technician, and a dental assistant[9,10].

Materials and methods

- Impression and primary model: A primary, anatomical impression is made using a stock tray for dentulous patients, a distal extension of the same is customized with pink wax in order to include the area where the velopalatal tissue was lost. Alginate is used for the impression, being careful to place a piece of gauze on the surface, of the same size as the velopalatal defect. The main aim of the gauze is to prevent the alginate from going up the upper airways (Fig. 1 and 2).

Fig 1: Velopalatal defect. Surgical resection of oncological etiology.

Fig 2: Impression in irreversible hydrocolloid. Note the distal velopalatal extension.

- **Survey:** once the primary model has been obtained, a parallelometer is used to survey the teeth and the mucous areas, including the equator of the velopalatal defect. It is essential to survey the mucous areas and the area of the defect, for the size of the input and output shaft for the prosthesis and its corresponding shutter is determined by the hard dental structures, keeping the length of the base and sealing the communication following this axis. The axis for the introduction and removal of the future prosthesis is determined, and the carving of support beds, plans and guiding surfaces for the abutments is planned, all of which will be done in the following clinical session. We decided to make a plaque prosthesis with 0.9 mm stainless steel carved retainers[11].
- **Individual tray:** a unique individual tray is made for the dental and mucous areas; the pyriform papilla and the median raphe are relieved for biological reasons, using pink wax. An individual acrylic tray will be made later, which is loose on the teeth and tight on the mucosa.
- **Preparation of the mouth:** the planned cutting is done with round and cylindrical diamond burs mounted on a handpiece.
- **Impression and final model:** the final impression is made with silicone elastomer, and the final survey is made with the final model, for a plaque prosthesis should follow the same guidelines as a cobalt-chromium structured one.
- **Preparation of the final model:** The retention areas are determined, with a 0.75 mm retentive undercutting to place 0.9 mm stainless steel active arms on pillars 17 and 24.

In plaque prostheses, the reciprocation of the appliance is set on the guiding surface of the lingual sides of the teeth, the guiding planes contributing in a single entry and exit axis. All the retention areas of the teeth as well as the ar-

eas which protect the gingival margin must be blocked below the guiding surfaces and planes with plaster of Paris or overlay[12]. This is an essential step, for these areas should not be filled with an acrylic base; this would hinder the installation of the appliance and its retouching at the clinic, thus losing the corresponding reciprocation. It should be noted that the anatomy of the loss of tissue must always be analyzed in the survey in order to find the equator of the defect, and to make the shutter enter the area with the necessary retentive slope, without injuring the patient when introducing or withdrawing it.

- **Final base:** the final base technique is applied. The base is made from transparent acrylic based on the model obtained. This provides a stable base for the prosthesis as well as for the tertiary impressions (functional modeling of the velopalatal area).

An omega-shaped 0.9 mm wire loop is made and projected from the distal end of the base of the prosthesis towards the velopalatal area. It will be used for the functional impression of the remaining tissue in the posterior oral sphincter. This is done by the prosthesist together with the speech therapist (see a detailed explanation of the procedure below).

The final base is processed in transparent heat-curing acrylic by means of a muffling process.

Transparent or colorless heat-curing acrylic is chosen because it makes it easy to see the supporting points in the remaining structures, as well as the potential compression areas (fig. 3 and 4).

Fig 3: Model and delimitation of the base.

Fig 4: Processing of the base of the prosthesis. Omega loop made of 0.9 mm wire.

A dynamic or functional impression (tertiary impression) is made by requesting the patient to utter certain phonemes, pronounce words or phrases as well as to make swallowing and cervical movements in order to reproduce the missing material following a tumor excision.

The processed prosthetic base is placed in the mouth and the space remaining between the wire omega and the surrounding tissues of the oropharynx are visually assessed. The patient is previously trained by requesting her to move her head to the side and forward towards the chest, to utter phonemes, preferably those which are articulated at the back in isolated syllable combinations, forming words, and associated with simple phrases, together with saliva swallowing movements.

Sealing wax is placed on top of the wire omega, thus creating a first approximate shape of the shutter bulb (figures 5 and 6). The next step is to add a regular consistency silicone elastomer. The aim of this particular technique is to record the stylopharyngeus muscles and the superior pharyngeal constrictor muscle (Passavant's ridge).

Fig 5: Processing of the base of the prosthesis. Omega loop made of 0.9 mm wire. (a different angle)

Fig: 6. Functional record of the velopharyngeal muscles with modeling wax.

The palate and the tongue are closely linked from the womb; it is thus not difficult to understand the reciprocity between the shape and the function of the oral cavity. Furthermore, as the child acquires language, she acquires the different phonemes in a sort of vocal play, where the tongue is in contact with the other oral components. This is how phonetic engrams and oropharyngeal proprioceptivities are formed and recorded in the brain, together with auditory perception.

Losses or alterations in the mouth after tumor excision alter the sensitivity, the intraoral proprioceptivities, the speech articulation points, the placement of the tongue to swallow, and even the saliva.

The alterations caused by this kind of surgery are manifold: the quality of the voice is altered with a nasal resonance (hypernasal resonance), as is the intensity of the voice (hypophonia), breathing is more frequent, the utterance of language tends to be blurred but not necessarily unintelligible, swallowing and hearing are also altered.

This is why it is so important to act early, resorting to the patient's neuromuscular and auditory memory, providing her with a structure that is as similar as possible to what she had prior to the surgery, allowing for the new intraoral propioception, and facilitating the compensatory maneuvers which occur naturally in the patient.

The selection of phonemes and combinations of phonemes in syllables and phrases for the patient to repeat, and the oral movements that increase intraoral pressure are selected based on the location and the size of the tumor excision and the patient's possibilities[13].

The phonemes used in this case were posterior articulated phonemes /k/, /g/, /x/, combined with the repetition of the vowel sounds /e/, /i/. The transitional phonemes chosen, articulated from the middle to the posterior area of the palate were /tʃ/ and /ɲ/ combined again with the above vowels.

Considering that speech is part of the dynamic communication process, she was asked to repeat words with the phonemes mentioned above.

The functional record is made by the dentist in the presence of the speech therapist. The patient is sitting upright, regular silicone is placed on the wire loop, and with the prosthesis in place, she is asked to utter phonemes, words, and phrases during the active period (vulcanization) of the silicone elastomer (figs. 7 and 8). The different combinations of phonemes are uttered during the 3:30 minutes it takes the elastomer to be vulcanized, in order to record the different groups of muscles (lingual and pharyngeal).

The patient must be sitting on the dental chair at a 90 degree angle for the dynamic impression, for the normal position we are in when we speak is standing or sitting at the axis of gravity.

