Official Publication of AÇBİD
(Association of Oral and Maxillofacial Surgery Society)

EurAsian Journal of Oral and Maxillofacial Surgery

January 2023 Volume 2 Number 1
e-ISSN: 2687-5497

EurAsian Journal of Oral and Maxillofacial Surgery

Official publication of AÇBİD (Association of Oral and Maxillofacial Surgery Society)

e-ISSN: 2687-5497

EurAsian Journal of Oral and Maxillofacial Surgery
Official publication of AÇBİD
(Association of Oral and Maxillofacial Surgery Society)

EurAsian Journal of Oral and Maxillofacial Surgery
Official publication of AÇBİD (Association of Oral and Maxillofacial Surgery Society)

RESEARCH ARTICLE

Assessment of the State-Trait Anxiety Relationship in Patients with Myalgia of Masticatory Muscles

Şakir KATI, DDS,PhD[1], Cahit ÜÇOK,DDS,PhD[2], Mustafa Ercüment ÖNDER,DDS,PhD[3]

[1]Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Ankara University, Ankara, Turkey,
[2]Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Ankara University, Ankara, Turkey,
[3]Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Kırıkkale University, Kırıkkale, Turkey

Abstract

Background

In clinical practice, non-dental facial pain patients are often seen. The hypothesis of this trial was that a link exists between trait anxiety and local myalgia of masticatory muscles. The study aimed to determine both the pre- and post-treatment anxiety levels of patients diagnosed with local myalgia.

Materials and Methods

Sixty patients diagnosed with local myalgia were asked to complete the State-Continuous Anxiety Inventory questionnaire before and 2 months after treatment. Scores were compared with a reference standard previously standardized for Turkish people on dental anxiety.

Results

State and trait anxiety levels were lower after the treatment than before the treatment. Trait anxiety before the treatment was the most intensive type, whereas that after the treatment was the least intensive type.

Conclusion

Although establishing a cause–effect relationship between local myalgia and anxiety is difficult, patients presenting with local myalgia symptoms should also be evaluated for clinical anxiety.

Practical Implications

It should be known by dentists that local myalgia patients who do not resolve with standard procedures should be evaluated for anxiety.

Key Words: Anxiety, State–trait anxiety, Myalgia, Temporomandibular disorders, TMD

Introduction

The term "temporomandibular disorders," (TMDs) first defined by Bell[1] and currently used by the American Dental Association[2], refers to all functional disorders of the mastication system. Temporomandibular disorders may originate from a joint pathology or various conditions involving the mastication muscles. According to DC/TMD classification updated by Schiffman et al in 2014, most common types of pain-related temporomandibular disorder is myalgia. Myalgia can be considered in two subgroups are myofascial pain which is the refers to a distant site and local myalgia which is the refers to overall muscle pain[3,4]. Local myalgia (LM) is characterized by pain or associated muscular spasms, tenderness, limited articular range of motion, stiffness, fatigue, or sometimes autonomic dysfunction originating from the trigger points found in stiff bands emerging from muscles and/or fascia. Its etiology is controversial and remains incompletely understood; although many factors may cause LM, psychiatric disorders such as anxiety, depression, personality disorders, fatigue, and stress appear to be the most critical[5].

State anxiety (SA) defines subjective fear in response to conditions of repression, which can be described as a state of perturbation, tension, fear, or unhappiness, and which disappears once the threatening factor is eliminated. Conversely, trait anxiety (TA) is defined as the tendency toward an oversensitive demeanor under stress or pessimism and intense emotive reactions independent of environmental

Corresponding Author: Şakir Katı
Address: Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Ankara University, Ankara, Turkey,
Mobile: +90(535) 4906886
E-mail: sakirkati@gmail.com

conditions[6]. The prevalence of psychiatric disorders associated with TMD has been reported to be approximately 66%–76%. The majority of accompanying psychiatric disorders have been emphasized as anxiety, atypical depression, somatoform disorder, and hypochondriasis alongside mild depression[7].
Previous studies have revealed a clear relationship between TMD and various psychiatric disorders. However, few studies have assessed psychiatric disorders specific to Turkish people, the validity and reliability of which have not been tested. The hypothesis of this trial was that a link exists between state–trait anxiety and LM. It aimed to determine both the pre- and post-treatment anxiety levels of patients diagnosed with LM.

Materials and methods

This study followed the Declaration of Helsinki regarding medical protocols and ethics and the Regional Ethical Review Board of Ankara University Faculty of Dentistry approved the study (2013/36290600/05). This study included 60 patients older than 18 years of both sexes, who presented to the Department of Oral and Maxillofacial Surgery of Ankara University Faculty of Dentistry between 2011 and 2014 with acute facial pain, limited mouth opening, and difficulty eating. Clinical and radiological examinations confirmed that the patients were free of intraarticular disorders such as internal derangement, osteoarthritis, or degeneration; they had accompanying complaints such as muscular spasm, tenderness, stiffness, fatigue in the facial musculature and no muscle pain to refers to distant site and thus all of them were diagnosed with LM. Thirty patients with internal derangement with joint noises, osteoarthritis, suspected pregnancy, or age under 18 years were excluded from the study.
This study involved a treatment group and a reference standard group. The treatment group consisted of the 60 aforementioned patients, whereas the reference standard group comprised values from a study conducted by Öner et al. that are regarded as the norm for Turkish society on dental anxiety[8].
Patients were re-evaluated by a more experienced clinician after being examined by the same investigator. Patients diagnosed with LM who agreed to participate in the trial were informed about the study and signed informed consent forms. The patients were asked about complaints and medical history and their answers were recorded in detail. The visual analog scale was used to determine how current complaints affected each patient's life, and the utmost care was taken to record patient conditions as accurately as possible. Muscle palpations (masseter and temporal muscle); joint sounds; maximum mouth opening; quantity of leftward, rightward, and protrusive joint motions; and any notable intraoral examination findings were recorded in detail. The patients were administered the Signed Trait–State Anxiety Inventory (STAI FORM TX-1) and the Signed Trait–Anxiety Inventory (STAI FORM TX-2). These inventory forms were validated and tested by Öner et al. for the Turkish population[8].
The patients who completed the STAI FORM TX-1 underwent the first session of infrared treatment (a thermotherapy agent) for 30 min, which targeted the tender points of the chin and face muscles that were detected during the examination and identified as trigger points. Trigger points are the oversensitive points inside palpable nodes located on stiff bands that are situated inside the musculoskeletal system. On the same day, the patients were prescribed tenoxicam 1 tablet per day and phenprobamate 1 tablet per day for 15 days to support thermotherapy with pharmacological agents. The patients were informed of the sedative and addictive effects of the phenprobamate. No further anxiolytic agents were used in our study. Thermotherapy can be described as the application of moist-hot and dry-hot agents to a patient's skin. Hot application evokes vasodilatation through direct or reflex pathways, thereby reducing the pressure on trigger points and relieving pain. After the first session, the patients were provided with a Points to Take into Consideration information sheet for home treatment, and they were instructed to comply with the written recommendations as far as possible. The patients received 30-min thermotherapy sessions using the infrared device for 5 sessions with 3-day intervals for a total of 15 days. The patients were instructed to stop their medications at the end of 15th day but to continue home treatment for 1 month from the start of treatment. Home therapy promotes habits relating to local formants, eating, mastication, and cushion and lying positions, as well as parafunctional habits that should not be performed. Patients received an explanation and were provided with a form that clarified these procedures. At the end of the 1-month period, the patients were invited to a control visit and any changes in their complaints were assessed. No other procedure was performed by the end of the 2-month period from the start of the treatment. When the 2-month period was completed, the patients were requested to complete STAI FORM TX-1 and STAI FORM TX-2.
The mean pre-treatment and post-treatment SA and TA scores were calculated separately. The scores were evaluated according to the Spielberger classification: a STAI score of 20–37 indicated no anxiety or low anxiety; a score of 38–44 indicated moderate anxiety; and a score ↗45 indicated high anxiety[6].
Prior to data analysis, the patient responses to the state–trait anxiety inventory, accuracy of the study data, completeness of the responses, and missing data were checked using various subprograms of the Statistical Package for the Social Sciences (IBM Corp. Released 2013. IBM SPSS Statistics for Windows, Version 22.0. Armonk, NY: IBM Corp).
The present study used data obtained from the norm groups defined by Öner et al.[8] as the reference standard. A sample in the state–trait anxiety inventory handbook was used as the norm group, which comprised 48 patients (Xtrait=35.13, standard deviation (SD)=9.43; Xstate=32.04, SD=10.7) who presented to a dentistry unit for toothache. The study data were analyzed using a one-sample t-test, an independent samples t-test with Spearman's correlation analysis, a decision tree analysis, and a dependent t-test with Spearman's correlation analysis. A p-value ←0.05 was considered statistically significant. In the decision tree analysis, further power analysis was deemed unnecessary because nonsignificant variables were automatically excluded from the analysis.