Fig. 7. Final functional record with silicone elastomer

Fig. 8. Final functional record with silicone elastomer (another angle)

The velopalatal section of the prosthesis or the shutter bulb must have the following features:
a) In the silent resting position of the oropharyngeal muscles (while the person breathes) the prosthesis must be separated from the muscles, allowing the air to pass towards the pharynx when breathing through the nose.
b) The oropharyngeal muscles are contracted both when eating and speaking. For these movements, the shutter must touch said structures softly in order not to damage the mucous tissues.
The velopalatal extension of the prosthesis, made based on the records created in the clinic, is created with a mold and a plaster countermold which are connected by means of guides, which reproduce both the nasal side and the mouth side, respectively, and between both peripheral edges of the shutter bulb. This molding chamber is loaded and processed with transparent heat-cured acrylic. The prosthesis must be carefully finished so as not to damage the remaining tissues involved (fig. 9), both on the oral and the nasal side of the shutter bulb (fig. 10, 11 and 12).

Fig. 9. Processed prosthesis

Fig. 10. Processed prosthesis (mouth side).

Fig. 11. Processed prosthesis, mouth side (another angle)

Fig. 12. Processed prosthesis, nasal side (another angle)

The prosthesis is finally installed, with the relevant prosthetic and speech therapy controls[14].
The rehabilitation will not be comprehensive if it is not approached from the psychological perspective as well, in which sense, rehabilitation shall be understood as "re-enabling the person, restoring someone back to their previous state", making reference to the healthcare process aimed at caring for the consequences of an illness or trauma which cause dysfunction or disability, to restore the person back to their social, working, and comprehensive functions.
The psychologist is in charge of seeing the patient through the process of mourning for the body part they lost. This loss or mutilation, whose impact depends on the patient, is also

discussed in terms of their fantasies and expectations[15].

The psychologist accompanies the elaborate process of coming to terms with the loss. This is not just any loss: the patient has lost a part of herself, which is experienced as an anticipation of death or a loss of integrity.

Fig.13: Installed prosthesis

There are five stages or mourning:

Denial stage: The person protects herself from what has happened, and uses denial as a defense mechanism.

Anger stage: She cannot avoid what happened and this creates discontent. She expresses her anger towards her surroundings or her own self. The fantasies associated to the loss and how the person experiences them will define where she projects her anger. She will look for reasons or people to blame.

Bargaining stage: She tries to negotiate with her environment or with herself, assessing the pros and cons of what happened.

Depression stage: She experiences grief.

Acceptance stage: At this stage, the person accepts that the loss is inevitable[16].

A good and healthy mourning process allows the patient to accept the replacement, the prosthesis in this case, as part of her new body image. Raich defines body image as a complex construct which involves the whole body and its parts, as well as the perception of movement and the subjective perception of feelings, thoughts, and appreciations, as well as the way in which the individual behaves in relation to her cognitions and feelings[17].

Multidisciplinary team
Oral and Maxillofacial Prosthesist
Speech therapist
Psychologist
Dental Lab Technician
Dental Assistant

Chart 2: Members of the multidisciplinary team

The members of the Oral and Maxillofacial Prosthesis Service team involved in the resolution of this particular case followed a care protocol that was put together over time in the team's monthly meetings (R. Soler, oral communication).

This protocol establishes the role played by each one of the professionals, as well as their interaction with the others, at the different stages of the treatment (Chart 3).

The main features of the protocol are as follows:

- The treatment plan is put together based on the prosthetic, speech therapy, and psychological assessment.
- Essential interaction between the prosthesist and the specialized dental lab technician.
- Joint clinical work of the prosthesist and the speech therapist.
- Ongoing pshychological assessment.
- Joint discharge.

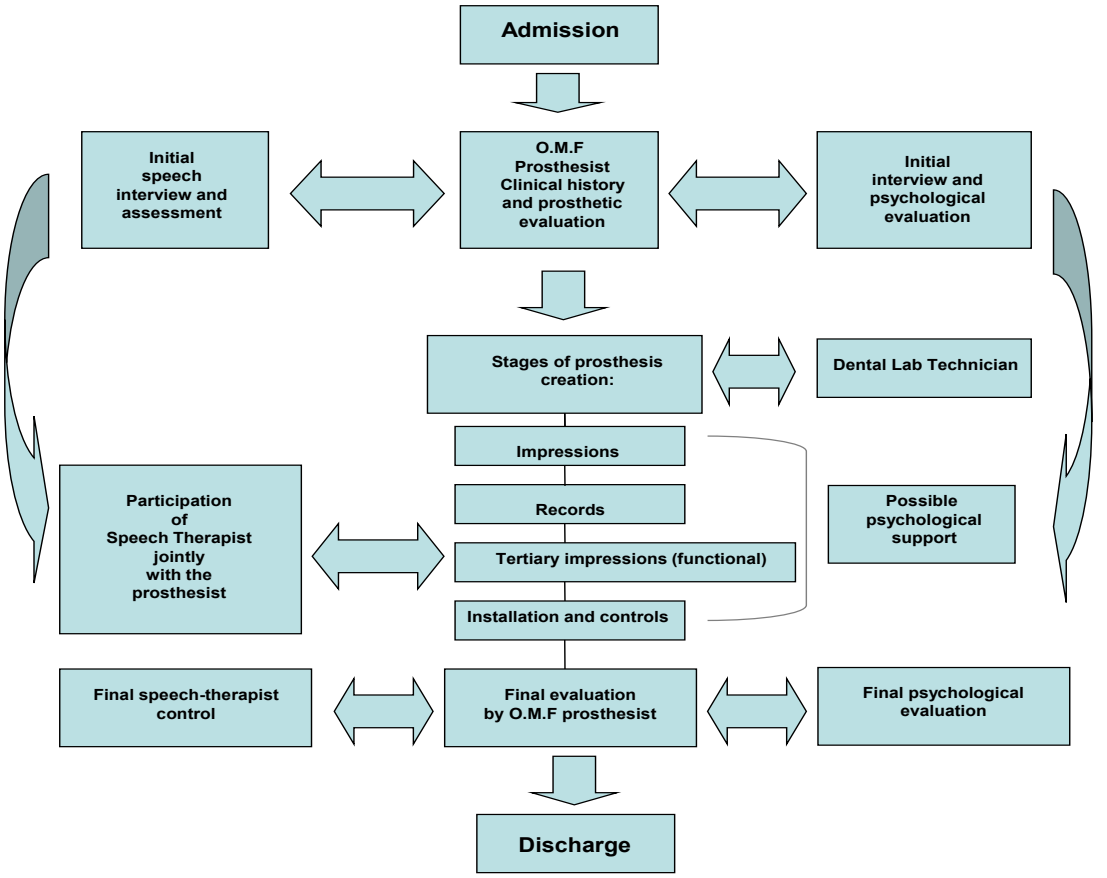

Chart 3: Oral and Maxillofacial Prosthesis Service Care Protocol

The patient receives a satisfaction survey form to assess the care provided. The form collects the patient's opinion and that of her friends and family; it is given to the patient at the moment of installation, and she is expected to return it to the team in the subsequent check-up sessions during the adaptation period.

Discussion

The technique applied emphasizes the importance of the functional impression of the remaining tissues of the soft palate with the participation of the speech therapist, thus obtaining a shutter bulb which adapts to the many ways in which these tissues change according to the different functions they perform.