Results

Among the study patients, 48 (80%) were women and 12 (20%) were men. The youngest patient was 19 years old and the oldest was 66 years old. The mean age of the study population was 38.5 years. The minimum duration of pain was 1 month, the maximum duration was 36 months, and the mean duration of

7.15 months. Forty-five (75%) patients stated that pain caused limitations in their lives, whereas 15 (25%) patients stated the opposite. Twenty-four (40%) patients experienced pain in the morning, 22 (36.7%) in the evening, 1 (1.7%) at night, and 13 (21.7%) through the day. Forty-five (75%) patients could open their mouth less widely than before, whereas 15 (25%) patients could open their months as wide as before. Fifty-nine (98.3%) patients had tenderness of the masseter muscle, whereas 1 (1.7%) patient did not. Forty-five (75%) patients had temporal muscle tenderness, whereas 15 (25%) did not. The mean mouth opening was 43.42 mm. Forty-four (73.3%) patients had class I occlusion, 13 (21.7%) had class II occlusion, and 3 (5%) had class III occlusion. One (1.7%) patient stated that joint complaints did not affect daily life; 1 (1.7%) patient stated that joint complaints barely affected daily life; 16 (26.7%) patients stated that joint complaints mildly affected daily life; 30 (50%) patients stated that joint complaints markedly affected daily life; and 12 (20%) patients stated that joint complaints excessively affected daily life. The pre-treatment and post-treatment SA and TA scores of the study group are shown in Figure 1.

Tables 1–8

Table 1. Comparison of pre-treatment SA scores of the study and control arms (Single sample t-Test)

Group	N	Minimum	Maximum	Mean	SD	
Study pre-SA	60	21	69	54.97	10.13	
	Kolmogorov-Smirnov Normality Test					
	Test value	Degree of freedom		Asymptotic significance		
	0.14	60		0.055		
	t-Test					
	Reference standard group mean=32.04			Confidence interval		
	T test value	Degree of freedom	Asymptotic significance	Difference of means	Lower	Upper
	17.517	59	0.000	22.927	20.31	25.55

Since Kolmogov-Smirnov normality test results had a p=0.055→0.05, it can be stated with a 95% confidence level that the distribution of patients in the treatment arm conformed with a normal distribution and t-Test was applicable. It can be stated with 95% confidence level that the mean pre-treatment and post-treatment SA score of the study arm (54.97-anxious) was **significantly greater** than that of the control arm (32.04- less anxious). (SA: State anxious, SD: Standard Deviation)

Table 2. Comparison of pre-treatment TA scores of the study and control arms (Single sample t-Test)

Group	N	Minimum	Maximum	Mean	SD	
Study pre- TA	60	33	72	58.08	8.96	
	Kolmogorov-Smirnov Normality Test					
	Test value	Degree of freedom		Asymptotic significance		
	0.08	60		0.2		
	t-Test					
	Reference standard group mean=35.13			Confidence interval		
	T test value	Degree of freedom	Asymptotic significance	Difference of means	Lower	Upper
	19.835	59	0.000	22.953	20.64	25.27

Since Kolmogorov-Smirnov normality test results yielded p=0.2→0.05 and as the distribution of the pre-TA values of the patients in the treatment arm was normal in a confidence level of 95%, t-Test was applicable. It can be stated with 95% confidence level that the mean pre-treatment TA score of the study arm (58.08- most anxious) was significantly greater than that of the control arm (35.13-less anxious). (TA: Trait anxious, SD: Standard Deviation)

Table 3. Comparison of the post-treatment SA scores of the study arm and the SA scores of the control arm

Group	N	Minimum	Maximum	Mean	SD	
StudyPost-SA	60	21	48	33.62	5.04	
	Kolmogorov-Smirnov Normality Test					
	Test value	Degree of freedom		Asymptotic significance		
	0.075	60		0.2		
	t-Test					
	Reference standard group mean=32.04			Confidence interval		
	T test value	Degree of freedom	Asymptotic significance	Difference of means	Lower	Upper
	2.42	59	0.059	1,577	0.27	2.88

Since Kolmogorov-Smirnov normality tests results showed p=0.2→0.05, the distribution of SA values of patients in the treatment arm was normal with the confidence level of 95%, t-Test was applicable. It can be stated with 95% confidence level that there was no significant difference between the mean post-treatment SA score of the study arm and the mean state anxiety score of the control arm. (SA: State anxious, SD: Standard Deviation)

Table 4. Comparison of the post-treatment TA score of the study arm and the post-treatment TA scores

Group	N	Minimum	Maximum	Mean	SD	
Study postTA	60	34	57	46.05	4.63	
	Kolmogorov-Smirnov Normality Test					
	Test value	Degree of freedom		Asymptotic significance		
	0.098	60		0.2		
	t-test					
	Reference standard group mean=35.13			Confidence interval		
	T test value	Degree of freedom	Asymptotic significance	Difference of means	Lower	Upper
	18.254	59	0.000	10.920	9.72	12.12

Since Kolmogorov-Smirnov normality tests results showed p=0.2→ 0.05, TA Values of patients in the treatment arm showed a normal distribution at a confidence level of 95%, t-Test is applicable. It can be stated with 95% confidence level that the study arm had a greater mean post-treatment TA score than the control arm. (TA: Trait anxious, SD: Standard Deviation)

Table 5. Comparison of post-treatment SA scores of the study and control groups

Group	N	Minimum	Maximum	Mean	SD	
Study post SA	60	21	48	33.62	5.04	
	Kolmogorov-Smirnov Normality Test					
	Test value	Degree of freedom		Asymptotic significance		
	0.075	60		0.2		
	t-Test					
	Reference standard group mean=32.04			Confidence interval		
	T test value	Degree of freedom	Asymptotic significance	Difference of means	Lower	Upper
	2.42	59	0.059	1.577	0.27	2.88

Since Kolmogorov-Smirnov normality tests results showed p=0.2→0.05, distribution of SA Values of patients in the treatment arm had a normal distribution at a confidence level of 95%, t-Test was applicable. According to t-Test results, since p=0.059→0.05, it can be stated with a confidence level of 95% that there was no statistically significant difference between groups. (SA: State anxious, SD: Standard Deviation)

Table 6. Comparison of pre- and post-treatment SA scores

Study Group	N	Minimum	Maximum	Mean	SD	
Pre-SA	60	21	69	54.97	10.13	
Post-SA	60	21	48	33.62	5.04	
Dependent t test	Test value	Degree of Freedom	Asymptotic significance	Difference of Means	Lower confidence interval	Upper confidence interval
	19.71	59	0.000	21.35	19.18	23.51

Since Dependent T-Test results showed p=0.000←0.05, it can be stated with ← confidence level of 95% that a statistically significant difference was present between the two groups. It can be stated with 95% confidence level that the mean pre-treatment SA score (54.97-anxious) was greater than the mean post-treatment SA score, and the post-treatment SA was reduced compared to the pre-treatment level. (SA: State anxious, SD: Standard Deviation)

Table 7. Comparison of pre- and post-treatment TA scores

Study Group	N	Minimum	Maximum	Mean	SD	
Pre-TA	60	33	72	58.08	8.96	
Post-TA	60	34	57	46.05	4.63	
Dependent t test	Test value	Degree of Freedom	Asymptotic significance	Difference of Means	Lower confidence interval	Upper confidence interval
	13.332	59	0.000	12.033	10.227	13.839

It can be stated with confidence level of 95% that the mean pre-treatment TA score (58.08-anxious) was greater than the post-treatment TA score (46.05-anxious), and that it was reduced after the treatment. (TA: Trait anxious, SD: Standard Deviation)

Table 8. Comparison of the Pre-treatment TA, post-treatment SA, pre-treatment TA, and post-treatment TA scores (ANOVA)

	Difference of means	Standard error	Asymptotic significance	Confidence interval	
				Lower	Upper
Pre-treatment SA					
Post-treatment SA	21.35	1.385	0,00	18.62	24.08
Pre-treatment TA	-3.117	1.385	0,25	-5,84	-0,39
Post-treatment TA	8.917	1.385	0,00	6.19	11.64
Post-treatment SA					
Pre-treatment SA	-21.350	1.385	0,00	-24.08	-18.62
Pre-treatment TA	-24.467	1.385	0,00	-27.19	-21.74
Post-treatment TA	-12.433	1.385	0,00	-15.16	-9.71
Pre-treatment SA					
Pre-treatment SA	3.117	1.385	0,25	0,39	5.84
Post-treatment SA	24.467	1.385	0,00	21.74	27.19
Post-treatment TA	12.033	1.385	0,00	9.31	14.76
Post-treatment SA					
Pre-treatment SA	-8.917	1.385	0,00	-11.64	-6.19
Post-treatment SA	12.433	1.385	0,00	9.71	15.16
Pre-treatment TA	-12.033	1.385	0,00	-14.76	-9.31

It can be stated with a confidence level of 95% and a significance level of p←0.05 that the magnitudes of the scores were in the order of Pre-treatment TA→Pre-treatment SA→Post-treatment TA→post-treatment SA. (TA: Trait anxious, SA: State anxious)

Discussion

The importance of psychological factors in the development of temporomandibular disorders is well-known[7,10]. Numerous studies have also reported that temporomandibular disorders accompany some psychological disorders, such as anxiety, depression, and personality disorders[11-17]. The hypothesis of the current study was that a cause and effect relationship exists between LM and state–trait anxiety levels. The present study results showed that the patients in the treatment group had higher levels of both anxiety types compared to those of the reference standard. The mean post-treatment TA scores of the treatment group were higher than those of the reference standard, indicating that TA persisted after treatment. SA is thought to be resolved by LM treatment, but TA requires other advanced treatment modalities, such as cognitive behavior therapy or long term used anxiolytic medication.

Although epidemiological data on LM in temporomandibular disorders remain unclear, the disorder has been reported to typically affect women 3–6 times more often than men. This difference has been explained by women seeking treatment more commonly than men, but also by hormonal and ergonomic factors. A study of patients with acute temporomandibular joint disorders revealed that pain-related TMD patients seeking treatment was predicted by sex, pain intensity, and psychosocial stress[18]. Friction et al.[19] reported that 135 (82.3%) of 164 patients with myofascial pain in the neck or face were women. The present study also demonstrated that 48 (80%) of 60 patients were women, thus supporting previous studies suggesting that the disorder is more common among women. Myofascial pain most commonly occurs between 20 and 40 years of age. In accordance with previous reports, the patients in the current study had a mean age of 38.5 years[20-22].

In a study by Arı[23], patients with myofascial pain had the most severe pain in the morning immediately after waking up, and it recurred following both excess activity and prolonged periods of inactivity. The present study showed that 40% of patients had pain in the morning, 36.7% in the evening, 1.7% at night, and 21.7% throughout the day.

Questioning the patients about the duration of their symptoms revealed that the majority of patients had a chronic disease. Their myofascial pain lasted between 1 month and 36 months, with a mean duration of 7.15 months. The duration of myofascial pain was 1–9 months in 76.7% of patients; 10–19 months in 11.7%; 20–29 months in 5%; and 30–39 months in 6.7%. These findings are in agreement with previous reports, which have shown that the age of onset of the disorder is 18–26 years and the age at the time of presentation to a physician is 20–50 years[24].

Among the whole study group, 66.7% (40 patients) considered their disorder to have started because of stress. The patients commonly had difficulties with personal relationships. Emotional stress has been reported as a predictor of myofascial pain or local myalgia because it causes the emergence of parafunctional behaviors[25]. In a study of the effect of stressful life events on pain intensity and depression, anxiety levels, and treatment outcomes, social interaction problems originating from interpersonal problems were significantly more common in patients with myofascial pain than in those with non-muscular temporomandibular joint disorders[14].

Simons et al.[26] described myofascial pain as a syndrome in which limited mouth opening may or may not be present. According to the statements of the patients in the current study, 68.3% of patients had difficulty in opening their mouth, whereas 31.7% had no difficulty; in general, 75% of patients could open their mouth less than before, whereas 25% had the same mouth opening as before. Simons et al.[26] defined myofascial pain as a syndrome originating from the trigger points found in the stiff bands in which facial pain accompanies muscle tenderness on palpation, which is formed by local and referred pain, and which limits daily life. Seventy-five of our patients reported a limitation in daily life caused by pain, whereas 25% of them did not. We did not formally detect or evaluate trigger points within stiff bands. Regarding the findings of muscular tenderness upon palpation, 98.3% of patients had tenderness in the masseter muscle, whereas 1.7% of them did not; additionally, 75% of patients had tenderness in the temporal muscle, whereas 25% of them did not.

The results of the chi-square test suggested, at a confidence level of 95%, a significant relationship between sex and pre-treatment TA. Thus, pre-treatment TA varies by sex. The coefficient of this correlation (i.e., the Phi coefficient) was 0.379. Considering that the Phi coefficient can have a value between 0 and 1, 0.379 may be considered small, but it was statistically significant. Based on pre-treatment TA levels, 66.7% of women and 25% of men had high anxiety.

Frederiksson et al.[27] conducted a pain threshold study and advocated that evaluating men and women separately would be more useful. Women have an increased incidence of 30–49 years of age and the trigger points are more common at a ratio of 3:1[28].

Although myofascial pain occurs in both sexes, its prevalence was higher among women than men and has been reported to be more common in the second half of the menstrual cycle, reaching a maximum prevalence between 30 and 39 years of age, and has a lower prevalence in elderly people[26,28,29].

Numerous studies have indicated that temporomandibular disorders accompany psychological disorders such as anxiety, depression, and personality disorders [10-16]. A study by Merksey[30] showed that anxious and depressive symptoms increased in prevalence in facial arthromyalgias, albeit to a lesser degree than other conditions characterized by chronic pain. Although many factors lead to myofascial pain, psychiatric disorders, fatigue, and stress appear to be the most critical. Suvinen et al.[31] and Koh[32] reported that among temporomandibular disorders, myofascial pain or local myalgia is the most thoroughly studied from the psychiatric aspect; the researchers also added that no definitive conclusions could be drawn from available studies because of a lack of consensus regarding the diagnosis, diversity of psychiatric assessment and evaluation systems, and the multidimensional property

of myofascial pain. Generally, however, myofascial pain is emphasized as a disorder that is closely related to stress, in which both first axis and second axis psychiatric disorders are common.

Similar to our study, Krishnan et al.[33] and Brown[34] found that depressive and anxious symptoms were more common in chronic pain populations than in control groups.

Conclusion

The current study results suggest that the mean pre-treatment SA score regressed following treatment, indicating that the applied treatment protocol was effective against SA. The mean pre-treatment TA score was higher than the mean post-treatment TA score, and post-treatment TA was lower than pre-treatment TA. Similarly, the applied protocol exerted a beneficial effect on the TA level but failed to completely abolish TA.

In patients with myofascial pain or myalgia, a prominent psychopathological condition may be correlated to anxiety levels. Although establishing a cause–effect relationship between myalgia and anxiety is difficult, patients presenting with myalgia symptoms should also be evaluated for anxiety. Our findings suggest that clinicians should carefully assess and provide guidance for reducing muscle tension, parafunctional activity, emotional distress and high levels of overall stress in their patients experiencing TMD-related myofascial pain.

Declaration of conflicting interests

The authors declared no conflicts of interest with respect to the research, authorship and/or publication of this article.

Funding

The authors received no financial support for the research, authorship and/or publication of this article.

ORCIDs:

Şakir Katı: https://orcid.org/0000-0002-9688-6058
Cahit Üçok: https://orcid.org/0000-0003-3566-4731
M Ercüment Önder: https://orcid.org/0000-0002-1488-616X

References

1. Bell WE. Clinical diagnosis of the pain dysfunction syndrome. J Am Dent Assoc 1969; 79(1):154-160.
2. Simmons HC. A Critical Review of Dr. Charles S. Greene's Article titled "Managing the Care of Patients with Temporomandibular Disorders: A New Guideline for Care" and A Revision of the American Association for Dental Research's 1996 Policy Statement on Temporomandibular Disorders, approved by the AADR Council in March 2010, Published in the Journal of the American Dental Association September 2010. J Craniomand Pract.2012;30(1):9-24.
3. Peck CC, Goulet JP, et al. Expanding the taxonomy of the diagnostic criteria for temporomandibular disorders. J Oral Rehabil. 2014;41(1):2–23 .
4. Schiffman E, Ohrbach R, et al. Diagnostic criteria for temporomandibular disorders (DC/TMD) for clinical and research applications: recommendations of the international RDC/TMD consortium network and orofacial pain special interest Groupdagger. J Oral Facial Pain Headache. 2014;28(1):6–27.
5. Önder E, Işıklı S, Pampu A. Miyofasiyal ağrı disfonksiyon sendromu ile durumluk sürekli kaygının ilişkisi. [Relationship between Myofascial Pain-Dysfunction Syndrome and State-Trait Anxiety] TDB Derg. 2009; 76:130-132. Turkish.
6. Spielberger CD. Manual for the State-Trait Anxiety Inventory STAI. Consulting Psychologists Press, Inc, Redwood City, CA (1983)
7. Epker J, Gatchel RJ. Coping profile differences in the biopschosocial functioning of patients with temporomandibular disorder. Psychosom Med.2000;62:69-75.
8. Öner N, Le Compte A. Durumluk-sürekli kaygı envanteri el kitabı. [A Handbook of State-Trait Anxiety Invantory] İstanbul: Boğaziçi Üniversitesi Yayınları, 1985. Turkish
9. Turner JA, Dworkin SF, Mancl L, et al. The role of beliefs, catastrophizing, and coping in the functioning of patients with temporomandibular disorders. Pain.2001;92:41-51.
10. Dworkin SF, Sherman J, Mancl L, et al. Reliability, validity, and clinical utility of the research diagnostic criteria for temporomandibular disorders axis II scales: depression, non specific physical symptoms, and graded chronic pain. J Orofac Pain.2002;16:207-220.
11. Madland G, Feinman C, Newman S. Factors associated with anxiety and depression in facial arthromyalgia. Pain. 2000;84:225-232.
12. Gatchel RJ, Garofalo JP, Ellis E, et al. Major psychological disorders in acute and chronic TMD: an initial examination. J Am Dent Assoc.1996;127:1365-1370.
13. Auerbach S, Laskin D, Frantseve LME, et al. Depression, pain, exposure to stressful life events and long term outcomes in temporomandibular disorder patients. J Oral Maxillofac Surg. 2001;59:628-633.
14. Manfredini D, Bandettini diPA, Romagnoli M, et al. A spectrum approach for the assessment of manic-depressive symptoms accompaying temporomandibular disorders. Minerva Stomatol. 2003;52: 231-240.
15. Mongini F, Ciccine J, Ibertis F, et al. Personality characteristics and accompaying symptoms in temporomandibular joint dysfunctions, headache and facial pain. J Orofac Pain. 2000;14:52-58.
16. Yap AU, Dworkin SF, Chua EK, et al. Prevalance of temporomandibular disorders subtypes, psyhcologic distress and psychosocial dysfunction in Asian patients. J Orofac Pain. 2003;17:21-28.
17. Nifosi F, Violato E, Sifari L, et al. Clinical psychiatric assessment and pyscopathological measure of patients with myofacial and temporomandibular joint pain. Int J Pyschiatry Med.2007;37:238-240.
18. Miyachi H, Wake H, Tamaki K. Detecting mental disorders in dental patients with occlusion related problems. Phychiatry Clin Neurosci.2007;61:313-319.