The patient starts working with the speech therapist prior to the surgery. The professional in charge records the patient's voice and assesses the different functions of the mouth to compare pre-surgery records to post-surgery records.

Once the prosthesis has been made and installed, it will be possible to assess its functional gain by using the speech therapy file to compare the quality of the voice achieved, eating, mastication, speech intelligibility, intraoral pressure (kissing, blowing, whistling, clicking, sucking). One of the main concerns of the patient, who is a working teacher, was the quality of the voice she would have to use in front of her students[18]. With this in mind, the team tried to improve the hypernasal resonance, the articulation of words, and to prevent the reflux of food and liquids to the rhinopharynx.

From the prosthetic point of view, the distal extension provided by the velopalatal prosthesis makes it necessary to consider its mechanics. The extension creates additional weight, apart from an inevitable lever arm. For this purpose, the distal section of the prosthetic base must be as light as possible, while providing the highest possible retention.

The height of the palate must follow the curvature of the palatal vault (the tongue has the same mobility); a low palate may trigger the gag reflex and a feeling of a full mouth due to insufficient intraoral space. An average person swallows about 70 times per hour during the day, and produces 1 to 2 liters of saliva per day, which must be swallowed; this must be taken into account for the tertiary impression (dynamic or functional impression).

Finally, all the clinical procedures are carried out with the support of a psychologist, who is one of the first professionals to establish contact with the patient and the case, and is the one that discharges the patient.

Conclusions

Working with a multidisciplinary team helps the patient recover from the aesthetic and functional point of view, as well as in a more comprehensive manner (from the psychological and social perspective), offering her the possibility of going back to work and reactivating family, social, and work relations.

The multidisciplinary team must be in touch with the patient before the surgery, plan a solution for the case with the surgical team and prepare the pre-surgical prosthetic and speech therapy records.

The work of the psychologist is essential not only for the patient but also for the prosthesist, as it will allow him to treat the patient in a more personalized manner.

The prosthesis is designed jointly by the dentist and the speech therapist, and it is assessed by both at the different clinical stages; the patient should be ready to be discharged once she has undergone all the dental, speech therapy, and psychological assessments.

Within this framework, the patient can receive support during the treatment, making it easier for her to express her fears and fantasies, thus allowing her to better accept what happened, taking the person as a bio-psycho-social being.

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Skeletonized body identified by analysis of frontal sinus morphology and characteristics of osteosynthesis material: a forensic case report

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Abstract

Forensic dentistry is essential for the identification of highly decomposed and charred bodies, as well as skeletal remains. This study reports a case of human identification by analyzing the morphology of the frontal sinuses and osteosynthesis material. In the anthropological assessment of skeletal remains a surgical plate used for osteosynthesis was detected in the periorbital regions. Relatives of the potential victim provided ante-mortem (AM) radiographs which revealed the presence of an osteosynthesis plate. Post-mortem (PM) imaging exams were performed to reproduce the AM data. Similarities were observed between the AM and PM radiographs, especially regarding the morphology of the frontal sinuses and the position and outline of the surgical plate used for osteosynthesis. The comparison of AM and PM images made it possible to identify the victim and to aid the criminal investigation. It also highlighted the role of radiographs and anatomical characteristics in the process of human identification.

Keywords: forensic dentistry, forensic anthropology, frontal sinus, radiography.

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Introduction

With the rise of urban violence, traffic accidents and crime, the number of charred, mutilated or highly decomposed bodies and skeletal remains that reach forensic medicine departments around the world has increased significantly[1,2]. The cause of death and the identity of the victim could be difficult to determine in such conditions, as when soft tissues are destroyed it is not possible to conduct a fingerprint analysis[1,3]. This is why forensic anthropology and forensic dentistry play a major role in criminal investigations, where the relatives of the missing person will be interviewed and asked to provide medical and dental records, such as clinical records, radiographs, CT scans, cast models and pictures to aid in the human identification process[1,3-6].

Among the many methods available it is worth mentioning those comparing AM and PM radiographic imaging of the skull[4], as the morphological variability of its many anatomical structures, like the frontal sinus, is very useful in forensics to determine a person's identity[7].

The specialized literature presents cases, although few, where human remains were identified by analyzing the frontal sinus[7,8], comparing images of the anatomical characteristics of the sinus and those of the osteosynthesis material[9]. The aim of this study is to present a forensic case report of a skeletonized body identified by analysis of frontal sinus morphology and the type and location of the material used to reduce trauma-induced fractures.

Case description

A skeletonized body was found in a forested region in the State of Goiás, Central-West region of Brazil. The forensic anthropological analysis (biological profiling) and cause of death determination were conducted after the examination at the scene had taken place and the remains were transported to the Institute of Forensic Medicine (IML) in the state capital.

During the external examination of the body we observed the pelvis and skull had male characteristics[10], closed medial clavicular epiphysis[11], no osteophytes in the vertebral bodies[12], sagittal suture with few points of synostosis and third molars with full rhizogenesis, so we determined the age to be between 30 and 40. We estimated the height (between 1.67 m and 1.74 m) by measuring the length of the long bones of the upper and lower limbs and by using Trotter & Gleser's[13] 1952 table. There were three osteosynthesis metal plates on the skull, fixed with screws, which suggests the occurrence of AM trauma (characteristics and location shown in Table 1 and Figure 1).

The cause of death was determined to be traumatic brain and face injury by means of a per-

Table 1 - Characteristics of the osteosynthesis material found in the skull

Plate:	Anatomical Region	Shape of the plate	Number of fixing screws	Presence of bone on the plate
1	Lateral wall of the right orbital ridge	Linear	4	No
2	Body of the right malar bone	Linear	4	Yes
3	Left infraorbital rim	Linear	3	Yes

Fig. 1– right-hand side (A) and front (B) of the skull, with male anthropological features and plates, fixed with screws, in the periorbital region, bilaterally.

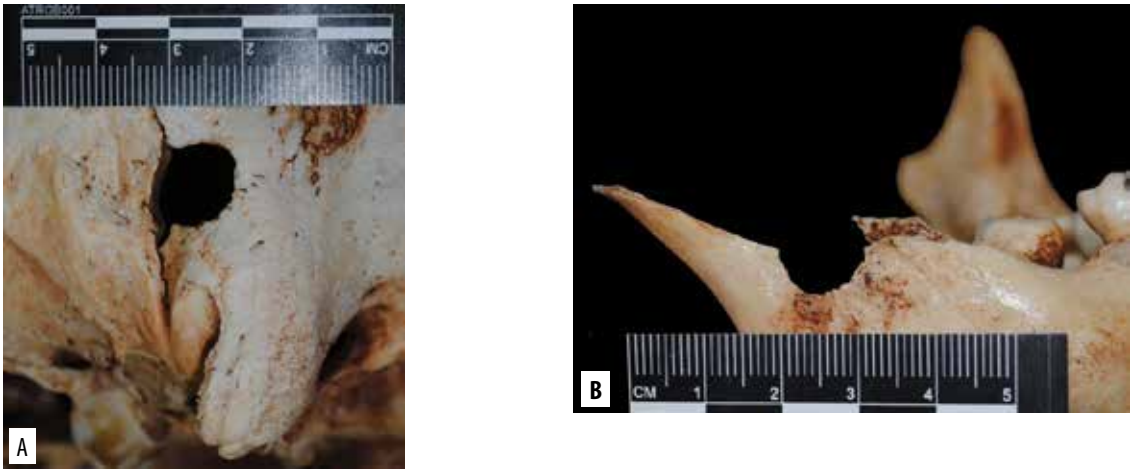

Fig. 2 - Entry gunshot holes on the right-hand side of the occipital region (A) and right ramus (B), fractured.