19. Friction JR, Kroening R, Haley D, et al. Myofacial pain syndrome of the head and neck: A review of clinical characteristics of 164 patients. Oral surgery, Oral Medicine, Oral Pathol.1985;60: 615-623.
20. Fricton JR. Clinical care for myofacial pain. Dent Clin North Am.1991;35:1-8.
21. Marbach JJ. Temporomandibular pain and dysfunction syndrome. History, physical examination and treatment. Rheumatic Dis Clin North Am.1996;22:477-498.
22. Nekora-Azak A, Evlioğlu G, Ordulu M, et al. Prevalance of symptoms associated with temporomandibular disorders in a Turkish population. J Oral Rehab.2006;33:81-84.
23. Ari, H. Miyofasyal ağrı sendromunda laser ve tetik nokta enjeksiyonunun karşılaştırılması. [Comparison of Laser and Trigger Point Injection Methods in Myofascial Pain Syndrome] PhD Thesis (2002) Gülhane Askeri Tıp Akademisi, İstanbul. Turkish
24. Bessete RW, Jacobs JS. Grabb and Smith's Plastic Surgery, Temporomandibular Joint Dysfunction, Lippincott-Raven Publishers 1997; pp:88-90.
25. Glaros AG, Williams K, Lausten L. The role of parafunctions, emotions and stress in predicting facial pain. J Am Dent Assoc.2005;136(4):451-458.
26. Simons DG, Travell JG, Simons LS. Travell&Simons Myofascial Pain and Dysfunction. The triger point manual, Vol 1. Upper Half of Body.2nd ed. Baltimore, Philadelphia. Wilkins&Wilkins, 1999; pp:120-124.
27. Frederikson L, Alstergen P, Kopp S. Absolute and relative facial pressure pain thresholds in healthy individuals. J Orofac Pain.2000;14(2):98-104.
28. Han SC, Harrison P. Myofascial pain syndrome and triger point management. Reg Anest.1997;22: 89-91.
29. Cooper BC, Alleva M, Cooper DL, et al. Myofascial pain dysfunction analysis of 476 patients. Laryngoscope.1986;96:1099-1102.
30. Merskey H, Brown J, Brown A, et al. Psycological normality and abnormality in persistent headache patient. Pain.1985;Sep23(1):35-37.
31. Suvinen TI, Reade PC, Kemppainen P, et al. Review of aetilogical concepts of temporomandibular pain disorders: towards a biopsychosocial model for integration of physical disorder factors with psycological and psycosocial illness impact factors. Eur J Pain.2005;9:613-633.
32. Koh KB. Anger and somatization. J Psychosom Res.2003;55:113-120.
33. Krishnan KR, France RD, Pelton S, et al. Chronic pain and depression.I. Classification of depression in chronic low back pain patients. Pain.1985;Jul22(3):279-287.
34. Brown GK. A casual analysis of chronic pain and depression. J Abnorm Psychol.1990;99(2):127-137.

EurAsian Journal of Oral and Maxillofacial Surgery
Official publication of AÇBİD (Association of Oral and Maxillofacial Surgery Society)

CASE REPORT

Cemento-Ossifying Fibroma of the Mandible: A Case Report

Kadircan KAHVECİ,DDS[1], Ferhat AYRANCI, DDS PhD[1], Mehmet Melih ÖMEZLI, DDS PhD[1], Mustafa AY, DDS[1]

[1]Ordu University, Faculty of Dentistry, Department of Oral and Maxillofacial Surgery, Turkey

Abstract

Objective

Cemento-ossifying fibroma is a rare benign fibro-osseous lesion that originate from the mesenchymal cells of the periodontium. Enucleation or resection may be considered as treatment options of this lesion. Here we present surgical treatment of a case of cemento-ossifying fibroma located in the ramus of the mandible.

Case

A 12-year-old male patient was admitted to our clinic for routine examination. After clinical and radiological examinations, multilocular and non-expanding lesion was detected in the posterior region of the right mandible. Enucleation was performed under general anesthesia with intraoral approach.

Conclusion

Cemento-ossifying fibroma can exhibit different behaviors clinically and radiologically. After performing differential diagnosis from other fibroosseous lesions conservative treatment such as enucleation with follow-up will be more appropriate treatment option than radical surgeries like resection, especially in young patients.

Keywords: Cemento-ossifying fibroma, mandibula, enucleation

Introduction

Cemento-ossifying fibroma (COF) is uncommonly seen fibro-osseous lesion originate from mesenchymal cells of the periodontal ligament [1,2]. It consists of fibrous tissue matrix and contains calsified or cementoid structure[3]. The COF usually occurs premolar and molar region of the mandible without any symptoms or rarely seen with some symptoms such as expansion, paresthesia or anesthesia [3,4]). It has slow growth pattern and radiologically, is shown well-demarcated, radiolucent or mixed image [2,5]. It is central neoplasm of the bone and induced confusing of terminology because of the clinical and radiological behavior [1]. Thus, it should be separated from other fibro-osseous neoplasm and determined the most appropriate treatment option. Enucleation or surgical resection which is used in treatment of large or aggressive lesions, may be considered as the treatment methods in the management of COF [6,7]. In this case report, we present surgical treatment of a case of cemento-ossifying fibroma located in the ramus of the mandible.

Case Report

A 12-year old male patient admitted to Oral and Maxillofacial Surgery Service of Ordu University for the routine examination. After clinical and radiological examination, well-demarcated radiolucent lesion in the ramus of the right mandible which includes calcified material and impacted second and third molars, was observed (Figure 1). The patient had no swelling, expansion, paresthesia and lymphadenopathy.

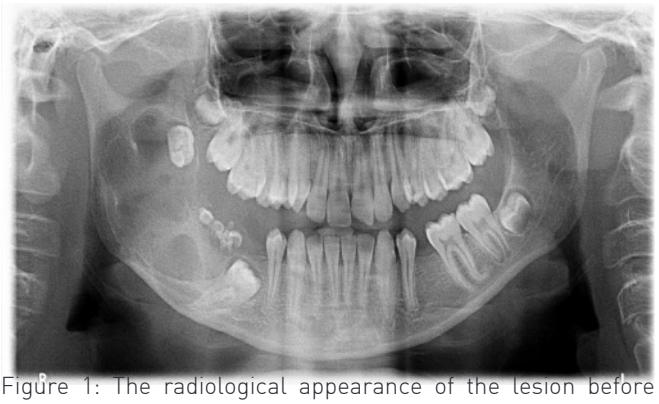


Figure 1: The radiological appearance of the lesion before surgery.

Corresponding Author: Kadircan Kahveci
Research Assistant
Address: Ordu University Faculty of Dentistry, Cumhuriyet Mah. Mustafa Kemal Bulvarı No: 254, Ordu
Mobile: +90(538).9783224
E-mail: kahveci_kadircan@hotmail.com

Surgical procedure

Conservative surgical exsicion with intraoral approach was planned under general anesthesia. After the endotracheal intubation, local anesthesic infiltration with 2% articain was done. A full thickness flap was raised buccally and lingually to identify the tumour mass. The lesion was enucleated totally, the impacted tooth were removed and then, the cavity was irrigated with saline solution. The primary closure was done and the enucleated specimen was sent to histopathological examination within 10% formalin solution. No complication and recurrence occur during the periodic follow-up of the patient through 18 months (Figure 2).

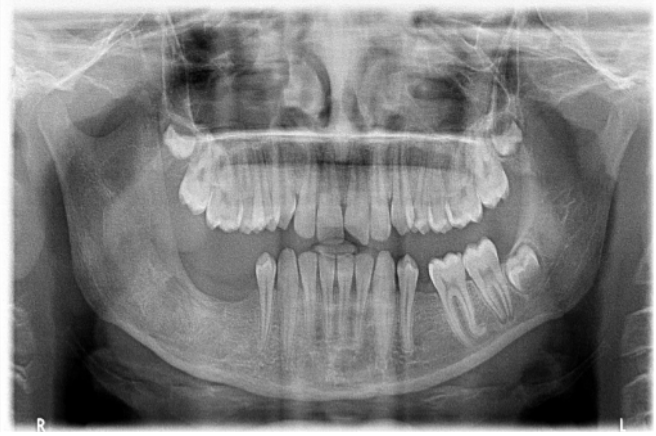


Figure 2: The post-operative 6th-months control shows the new bone formation.

Discussion

The COF is a benign neoplasm whose etiology is still under debate. It considered to occur as a result of the differentiation of the periodontal ligament cells associated with infection, trauma or tooth extraction[4,8]. However, the presence of this lesion in other anatomic regions such as maxillary antrum, temporal or frontal bone outside the jaws suggests that the ectopic periodontal membrane or undifferentiated mesenchymal cells may be considered as the other possible etiological factors [2,9]. In this case, we think that the history of trauma reported by the patient may be the possible etiologic factor that contribute the occurrence of the tumor.

The COF has a characteristic centrifugal growth pattern that represents the growth of the tumor equally in all directions [1,9]. It is usually treated with enucleation; however, a recurrence rate of % 28 in patients who treated with enucleation or curettage, has been reported in the literature[10]. The resection may be considered for aggressive lesions that tend to grow and cause significant facial asymmetry [6,10]. We preferred the enucleation which was the minimally invasive treatment method, as the treatment option for the young patient in present case.

Considering the various clinical behaviors of the COF the most appropriate treatment modality should be chosen. Long-term follow-up is necessary because of the potential of recurrence.

References

1. Sarwar HG, Jindal M, Ahmad S. A case report of cemento-ossifying fibroma. Journal of maxillofacial and oral surgery. 2010;9(2):178-81.
2. Cecchetti F, Luciani F, Bramanti E, Bartuli F, Ottria L, Arcuri C. Cemento-ossyfying fibroma juvenile of the oral cavity. ORAL & implantology. 2010;3(1):33.
3. Bertrand B, Eloy P, Cornelis JP, Gosseye S, Clotuche J, Gilliard C. Juvenile aggressive cemento-ossifying fibroma: Case report and review of the literature. The Laryngoscope. 1993;103(12):1385-9.
4. Naik RM, Guruprasad Y, Sujatha D, Gurudath S, Pai A, Suresh K. Giant cemento-ossifying fibroma of the mandible. Journal of Natural Science, Biology, and Medicine. 2014;5(1):190.
5. Mohapatra M, Banushree C, Nagarajan K, Pati D. Cemento-ossifying fibroma of mandible: An unusual case report and review of literature. Journal of oral and maxillofacial pathology: JOMFP. 2015;19(3):405.
6. Khanna M, Buddhavarapu SR, Hussain SA, Amir E. Cemento-ossifying fibroma of paranasal sinus presenting acutely as orbital cellulitis. Journal of radiology case reports. 2009;3(4):18.
7. Katti G, Khan MM, Chaubey SS, Amena M. Case Report: Cemento-ossifying fibroma of the jaw. BMJ case reports. 2016;2016.
8. Dalghous A, Alkhabuli JO. Cemento-ossifying fibroma occurring in an elderly patient. A case report and a review of literature. Libyan journal of Medicine. 2007;2(2):95-8.
9. Bernier JL, Thompson HC. The histogenesis of the cementoma: Report of 15 cases. American journal of orthodontics and oral surgery. 1946;32(9):A543-A55.
10. Eversole L, Leider A, Nelson K. Ossifying fibroma: a clinicopathologic study of sixty-four cases. Oral surgery, oral medicine, oral pathology. 1985;60(5):505-11.

EurAsian Journal of Oral and Maxillofacial Surgery
Official publication of AÇBİD (Association of Oral and Maxillofacial Surgery Society)

CASE REPORT

Dentigerous Cyst Associated With a Transmigrated Canine of the Mandibular Symphysis: A Case Report

Rıdvan GÜLER[1], Osman HABEK[1], Bekir İLYAS[1], Kamil Serkan AĞAÇAYAK[1]

[1] Dicle University, Faculty of Dentistry, Departmant of Oral and Maxillofacial Surgery, Diyarbakır, Turkey

Abstract

Introduction

Transmigration is a infrequent phenomenon seen almost exclusively in the mandibular canines. Transmigration, is an intraosseous displacement of an unerupted teeth in which a movement phenomenon causes it to cross midline by more than half. Dentigerous cyst(DC) is one of the most common types of odontogenic cyst. Accumulation of fluid among the reduced enamel epithelium and the tooth crown is suggested as the possible etiopathogenesis of this particular cyst by most authors. Clinically, it is asymptomatic but can cause cortical bone expansion. DC is usually associated with an unerupted or developing teeth bud, and is found most often associated ith crowns of mandibular third molars followed by maxillary canines and then maxillary third molars. Early detection of a transmigrant tooth is essential for the treatment, planning and prevention of more complicated situations.

Case Report

An 30-year-old male was admitted to our department with a complaint of moderate pain in the left canine region of the mandible. On an intra-oral examination, we noted that the mandible left canine were clinically unerupted. The panoramic radiograph and 3D computed tomography revealed that the left mandibular canine had migrated to left side crossing the midline below the apices of the incisors, and showed pericoronal radiographic changes suggestive of cystic degeneration. DC was enucleated and the associated impacted teeth extracted under general anesthesia.

Conclusion

As a result is therefore important to perform radiographic-clinic examination of all unerupted teeth.

Keywords: transmigration, mandible symphysis, dentigerous cyst, canine

Introduction

Migration of a canine from its normal position to the contralateral hemiarch, crossing the midline is known as transmigration. This phenomenon is a infrequent, unusual developmental anomaly of unknown origin and it occurs almost exclusively with mandibular canines but also develops infrequently in maxillary canines as well.[1] The etiological factor that causes the migration of a tooth is still not clear.[2]

This condition was first reported by Aydin and Yılmaz[3] in 2003. In a review of 4,500 panoramic radiographs, Aydin et al.[6] identified eight mandibular canines and six maxillary canines (0.31 %). This anomaly is generally asymptomatic, with no pain or over pathology, and usually cannot be detected during a clinical examination. Transmigrating tooth can occasionally give rise to resorption of roots and tilting of adjacent teeth.[4]

DC are mostly asymptomatic and found incidentally during the evaluation of an unerupted teeth.[5] If the patient has infection and expansion, they become symptomatic. In the event of infection, it can cause a painful swelling. In the case of an expansion into cortical bone, DC can cause facial asymmetry, and destruction of the adjacent tissues. Histological diagnosis plays a key role in the definite diagnosis.This study discusses the case report of a transmigrated mandibular canine, which was also associated with a DC.

Case Report

An 30-year-old male was admitted to our department with a complaint of moderate pain in the left canine region of the mandible. He was systemically healthy and extra-oral examination was within normally. There was no sign of any regional lymphadenopathy. His mouth opening was normally.

Corresponding Author: Rıdvan Güler
Research Assistant
Address: Dicle University Faculty of Dentistry, Oral and Maxillofacial Surgery, Diyarbakır
Mobile: +90(534).7644921
e-mail: ridvanguler06@gmail.com

On an intra-oral examination, we noted that the mandible left canine were clinically unerupted.

For the radiological evaluation, a panoramic radiograph and CBCT was taken. The panoramic radiograph and 3D computed tomography revealed that the left mandibular canine had migrated to left side crossing the midline below the apices of the incisors, and showed pericoronal radiographic changes suggestive of cystic degeneration (Fig 1).

DC was enucleated and the associated impacted teeth extracted under general anesthesia (Fig 2-3). Surgical exposure of the canine with an excisional biopsy of the surrounding dentigerous cyst was performed.(Fig 4) The patient tolerated the procedure well. Patient was given a week course of antibiotic and analgesic. Sutures were removed on the ten day after operation and the postoperative course was uneventful. The extracted tooth with the cystic capsule was sent for histopathological examination. Hematoxylin and eosin-stained sections of the specimen were prepared. It showed thin connective tissue wall with a thin layer of stratified squamous epithelium (Fig 5). These features confirmed the features of dentigerous cyst. Post-operative clinical follow-up that was conducted after one and six month of the surgery was uneventful.

Figure 1

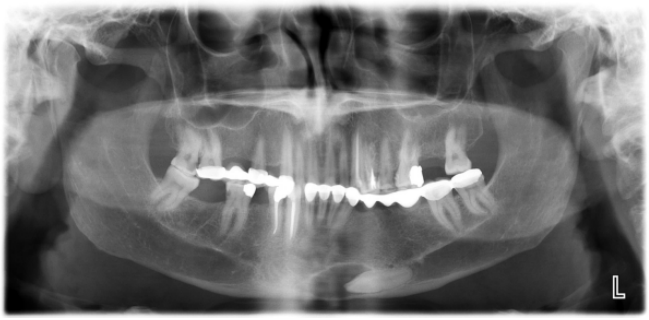


Figure 1: Panoramic radiograph showing horizontally impacted left mandibular canine located at symphysis of mandible crossing the midline.