forating, blunt instrument. There were transfixing wounds caused by firearms, with entry holes in the occipital and right jaw regions (Figure 2). In addition to expert examinations, police investigations revealed that the victim was probably a 29-year old man who had been kidnapped seven months earlier. The AM documentation submitted entailed a medical form and two posteroanterior (PA) skull radiographs (one presurgical and one immediately after surgery), taken seven years before his disappearance, which showed a fracture and an osteosynthesis plate in the right periorbital region. These also showed the morphology of the frontal sinus: expanded to both sides of the midline (Figure 3).

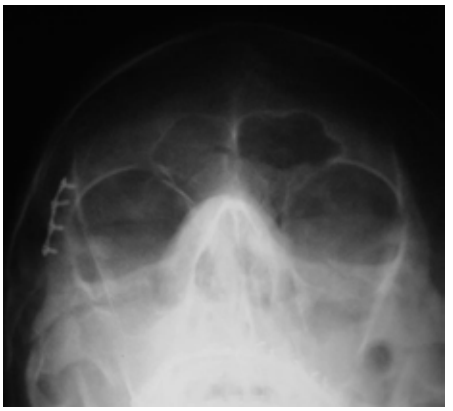

Fig. 3 - AM postsurgical PA radiograph of the skull, taken immediately after the first osteosynthesis plate was fitted.

We decided to conduct PM exams, a PA radiograph and a cranial cone-beam CT (Figure 4). This was done at a radiological laboratory that has signed an agreement with the IML. InVesalius® software was used to review the PM CT. The cranial image was reconstructed, rotated and processed in the same position as the AM immediate postsurgical PA radiograph.

After that, we compared the AM immediate postsurgical PA radiograph to the PA radiograph and PM CT. We found morphological similarity of the frontal sinus in terms of its area of expansion, number and location of the septa and lobes (Table 2) and the osteosynthesis plate present in the lateral wall of the right orbit, anatomic location, type and fixing screws (number, design, length, arrangement and fixing angle), as shown in Figure 5. This made it possible to positively identify the missing person.

Table 2. Comparison of frontal sinus characteristics in AM and PM radiographs.

Feature	AM radiograph	PM radiograph	Similarity
Bilateral Expansion	Yes	Yes	Yes
Greater expansion area	Left-hand side	Left-hand side	Yes
Number of lobes on the right-hand side	2	2	Yes
Number of lobes on the left-hand side	3	3	Yes
Presence of median septum	Yes	Yes	Yes
Presence of intermediate septa	No	No	Yes

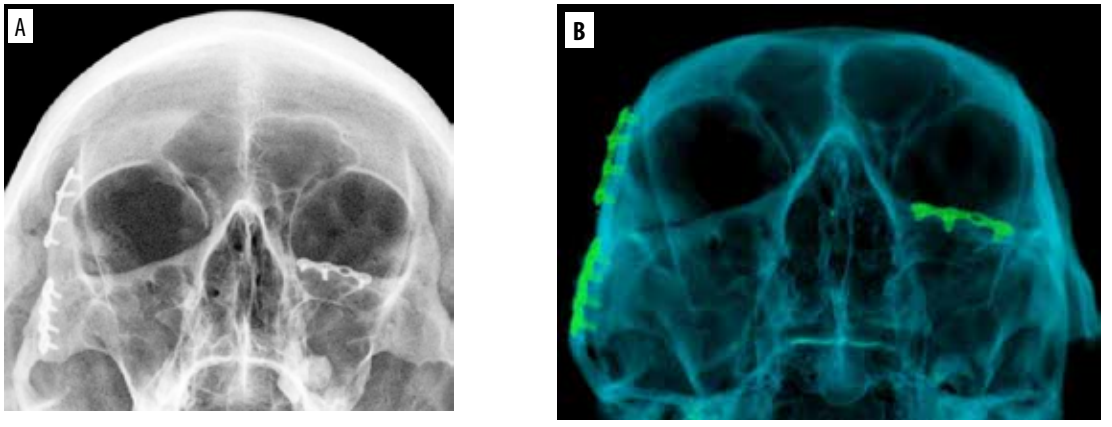

Fig. 4 - PA radiograph (A) and 3D CT reconstruction (B) of the PM skull.

Fig. 5 - Comparison of AM immediate postsurgical PA radiograph (A) and PA radiograph (B) and PM CT (C)

Discussion

Radiograph images are widely used for identification purposes, especially when a lophoscopy analysis cannot be performed, and are very useful in cases of charred, decaying and skeletonized bodies[4,5,9,14].

This is further enhanced by the number of skull x-rays performed on a daily basis, as the population has increased access to dental care, to diagnose, plan and/or check different clinical treatments[5,15].

In addition, the skull is highly resistant to environmental conditions and thus has demonstrated great forensic potential. In fact, PA radiographs make it possible to study the anatomical features (dental or bone), pathological and/or therapeutic characteristics, as well as the type of a possible osteosynthesis material[9].

However, we should remember that PA radiographs are not the best alternative for a detailed dental analysis, if compared to panoramic[5] or intraoral[1] radiographs. These should be used with caution to avoid misleading or inconsistent conclusions. In return, they become valuable in a morphologic, qualitative and quantitative assessment of the frontal sinus, bearing in mind that this expansive cavity is unique and does not change in adults[7-9,16,17].

In this case, the morphological similarity between AM and PM x-ray images of the frontal sinus, comparative procedure widely supported and described in the literature[7-9,16.17], made the positive identification of the victim possible, paving the way for the criminal investigation to arrest and prosecute those involved in his death. The presence of the osteosynthesis plate on the lateral wall of the right orbit was an additional element for comparison, with results consistent with those of the frontal sinus[9].

Finally, it should be mentioned that this method is faster and less expensive than a DNA test, showing how important it is to have a radiology service in the IMLs or, if this were not possible, to enter into agreements with public or private radiological laboratories. It is also necessary to have forensic dentists with the right expertise and training in imaging techniques, especially extraoral x-rays and CTs, usually used for criminal investigation purposes.

Conclusions

PA radiographs and skull CTs make it possible to analyze the morphology of the frontal sinus and the characteristics of a potential osteosynthesis material, facilitating the human identification process in complex cases of charred, decaying or skeletonized bodies.