Figure 2

Figure 2: Intra-operative view

Figure 3

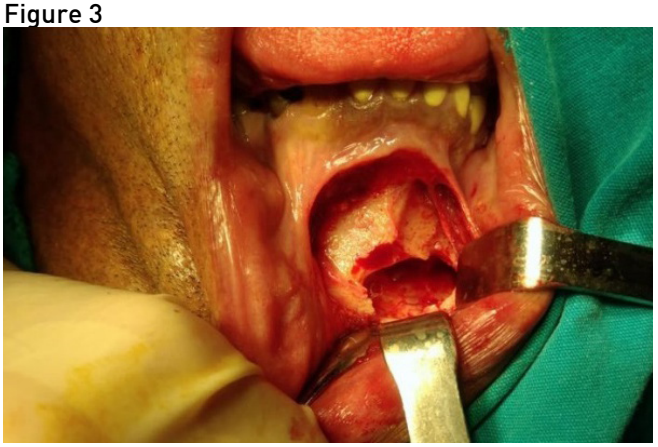


Figure 3: Intra-operative view

Figure 4

Figure 4: Histopathologic specimen

Figure 5

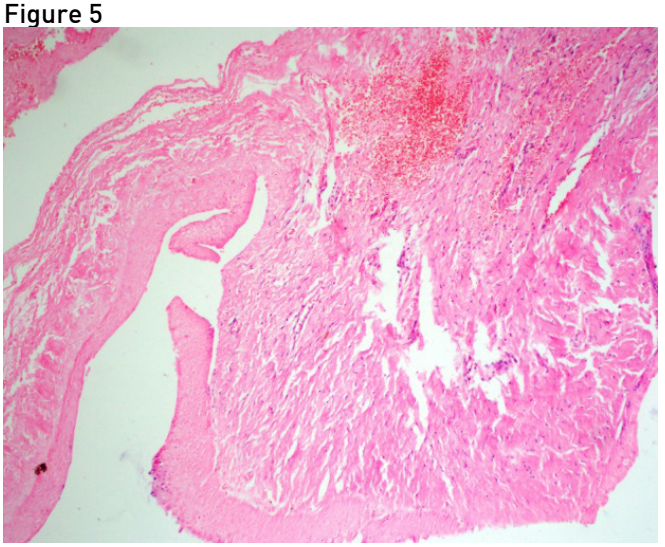


Figure 5: Histopathologic view of the lesion. (H&E, X 100)

Discussion

Canine impaction is more commonly in the maxilla than in the mandible, but canine transmigration is mostly in the mandible. The larger cross-sectional area of the anterior mandible compared with the anterior maxilla may be a reason for the higher frequency of mandibular canine transmigration.[6]

The etiology and exact mechanism of transmigration is still not clear, though a number of factors have been suggested. Tumors, cysts, supernumerer tooth and odontomas may cause malposition of teeth if they lie in the path of eruption of teeth. Other factors suggested by some authors as possible etiological factors are premature loss of deciduous teeth, retention of the deciduous canine and excessive length of the crown of the mandibular canines.[7]

Transmigrated mandibular canines are reported more frequently in females than males in the ratio of 1.6:1.8 In the present case impacted mandibular canines were classified based on angulations and depths of the involved teeth. In terms of angulation impacted mandibular canines can be classified as mesioangular, distoangular, vertical, or horizontal. Depth of the impactions were classified as Level A, Level B, and Level C as follows.[9]

Level A.The crown of the impacted caninetooth is at the cervical line of the adjacent tooth.

Level B.The crown of the impacted caninetooth is between the cervical line and rootapices of the adjacent tooth.

Level C.The crown of the impacted canines is beneath the root apices of the adjacent tooth.[9]

Transmigrated canines are classified by the criteria established by Mupparapu10 as follows.

- **Type 1:** Mesioangular canine with the crown crossing the midline, lateral or lingual to the anterior tooth.
- **Type 2:** Horizontal canine, near the lower edge of the mandible, under the apexes of the lateral incisors.
- **Type 3:** The canine is erupted, medially or distally to the opposite side.
- **Type 4:** The canine is horizontal near the lower edge of the mandible, under the apexes of the premolars and/or the contralateral molars.
- **Type 5:** The canine is positioned vertically, in the midline, with the long axis of the tooth crossing the midline.

Majority of times transmigrated canines are usually asymptomatic, although follicular cyst development surrounding the impacted tooth and chronic infection along with fistula creation have been recorded.[11] Nodine12, stated that migrated mandibular canines are frequently revealed without producing any obvious symptoms indicative of their existence. Ando et al.13 also described that they had not found any symptoms like pain or compression of mandibular nerve due to the transmigrated canines in their patient. As in our case, patient was accidentally diagnosed with impacted canine with absence of clinical symptoms.

Several treatment options are proposed for transmigrated canines, including surgical removal, transplantation, exposure and orthodontic treatment and in some cases plain follow.[4]

In our case, extraction was the ideal treatment option. Because presence of a cystic epithelium around the tooth attached to neck of tooth and the histopathological confirmation of a dentigerous cyst.

Conclusion

Transmigration of canine is a rare event caused by multiple etiologies. This case describes the presentation of the dentigerous cyst associated with impacted left lower canine crossing the midline which is a rare case in the literature. In conclusion is therefore important to perform radiographic-clinic examination of all unerupted teeth.

References

1. Joshi MR. Transmigrant mandibular canines: a record of 28 cases and a retrospective review of the literature. Angle Orthod. 2001; 71: 12-22.
2. Shapira Y, Borell G, Kuftinec MM, Nahlieli O. Bringing impacted mandibular second premolars into occlusion, Journal of the American Dental Association. 1996; vol. 127, no.7, pp. 1075–1078.
3. Aydın U, Yılmaz HH, Yıldırım D. Incidence of canine impaction and transmigration in a patient population. Dentomaxillofacial Radiology. 2004; vol. 33, no. 3, pp. 164–169.
4. Alexander B, Angel Morrison S, Swathy Anand PJ, Syamkumar V. Dentigerous Cyst associated with a Transmigrated Canine. International Journal of Preventive and Clinical Dental Research. January-March 2017;4(1):82-84.
5. Vasiapphan H, Christopher PJ, Kengasubbiah S, Shenoy V, Kumar S, Paranthaman A. Bilateral Dentigerous Cyst in Impacted Mandibular Third Molars: A Case Report. Cureus. 2018 Dec 5;10(12):e3691. doi: 10.7759/cureus.3691.
6. Özveri KB, Çetingül E. Transmigration of Impacted Canines: Six Case Reports. Hacettepe Diş Hekimliği Fakültesi Dergisi.2010; Cilt: 34, Sayı: 1-2, Sayfa: 23 -29.
7. Jayanthi P, Shaik Mohamed Shamsudeen SS, Rooban T, Umadevi M, Ranganathan K. Transmigration of Mandibular Canines: A Case Report. Indian Journal of Multidisciplinary Dentistry. 2011 May-June; Vol. 1, Issue 4.
8. Pinto L, Mengoni G, Ruscica C, Gastaldi G, Vinci R. Transmigration of a mandibular canine: case report. J Osseointegr.2020;12(3):236.
9. Bava FA, Azad AK, Farhan M, Siddeeqh S. A unilateral Dentigerous cyst associated with impacted mandibular canine crossing the midline: a rare case report. IOSR Journal of Dental and Medical Sciences. 2014 May; Volume 13, Issue 5 Ver. III.
10. Mupparapu M. Patterns of intra-osseous transmigration and ectopic eruption of mandibular canines: review of literature and report of nine additional cases. Dentomaxillofacial Radiology. 2002 Nov;31(6):355–60.
11. Rebellato J, Schabel B. Treatment of a patient with an impacted transmigrant mandibular canine and a palatally impacted maxillary canine. The Angle Orthodontist. 2003 Jun;73(3):328-36.
12. Nodine AM. Aberrant teeth, their history, causes and treatment. Dent Items Interest. 1943 Sep;65(2):440-51.
13. Ando S, Aizawa K, Nakashima T, Sanka Y, Shimbo K, Kiyokawa K. Transmigration process of the impacted mandibular cuspid. The Journal of Nihon University School of Dentistry. 1964;6(2):66-71.

EurAsian Journal of Oral and Maxillofacial Surgery
Official publication of AÇBİD (Association of Oral and Maxillofacial Surgery Society)

CASE REPORT

Soft and Hard Tissue Changes in the Oral and Maxillofacial Region in Patients with Neurofibromatosis: Rare Case Report

Günay YAPICI YAVUZ,DDS,Phd[1], Mahmut KOPARAL,DDS,Phd[1], Aydin KESKINRUZGAR,DDS,Phd[1], Şeyma BAYAZIT,Dr [1]

[1]Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Adiyaman University, Adiyaman, Turkey

Abstract

Introduction

Neurofibromatosis is a group of autosomal dominant genetic disorders characterized by multiple cutaneous lesions and tumors of the nervous system. NF is categorized into two, NF type-1 and NF type-2. Neurofibromatosis-1 is a neurocutaneous-skeletal disorder with variable phenotypic description. Craniofacial skeletal symptoms of the disease are observed in the jaw deformations and impacted, missing and displaced teeth can also be seen. The purpose of this report is to present a case of NF1.

Case report

A 23-year-old male patient was referred to the Department of Maxillofacial Surgery, Faculty of Dentistry, Adiyaman University due to abscess of molar teeth. As a result of the panoramic radiography taken from the patient, the second and third molar teeth were impacted in the left mandibula.

Conclusion

In neurofibromatosis, bone and soft tissue involvement can be seen in the maxillofacial region. They are also lesions that can cause the impacted teeth.

Keywords: Neurofibromatosis, maxillofacial region, autosomal dominant.