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Case report of Basal cell adenocarcinoma of the parotid gland: clinicopathological and immunohistochemical study

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Abstract

Basal cell adenocarcinoma is an epithelial neoplasm with the cytological characteristics of basal cell adenoma but with a morphological pattern of infiltrative growth indicative of malignancy. Due to its low incidence it is often difficult to diagnose a basal cell adenocarcinoma. The objective of the present study was to identify morphological and immunohistochemical characteristics that contribute to its diagnosis. A parotid tumor was resected in a 52-year-old patient; postoperative biopsy and immunostaining with Ki-67, CK19, p63 and alpha- smooth muscle actin were performed. It was diagnosed basal cell adenocarcinoma that invades the tumor capsule, periglandular fat and lymph nodes. Immunostaining with Ki-67, CK19, p63 and alpha- smooth muscle actin was positive. Subsequently, a maxillary sinus metastasis was diagnosed. The morphological characteristics, Ki-67 expression strongly positive and metastasis give the malignant character to this tumor, which differentiates it from the basal cell adenoma.

Keywords: parotid, basal cell adenocarcinoma, diagnosis.

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Introduction

The World Health Organization as well as Ellis and Auclair described basal cell adenocarcinoma (ICD-O Code 8147/3, International Classification of Diseases for Oncology) as a cytological epithelial neoplasm, and in terms of histomorphology it was found to be very similar to basal cell adenoma, but with infiltrative growth and a low incidence of metastasis[1-3].

Background

Some authors consider that this tumor stems from a basal cell adenoma while others argue that it originates de novo. Of all the cases, 80% are located in the parotid glands, 9% in the submandibular gland and 11% in minor salivary glands. It is a rare tumor, which is more frequent in adults between their 4th and 9th decade of life; it is very rare in children. Swelling is usually the only symptom but sometimes it also causes pain and occasionally remains undiagnosed for ten years. It has been categorized into four types based on its histological pattern of growth: solid, membranous, trabecular and tubular[4-6]. Given its low incidence and scarce bibliographic information, it is often difficult to diagnose it[7].

The aim of this study was to present a case of basal cell adenocarcinoma in the parotid deep lobe in a 52-year-old woman and to identify morphological and immunohistochemical characteristics that contribute to its diagnosis.

This work is part of the project called "The extracellular matrix and the myoepithelial component of epithelial tumors of human salivary glands: structural, histochemical, immunohistochemical and lectinhistochemical study", authorized by the Ethics Committee of the National Clinical Hospital. School of Medical Sciences. Universidad Nacional de Córdoba. Argentina (File 188/14).

Clinical case

In November 2002, a 52-year-old female patient visited the dentist and an otorhinolaryngology service since she felt pain in the left region of the mandible; a diagnosis was not reached. In December 2002 she went to a new appointment and a nodular mass was found in the left ascending ramus of the mandible, with increased pain on palpation. She was diagnosed with trigeminal neuralgia, ruling out the importance of the nodular lesion; physiotherapy was indicated for the cervical region. However, the pain continued to increase, so in December 2003 she underwent a biopsy puncture of an adenopathy, where an inflammatory process was identified.

In January 2004, a medical interconsultation was made with a head and neck surgeon who requested a CT scan of the neck which showed an image compatible with a tumor lesion. In February of the same year, a left parotid deep lobe tumor was excised with good postoperative evolution.

Anatomic pathology report: The macroscopy showed a slightly lobed, encapsulated mass, 5 cm x 1.7 cm x 1.2 cm of violet gray color with firm areas when cut. In the microscopic examination of the colored sections with H/E there was a fibrous capsule that surrounded the basaloid cells with pale eosinophilic cytoplasm and rounded or oval nuclei organized in sheets and nests of variable shapes and sizes, separated by bands of connective stroma. No areas of tumor necrosis, cellular atypia or mitotic figures were found.

The histological growth pattern was solid and tubular. The solid pattern presented nests and strands of contiguous basaloid cells that in the periphery of these structures formed a cellular layer organized in palisade cells.

In the tubular pattern there were basaloid cell islets that contained pseudocysts or prominent lumen lined with cuboid cells (Fig. 1 A and B).

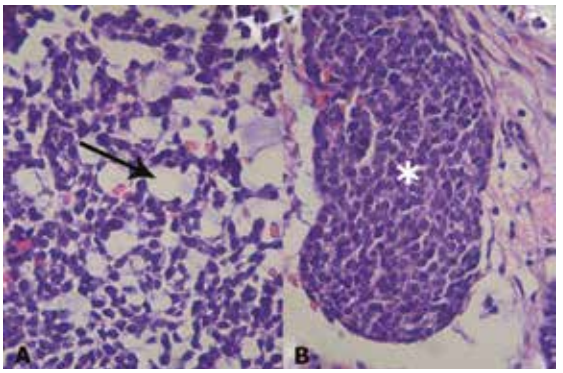

Fig. 1: HISTOLOGICAL PATTERNS OF BASAL CELL ADENOCARCINOMA
Tubular pattern. The arrow indicates the cells that contain lights and pseudocysts lined by cells with dense chromatin nuclei. H/E 400x.
Solid pattern. Nest of basaloid cells (asterisk). H/E 400x.

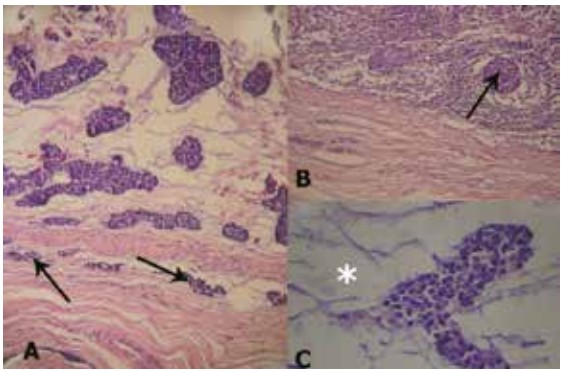

Fig. 2: BASAL CELL ADENOCARCINOMA WITH INFILTRATING GROWTH AND METASTASIS
The arrows show the invasion of the tumoral connective capsule. H/E 100x.
The arrow shows the metastasis in a regional lymphoid node (H/E). 100x.
The asterisk shows the infiltration of periparotid adipose tissue. H/E 400x.

The diagnosis of mixed type basal cell adenocarcinoma was made on the basis of its histological patterns (solid and tubular) and its malignant character, with an aggressive growth with infiltration of the tumor capsule and periparotid fat, perineural invasion and metastasis of a regional lymphoid node (Fig. 2A, B and C).
We then observed maxillary sinus involvement with bone metastasis.

Immunohistochemical labeling was used to find the differentiation of basaloid cells in epithelial (CK 19) and myoepithelial cells (p63 and α-smooth muscle actin) (Fig. 3A). The proliferating capacity of the tumor was studied with the marker for Ki-67. A positive score was given only if there were over 10% of marked cells[8]: the result was 25% of positive cells. (Fig. 3B).
The results are shown in Table 1.

Table 1: Immunohistochemical analysis of basal cell adenocarcinoma

Marker	Marked cell	Reaction *
Ki- 67 (prognostic marker of cell proliferation)	Epithelial cells (nuclei)	Strong
p63 (myogenic differentiation)	Myoepithelial cells (nuclei)	Strong
Alpha-AML (myogenic differentiation)	Myoepithelial cells (cytoplasm)	Moderate
CK19 (epithelial differentiation)	Epithelial cells (cytoplasm)	Strong

Alpha-AML: α-smooth muscle actin. CK19: cytokeratin 19
*** The qualitative assessment of the labeling was performed according to the intensity of coloration.**

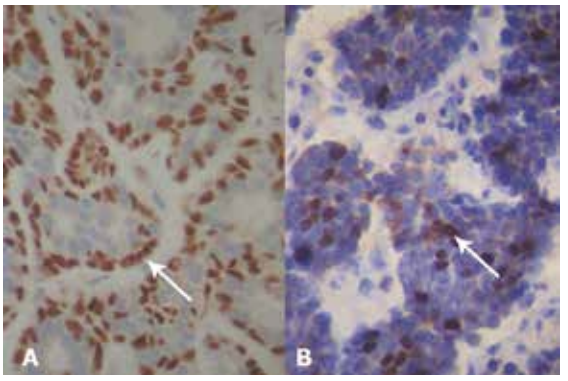

Fig. 3: IMMUNOHISTOCHEMICAL LABELING OF BASAL CELL ADENOCARCINOMA
The arrow shows the nuclei of myoepithelial cells positive for p63. 400x.
The arrow indicates the nuclei of the epithelial cells positively labeled for Ki-67. 400x.

Given the results of the histopathological study, the patient was referred for oncological treatment, with indication of postoperative radiotherapy for six weeks, which began in March 2004. The treatment plan included the irradiation of the parotid plus the cervical region left at a total dose of 60 Gy, with a daily dose of 2 Gy. The complications were mucositis, dysphagia and mycosis, which, upon receiving specific treatment, improved at the end of the radiotherapy sessions.

In September 2004, an orthopantomography showed a cyst in the left horizontal ramus of the mandible and a vegetative image in the homolateral piriform sinus. The surgery was performed and the biopsy was negative for malignancy.

Subsequently, in March 2005, the pain in the hard palate and the left region of the maxilla began. A CAT scan was indicated and a lesion was identified at the maxillary floor level. The surgery was performed and metastasis was diagnosed in the floor of the maxillary sinus. A new radiotherapy plan was indicated, which had mucositis and dysphagia as complications.

The general condition of the patient worsened and she presented dehydration and drowsiness. The radiation therapy scheme could not be completed; palliative care was continued until her death two months later.

Discussion

Primary basaloid tumors of the salivary glands are lesions characterized by a predominance of "basaloid" epithelial cells with round or ovoid nuclei surrounded by a thin cytoplasm. The term "basaloid epithelial cells" arose from observations with standard histological techniques that do not make it possible to distinguish if there are myoepithelial or ductal or basal epithelial cells. In these cases, immunohistochemical labeling is a useful tool to detect both cell types, since it improves diagnostic accuracy. The immunohistochemistry panel used in the case described (p63, α-smooth muscle actin and CK19) was important for the diagnosis of the "basaloid" cells of this carcinoma[9,10].

The heterogeneous tumoral group of basaloid epithelial cells includes benign tumors such as pleomorphic adenoma, and basal cell adenoma, and malignant tumors such as adenoid cystic carcinoma, undifferentiated small cell carcinoma, and basal cell adenocarcinoma[10,11].

Basal cell adenocarcinoma mainly affects the parotid gland (more than 90%) and is histologically similar to basal cell adenoma. However, it presents an infiltrating growth and a low incidence of metastasis[1,2,12].

It is generally considered a carcinoma of low degree of malignancy, locally destructive and with frequent recurrences. The recurrence period varies from six months to two years and only occasionally produces distant metastases[12,13].

In this case the evolution was atypical, presented a bone metastasis in an early form, less than a year after the end of its treatment. This led to the deterioration of the general condition of the patient and her death. This can be explained by the late diagnosis of the lesion, with lymphoid node metastasis; and histological factors of

poor prognosis such as capsular, perineural and periglandular fat invasion.

There is no predilection for gender, and 80% of tumors occur after 50 years of age with an average of 60 years of age[14]. This case corresponds to a 52-year-old woman.

Its histomorphological patterns are described as solid, membranous, tubular or trabecular. The solid pattern is the most frequent, characterized by solid cellular nests of different shapes and sizes. A collagen stroma, consisting of bands of different thickness, separates these nests that correspond to the most frequent histological pattern and with the highest risk of metastasis. In the membranous pattern there are abundant deposits of a strongly eosinophilic hyalinized basal membrane. The growth pattern is tubular when luminal spaces are formed between basaloid epithelial cells, whereas in trabecular growth, basaloid epithelial cells are arranged in interconnected cell bands[15].

In our study, the tumor was located in the parotid gland with a mixed growth pattern since the basaloid epithelial cells were placed in solid and tubular areas.

Regarding the prevalence of this neoplasia in Latin America, in the consulted databases we only found data in two studies by Ruiz-Godoy and Rivera et al. and Ito et al.[16,17].

In 1996 Ruiz-Godoy Rivera et al. conducted a clinicopathological study of six cases of basal cell adenocarcinoma analyzed at the Instituto Nacional de Cancerología, México[16].

Ito et al. examined the archives of the Department of Pathology of the Instituto del Cáncer de Londrina (Paraná State, Brazil) for the years 1972-2001, and analyzed a total of 496 cases of major and minor salivary gland tumors. These researchers found only three cases of basal cell adenocarcinoma[17].

Histologically, the main differential diagnosis is made with basal cell adenoma; but it also includes the solid variant of cystic adenoid carcinoma, basaloid squamous cell carcinoma and cutaneous basal cell carcinoma with deep invasion[12,15].

Wilson and Robins claim that local invasion of the surrounding soft tissues and the gland are the best markers for differentiating the basal cell adenoma from basal cell adenocarcinoma. They also consider it important to complement the recognition of these tumors on the bases of invasion with proliferation markers such as Ki-67 and apoptosis markers[18].

In a previous study we obtained a negative Ki-67 labeling in the basal cell adenoma, unlike the basal cell adenocarcinoma, where we observed a proliferation index greater than 25%, which also explains the unfavorable evolution of this patient[10].

Saluja et al. also claim that the definitive diagnosis of malignancy is established by the invasive nature of the neoplasm in the surrounding tissues[19].

On the other hand, Jung et al. report that basal cell adenoma with capsular invasion can also be considered an infiltrating tumor, which makes us question the category of basal cell adenocarcinoma. These authors say that the tumors do not have morphological differences, frequently develop a cribriform or solid growth pattern and are larger than the basal cell adenoma without capsular invasion. However, they consider that the cribriform structure of basal cell adenocarcinoma may correspond to a misdiagnosis of adenoid cystic carcinoma. In addition, the solid variant of adenoid cystic carcinoma presents cells with marked nuclear atypia and frequent mitosis[7].