Introduction

Neurofibramatosis is an autosomal dominant disease.[1] The term neurofibromatosis (NF) is used for a genetic disorders that affect cell growth of neural tissues. The most common are 2 NF forms: Neurofibromatosis type 1 (NF-1) and Neurofibromatosis type 2 (NF-2). [1,2]

Neurofibromatosis type 1 (NF-1), also known as Von Recklinghausen disease.[2] NF-1 is the most common type of NF and accounts for approximately 90% of all cases, with one occurrence at 3000 births.1-3 There is a mutation in the 17q11.2 chromosome known as the NF-1 gene.[4] Neurofibromatosis 2 (NF-2) results from mutations in chromosome 22 of the NF-2 gene.[5] 50% of NF-1 patients have a family history of the disease.[6]

The most common symptoms of NF-1 in the skin are cutaneous neurofibromas and cafe au lait points.[5] The cafe-au-lait spots are clinically yellow-brown spots. The presence of at least 6 diameters greater than 1.5 cm in diameter is pathognomonic for NF-1.[7]

Superficial nodular lesions resulting from the proliferation of neural tissues in the skin are called neurofibroma. It is especially localized in the head and neck region. Due to of their size, nodules may cause problems with aesthetics and function.[8] In this study, we present the findings of oral and maxillofacial areas in a patient diagnosed with NF-1.

Case Report

A 23-year-old male patient who was diagnosed as NF-1 by the dermatologist applied to the our clinic due to swelling and infection on the left side of the mandible. The patient was contacted with help from his family because of the difficulty of sensing and speaking.

In the extraoral examination, facial asymmetry was observed. Due to the mass in the left posterior region of the mandible, bone contact could not be palpated in the left mandibula. Left lateral canthus of the left eye and sagging in the left ear, as well as hypertrophy in the left ear were observed. There were cafe au lait spots on the neck. (Figure 1 and Figure 2)

In intraoral examination, revealed growth on the left side of the tongue. There were dilated tongue papillae on the related side. In the mandible anterior teeth region and left posterior region,

Corresponding Author: Aydin Keskinruzgar
Associate Professor
Address: Adiyaman University, Faculty of Dentistry, Oral and Maxillofacial Surgery, Adıyaman
Mobile: +90(416).2251920
e-mail: akeskinruzgar@gmail.com

normal colour, non-ulcerated, painless were observed gingival hyperplasia. The patient's oral hygiene was deficient. In the area of the patient's complaint, deep caries were detected teeth 33 and 34, also there was infection due to.

The patient's panoramic radiography revealed radiolucent lesion with together 2 and 3 molar teeth impacted in the left mandibular posterior region. And lesion appeared to infiltrative growth pattern with no prominent borders. Foramen mandible and foramen mentale had expansion. (Figure 3)

The patient was prescribed antibiotics due to infection. Then, under general anesthesia, teeth 16, 26, 33, 34, 46 and 47 were taken. Impacted teeth 37 and 38 were taken with surgical intervention. The lesion was excluded.

Figure1: NF-1 extraoral image

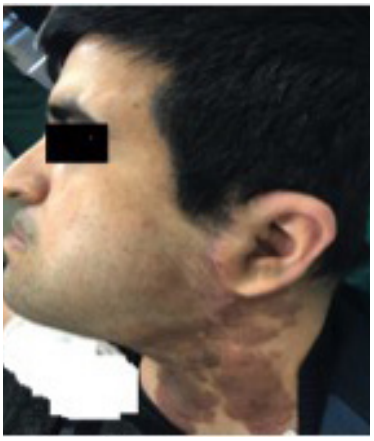


Figure 2: Cafe au lait spots on the neck

Figure 3: Left mandible affected by NF-1

Discussion

NF1 patients may exhibit hypoplasia or hyperplasia of the jaws and facial changes caused by the buccal bone and temporomandibular joint. Osseous changes can be in the bone or a product of soft tissues that grow against the bone.[6] In our case, due to the pressure on the surface of the NF bone, deformation of the bone was observed in the left mandible. Localized neurofibroma is the most common type of neurofibroma in NF-1 patients. The most frequently affected areas in the head and neck region are the scalp, cheek, neck and oral cavity.[2]

Oral findings can be found in almost 72% of NF1 patients.[6] Neurofibromas occurring in craniofacial structures can spread into the mouth, causing abnormalities that will prevent the function of oral structures, disrupting the teeth series and causing the teeth to be positioned incorrectly.[9] In our patient, NF-1 caused teeth 37 and 38 to be impacted. In particular, the expansion of the alveolar duct, foramen mandible and foramen mentality in the mandible is considered to be an oral manifestation of NF1.[3] Periodontal localizations may cause mobility in the teeth and hyperplasia in the gums. The most common oral manifestation of NF-1 is dilation of tongue fungiform papillae that occurs in about 50% of cases.[6] Dilation of tongue papillae was observed in our case report in accordance with the literature.

NF-1 patients may have abnormal speech rate, hoarseness and / or hypernasal speech.[5] In our case, mental retardation and perception deficits were observed in the patient.

Conclusion

NF-1 may show symptoms in the hard and soft tissue in the maxillofacial region. Diagnosis and follow-up of the disease is important for the patient's life functions.

References

1. Akoğlu G, Gazyağcı AS, Kutluer İ. Neurofibromatosis: Evaluation of Clinical Features of 11 Cases. Turk J Dermotol 2013;7:196-200.
2. Lee L, Yan YH, Pharoah MJ. Radiographic features of the mandible in neurofibromatosis A report of 10 cases and review of the literature. Surg Oral Med Oral Radiol Endod 1996;81:361-7.
3. Visnapuu V, Peltonen S, Tommissalo T, , Pentonen J, Happonen RP. Radiographic Findings in the Jaws of Patients With Neurofibromatosis. J Oral Maxillofac Surg 2012; 70:1351-1357.
4. Visnapuu V, Pienihäkkinen K, Peltonen S, Happonen RP, Peltone J. Neurofibromatosis 1 and dental caries. Clin Oral Invest 2011;15:119-121.
5. Visnapuu V , Peltonen S , Alivuotila L , Happonen RP, Peltonen J. Craniofacial and oral alterations in patients with Neurofibromatosis 1. Orphanet J Rare Dis. 2018; 13: 131.
6. Cunha K G, Barboza E P, Dias E P, Oliveira F M. Neurofibromatosis type I with periodontal manifestion. A case report and literature review. Br Dent J 2004;24;196(8):457-60.
7. Ucok C, Tuncer N, Günhan O. Neurofibromatosis (von reclinghausen's disease) a case report. Journal of Istanbul University, Fakulty of Dentistry 2012;31.76-79.
8. Çalık Yılmaz BC, Alaçam A. A Pathology mimicking dental abscess: facial plexiform neurofibroma. Acta Odontol Turc 2019;36(1):28-32.
9. Ayhan Bani A, Türköz Ç. Syndromes And Accompanying Craniofacial Anomalies. A.Ü.Dis.Hek. Fak. Derg. 2012; 39(1):35-47.

EurAsian Journal of Oral and Maxillofacial Surgery
Official publication of AÇBİD (Association of Oral and Maxillofacial Surgery Society)

CASE REPORT

Alveolar Distraction Osteogenesis in Wide Alveolar Cleft Patients

Yavuz FINDIK[1], Timuçin BAYKUL[1], Mehmet Fatih ŞENTÜRK[1], Tayfun YAZICI[1], Betül KIRAN[1]

[1]Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Süleyman Demirel University, Isparta, Turkey

Abstract

Introduction

Secondary alveolar cleft repair is commonly accepted for alveolar cleft patients, however, nowadays, controversy remains regarding the surgical technique, the timing of the surgery, and the donor site. Rehabilitation of the large alveolar clefts with autogenous graft or distraction osteogenesis is one of the most common treatment choices. The purpose of the report is to evaluate the surgical techniques for repairing the wide alveolar clefts.

Cases

Four patients with alveolar clefts were included in our case series. The width of the cleft was between 17 to 25 mm. All patients were treated with distraction osteogenesis. The segmental osteotomy was performed under general anesthesia. Distraction was started 5 days after surgery. All distractors were bone fixed but supported by a palatal arch for guiding the distraction. Dental cast models were used before the surgery. Pre and postoperative radiological examinations were performed through orthopantomogram and computed tomography (CT) scan.

Results

With distraction techniques, closure of the alveolar cleft was achieved. The desired movements with new bone formation were attained yet eventful in all cases.

Conclusion

The method of treatments described here is a prospect for treating extremely wide alveolar clefts. Further patients are needed to assess all effects, side effects, risks, and overall benefits of these techniques.