Although regional and distant metastases are not common, in this case there was metastasis in a regional lymphoid node and also in the maxillary sinus floor, which led to the patient's death[12].

In general, there is no consensus regarding its treatment, since some recommend local excision and others total parotidectomy, even in cases of membranous basal cell adenoma, as mentioned by Zhan et al.[12].

For the primary treatment of the tumor, when it originates in the minor salivary glands, some authors consider that surgical excision should be wide to ensure its complete removal[20].

In addition, there is no agreement on the role of radiotherapy. However, postoperative radiotherapy is recommended in the case of surgeries with positive surgical margins or surgical excision of recurrent tumors[21,22].

Conclusions

Epithelial and myoepithelial cells coexist in the basal cell adenocarcinoma of the salivary glands, identified through immunohistochemical markers.

It is essential to obtain histological sections of the tumor interface with the normal gland or with the surrounding fat to evaluate tumor invasion.

Capsular, perineural and periparotid adipose tissue invasion, Ki-67 positive immunolabeling with a proliferative index of 25% and metastasis in a regional lymphoid node and in the maxillary sinus are factors of poor prognosis that, associated with late diagnosis, explain the unfavorable evolution of the patient.

Although it is a tumor with local aggressiveness and low incidence of metastasis, early diagnosis and early oncological care are of crucial importance to give these patients a greater chance for a cure.

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Ascher syndrome: Clinical case care and treatment

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Abstract

We present a case report of patient with Ascher Syndrome. It is a rare entity, benig, with few cases related in the world.

Clinically Ascher Syndrome it is an anomaly wich may be either congenital or acquired, composed by three fillings: blepharochalasis, double lip (upper lip is involved more often than the lower, although on occasion both may be involved), and nontoxicthyroid goiter).

Apart from a deformity that interferes whit mastication and speech, surgical treatment may be indicated for cosmetic reasons.

Keywords: Ascher syndrome, double lip.

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Introduction

This work presents a case of Ascher Syndrome (AS), a condition characterized by concurrent double lip, blepharochalasis and nontoxic thyroid enlargement, though the thyroid enlargement is not evident in a large number of cases[1,2,3,4,5,6]. It is a syndrome of particular interest for dentists, because many times the patient wanders through different medical specialties without a positive diagnosis as the low incidence of cases makes identification difficult[1-3].
The occurrence of double lip has been described as a benign, rare or very uncommon congenital or acquired anomaly, without sex or racial predilection, which can sometimes be a manifestation of a specific syndrome[1,3,7,8].
Its etiology is unknown. According to Ascher[5-6], the upper lip is more frequently affected than the lower lip, at a ratio of 10:1. This developmental alteration can often appear alone or as part of a triad of clinical features, known as Ascher Syndrome (AS).

Background

A review of the scientific literature showed that Laffler[1,9] was the first to report a case of double lip associated with blepharochalasis in 1909. In 1920, Ascher[5] completes the clinical picture described by Laffler in 1909 by adding the thyroid hypertrophy. That triad of features is what is known today as Ascher Syndrome (double lip, enlargement of the eyelid skin and nontoxic thyroid enlargement).
Thyroid hypertrophy is not included consistently in most of the articles reviewed among the criteria for a positive diagnosis[1,2,3,4,5,6,7,8,9,10].
On occasion, in AS the extent of the lip and/or eyelid volume alterations is such that it can cause severe aesthetic and functional alterations, for which surgical correction is the only effective therapeutic approach for the area affected[1,10-12].

Clinical case

The patient is a 14-year-old male (Fig. 1) referred by the Clinic of Periodontics of the School of Dentistry to the Department of Oral & Maxillofacial Surgery III, Universidad de la República, because his upper and lower lip were enlarged, and this greatly affected him psychologically in his relationships. The enlargement had become very noticeable four years before. He visited several medical specialists and dentists that could not make a positive diagnosis or suggest a treatment plan.
His medical history was started in 2013, when he was admitted to the Department of Oral & Maxillofacial Surgery II. Psychosomatic development was consistent with his age. The patient has had asthma and allergic rhinitis for several years, and reports having used inhalers like Seretide®.
Visceral axis of the neck, temporomandibular joints and neck palpation were unremarkable.
The visual examination of the area showed bilateral enlargements in the upper and lower lips, which were strongly noticeable when the lips were not in resting position. The enlargements involved the mucosal aspect of both lips, were elliptical in shape with the major axis parallel to the free edge of the lip. Enlargements were found in each hemilip. The mucosa covering them was normal in color and texture, and they were painless and soft to the touch. There was no family history of this kind of malformation.
The intraoral examination showed permanent dentition in canine to canine cross-bite. Upper lateral incisors were conoid in shape. The patient was undergoing the planning stage of periodontal and orthodontic treatment.

Fig.1. Preoperative image of the patient (front view). Upper and lower lips in occlusion.

The following complementary tests were ordered: a complete blood count, which was normal; a thyroid blood test, which revealed normal levels of the thyroid stimulating hormone (TSH); and a thyroid ultrasound, which showed a slightly enlarged thyroid, as shown in Fig. 2.

Fig. 2. Ultrasound

Positive diagnosis of the case: Ascher Syndrome. The treatment plan chosen was surgical resection of lip enlargements with local anesthesia in two stages.

In the first stage we resected the upper lip enlargements (Fig. 3) and also performed a frenectomy of the upper lip for orthodontic reasons.

In the second stage, thirty days later, the surgical procedure was performed in the lower lip.

Both surgeries were performed using elliptical excision techniques with cold scalpel and the affected areas were sutured using separate stitches (Fig. 3).

Fig. 3. Surgical sequence in both lips: Preliminary elliptical markings of the lesion to be resected in the upper lip; removal of excess tissue; frenectomy; bloody surface after frenectomy; suture with separate stitches and twenty-five day postoperative follow-up of the upper lip. Preliminary marking of the lesion to be resected in the lower lip; resection of the excess tissue masses in the inner lip; and suture with separate stitches in the lower lip.

The histopathologic study of the surgical specimen obtained confirmed the clinical diagnosis. The two histological sections of the case presented here (Fig. 4) show, respectively:
- A panoramic view of labial mucosa.
- The submucosa of the lip with an abundance of collagen fibers, as well as a large number of mucous lip salivary glands.

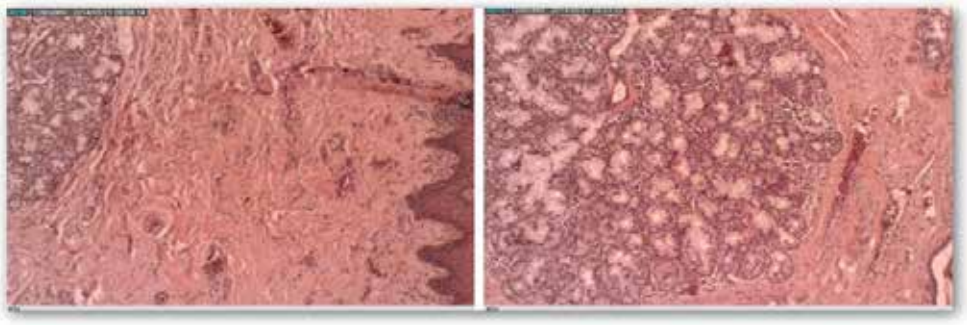

Fig. 4. Anatomo-pathological study of the surgical specimen.

The aesthetic surgical outcomes were very satisfactory for both patient and surgeon. This is shown in frontal images after thirty days (Fig. 5) and in profile images six months after surgery (Fig. 6). Frontal and profile images three years after surgery (Fig. 7)

Fig. 5. Front view after 30 days.

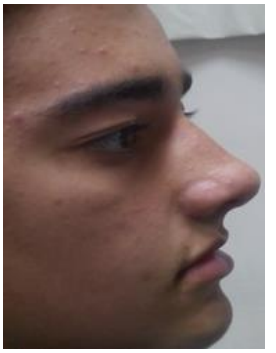

Fig. 6. Profile after 6 months.

Fig. 7. Clinical aspects three years after surgery. Orthodontic treatment in progress.

Discussion

The complete form of the AS, double lip, blepharochalasis and thyroid enlargement, was described by Ascher in 1920[5,6].

The first case of double lip with blepharochalasis was described and reported by Laffer in 1909[1,9].

As a consequence, some authors such as Lebuisson D, Leroy L, Aron JJ, Jeaneau E, J. Guillard J P. refer to it as the "Laffer-Ascher Syndrome"[13].

Because nontoxic thyroid goiter is not present in all cases (in only 10 to 50%)[1,10,12-15], many authors recommend not considering it when diagnosing AS.

Other authors[1,4,10,12-15] think nontoxic thyroid goiter is an inconsistent finding in some patients only during the initial stages of the syndrome, and that it can appear several years later, more slowly and delayed in relation to the lip and lid enlargements.

Blepharochalasis was first named and described by Fuchs in 1896, as an atrophic and progressive enlargement of the skin of the eyelids which can also include recurrent bouts of edema that may exacerbate the condition[12,16], leading indirectly to vision impairment caused by the closure or narrowing of the palpebral aperture. In general, only the upper lids are affected and make it difficult for patients to keep their eyes open[12]. It is treated surgically, with a blepharoplasty to remove a wedge of the excess skin.

In AS, blepharochalasis can be larger or smaller in relation to the enlargement found in the double lip. Blepharochalasis and double lip usually appear at the same time and in more than 80% of cases they manifest before the age of 20[17].

Double lip is a fold of excess or redundant hypertrophic tissue on the mucosal side of the lip and is the main element of the triad of features that characterize AS[1].

It consists of excess tissue in the labial mucosa, located towards the inner part of the vermilion border, and not, strictly speaking, as its name seems to indicate, a duplication of the lip organ, but rather a localized enlargement of the labial mucosa.

In most of the cases reported the enlargement appears in only one lip. It frequently occurs bilaterally in the upper lip, and it less commonly affects the lower lip (ratio of 10:1 in favor of the upper lip)[1-10,12-14].

The etiology of the syndrome is yet to be determined, and it can be of congenital or acquired origin[4,10-12]. Inheritance can be considered for some patients, due to the known familial cases reported by Franceschetti[17] and Viallefot[4,14].

Also, since most of the cases appear during puberty, some authors believe it could be triggered by hormonal disorders[1,4].

In AS, the alterations in lips and eyelids occur abruptly and simultaneously, simulating an angioneurotic edema which partially diminishes.

The thyroid enlargement becomes noticeable more slowly and later, with a normal uptake of radioiodine (131I) and is only present in 10 to 50% of cases[10-12,14,15].

As for double lip, the following have been described: a congenital form[14] which develops during the second or third month of intrauterine life[11], and an acquired form which may occur in isolation or as part of a syndrome such as AS[1].

Isolated forms can be triggered by different factors such as habit or trauma (biting, suction, orthopedic and/or orthodontic appliances, etc.)[11,18-20]. They usually remit or their volume diminishes once the habit or trauma-causing agent is removed.

In general, the lip deformity is not visible when patients keep their lips closed (except in the larger cases), but it appears when they open their mouths or smile.

Macroscopically, in the upper lip it can manifest as a soft, mobile, overhanging mass which on occasion can extend from one commissure to the other, or can frequently be divided in the middle by the labial frenum, resulting in two globular or oblong, middle-sized masses[1].The latter applies to the case presented here.

In the lower lip (very uncommon) it almost exclusively appears as two middlesized bunshaped masses, as shown in the patient's photograph.

It can exceptionally be found in both lips[1,4,14], as in this patient.

Histological examination showed masses of loose connective tissue, associated with numerous mucous salivary glands, like the ones normally occurring in the area[1,12], which is consistent with the histopathologic study of the surgical specimen obtained.

To make a positive diagnosis of AS, we must find the clinical triad described above, and differential diagnosis with other lip enlargements, such as trauma macrocheilitis, cheilitis glandularis, lymphangioma, angioedema, granulomatous cheilitis, inflammatory fibrous hyperplasia, sarcoidosis and salivary gland tumors.

Surgical treatment is indicated when there is interference with phonation or mastication, habit of suctioning the hyperplastic tissue and, mainly, for aesthetic reasons, when it affects the psychological state of the patient, as in this case.

As for the prognosis, these lesions tend not to relapse.

Different surgical techniques to correct a double lip have been described in the literature, all of which are for the upper lip, with the most common one being the elliptical excision of the enlargement, proposed by Digman in 1947[21].

This technique is used by many authors until today, like Cortes-Aroche[1], who published a description in 2007.

Reddy and Kotewara[22] and Dorrance[23] also advocate using the elliptical excision to remove the excess mucosa and the direct suture.

Guerrero Santos and Altamirano[24] suggest using a W-plasty to avoid removing too much mucous tissue, a mistake which, according to them, can be made with the elliptical excision.

Lamster[25] described a technique consisting of a triangular excision of mucosa, while Eski[26] described a technique with a bilateral elliptical excision for the cases in which there is a central bridle, associated with a Z-plasty at the level of the bridle.

Other authors describe combinations and small alterations of the techniques that have already been described[27-30].

As it was explained above, in this case we planned to resect in a first stage the upper lip enlargements and, on the following month, in a second surgical stage, those in the lower lip.

Both procedures were conducted using locoregional anesthesia.

We used elliptical excisions for both lips, suturing the affected areas using separate stitches, as described by Digman[21] Fig. 3.

We also performed a simultaneous labial frenectomy in the upper lip, as indicted by the treating orthodontists.

Conclusions

The Ascher Syndrome is a very rare condition with only a few cases reported in scientific publications.

It can even more rarely involve the upper and lower lips simultaneously, as in this patient, who suffers major psychological and emotional consequences.

Despite its low incidence, it is of particular interest for dentists, because they are often the first healthcare professionals to detect it, and they can make a diagnosis and treatment plan.

Treatment with the elliptical surgical excision technique, used in both lips in this specific case, provided very satisfactory psycho-emotional results for the patient in the short and long term (three years).

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