Keywords: alveolar cleft, distraction osteogenesis, wide cleft

Introduction

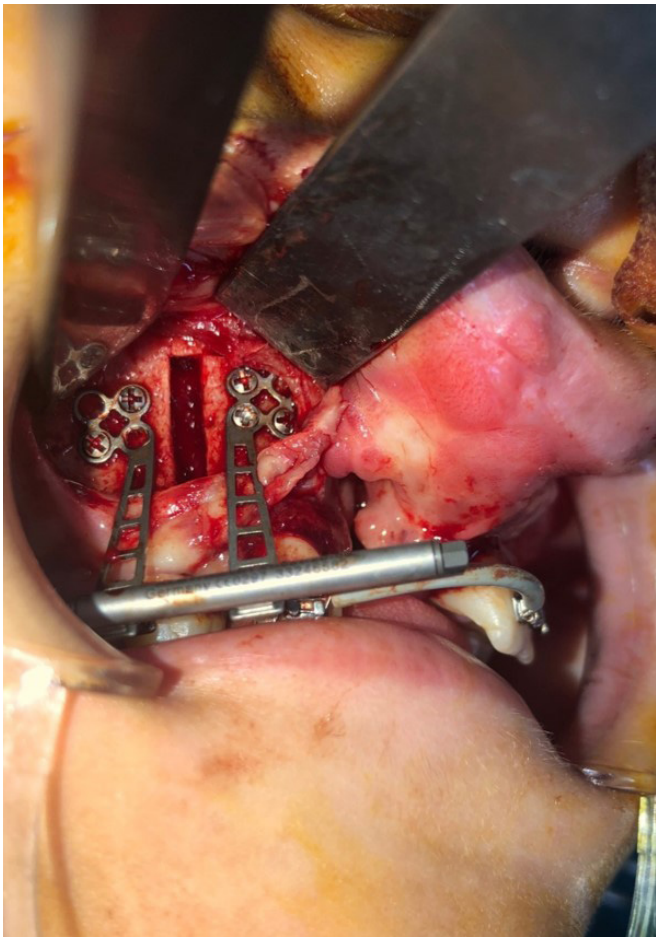
Rehabilitation of alveolar cleft is evolving era compared to other aspects of cleft lip and palate repair. Secondary alveolar bone grafting is the contemporary treatment of alveolar cleft and perialveolar fistula. Autogenous bone grafts from the iliac crest, proximal tibia or mandibular symphysis of the patient or synthetic grafts may be used for grafting.[1,2] In wide clefts failure rate is very high owing to the deficit of soft tissue in the cleft region. Besides, soft tissues affected by chronic inflamed oronasal fistulas may also increase failure rate. However, some cases are found unsuitable for secondary bone grafting. These are generally one of two types of deformities or their combinations: Vertical discrepancies across the cleft region and severe wide clefts. Both situations indicate a severe maxillary deficiency and tend to occur together though one may dominate the clinical picture.[2]

There is no classification for the treatment of ungraftable wide alveolar clefts in the literature. We classified wide cleft treatments in three methods.

1. Distraction osteogenesis

A unique method for new bone generation and soft tissue lengthening, which enables clinician to repair both soft and hard tissues at the same time with the patient's own tissues.[3,4] (Figure 1) Figure 1: intraoral view of the distractor

Corresponding Author: Betül Kıran
Research Assistant
Address: Süleyman Demirel University, Department of Oral and Maxillofacial Surgery, Çünür Mah. Merkez, Isparta
Mobile: +90(534).3597258
e-mail: betulkiran.294@gmail.com



2. Unilateral LeFort 1 osteotomy with elastic traction
In this technique, LeFort 1 osteotomy is performed unilaterally and osteotomized segment is moved with elastics to desired position in vertical discrepancies.

3. Free osteomuscle flap transfer
For wide ungraftable alveolar clefts, free flaps can be used, including bone, muscle and vascular support.

The aim of this case series was to present four patients with ungraftable wide alveolar cleft treated by distraction osteogenesis.

Cases and methods
The surgical procedures of these cases were designed by simulations performed on CT based 3D models and dental casts. Before the surgery, osteotomies were made and distractors was adjusted on the 3D models. All the patients were operated under general anesthesia and nasotracheal intubation.

Patient 1
Unilateral cleft with 17 mm bony defect. At the cleft side, a two teeth bearing segment was freed by interdental and horizontal osteotomies. KLS Martin Liou cleft distractor device was performed for bone-borne distraction. (Figure 2)
The segment was mesially transported and the cleft area was closed. (Figure 3) Alveolar grafting and removal of distractor were made in the same session.

Figure 2 : postoperative radiograph of the patient

Figure 3: panoramic radiograph after the distraction phase

Patient 2
Unilateral cleft including middle line was 25 mm. Because of economic reasons, custom made hybrid type distractor was performed. The cleft was closed desired size. Synthetic alveolar grafting will be going to while the removal of distractor. (Figure 4)

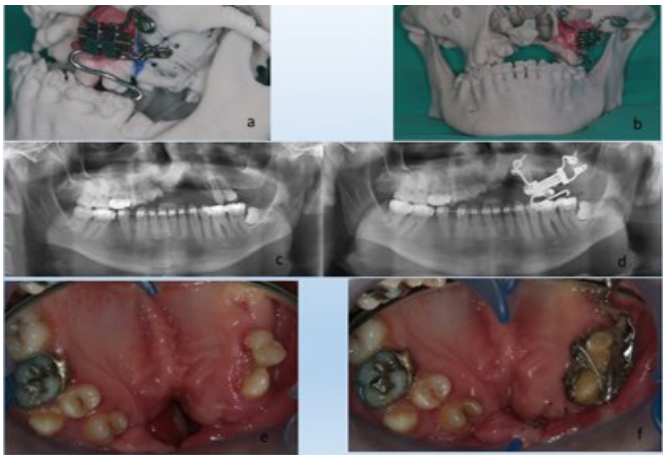


Figure 4: Patient's 3D casts, pre and post-operative radiographs and intraoral images

Patient 3

Unilateral cleft with 22 mm bony defect. At the cleft side, a one tooth bearing segment was freed by interdental and horizontal osteotomies. KLS Martin Liou distractor device was performed. The segment was mesially transported for 13 mm. (Figure 5) Distraction removal and alveolar grafting was performed in the same session.

Figure 5: Panoramic radiograph after the distraction phase

Patient 4

Unilateral cleft with 21 mm bony defect. They were crowding of teeth, therefore maxilla was expanded before the application of distractor and extraction of bad prognosis teeth was made. Interdental and horizontal osteotomies were made and KLS Martin Liou distractor device was performed. The segment was mesially transported for 13 mm. Alveolar grafting will be going to while the removal of distractor after consolidation period.

Results

The healing phase was uneventful and the activation period was completed in all patients. During the distraction, there was no device loosening issues and the segments was stable. No postoperative complication occured in the patients. The treatment of cases continues.

Discussion

Surgical closure of wide alveolar clefts is important not only in the improvement of articulation, establishment of functional occlusion, and aesthetic improvement, but also in the improvement of the oral environment. Secondary autogenous bone grafting for alveolar clefts provides continuity to the alveolar arch by closure of the cleft. Though this method is useful, failure of the bone graft can occur when the cleft is markedly large, or when covering with an oral mucoperiosteal flap is inadequate due to marked scar formation.[5]

Distraction osteogenesis is considered as the indicated approach for wide alveolar cleft patient, which can minimize risks of soft tissue breakdown and secondary bone graft failure. However, DO is a complicated procedure, particularly for alveolar bone, which has a limited place to set the distraction. It is important to design suitable distractor to make operations easier and to reduce morbidity.[6] Intraoral devices work better because they are securely fixed to adjacent bone or teeth, and the strength is easily controlled.[7] Liou et al. completed approximation of a wide alveolar cleft by creating a segment of new alveolar bone and attached gingiva with a tooth-borne intraoral distraction device.[8]

Many advantages have been reported for the distraction of alveolar bone to close large cleft gaps.[8,9,10] It is possible to decrease the size of the gap to a minimum which can be easily repaired. Even though the improvement of the bony cleft is limited, the soft tissue cleft is closed completely and enough attached gingiva is formed to cover the bone graft. Other advantages are that there is no need for a donor site and donor site surgery. Also can be avoided unpredictable resorption of free bone graft. Very large gaps might be closed successfully including new generated bone which can be used to move teeth into. The risk of relapse is less and the recovery time is shorter.[11,12]

There are many distractors on the market, such as bone borne, tooth-borne and hybrid types. The majority of the distractors on the market are bone borne devices so they perform the distraction through a straight line. This requires a secondary orthodontic treatment approach to create symmetric and ovoid arch form. Using a tooth-borne distractor is also advantageous because it makes the protocol simpler by avoiding the surgery necessary for removal of the distractor.[11]

Ding et al.14 studied changes in periodontal tissue during maxillary dentoalveolar distraction osteogenesis using an intraoral tooth-borne distractor to close wide alveolar defects in four dogs. They found that the morphological changes in the periodontal tissues of the supporting tooth were moderate. They could be reversed if the rate and duration of distraction were correct like the physiological changes of the periodontal ligament of the orthodontic tooth. Liou et al. recommend moving the teeth into the new generated bone as soon as possible to avoid shrinkage during maturation.[8]

Due to the abovementioned reasons, distraction osteogenesis remains as an exceptional treatment method generating new bone and following soft tissue utilizing the patients' own tissues. This is a remarkable advantage in cases with large cleft gaps to be closed or at least to diminish the size of the gap to a favourable dimension where further repair is easily feasible. The newly generated bone can also be used to move teeth into when needed.[13]

Conclusion

Recalcitrant alveolar clefts in permanent dentition are faced in all cleft centers.

If they are labeled as ungraftable, transport bone formation through distraction or the other techniques make these clefts amenable to grafting.

In conclusion, treatment of ungraftable alveolar clefts await more innovation.

References

1. Wang XX, Wang X, Li ZL, Yi B, Liang C, Jia YL, Zou BS. Anterior maxillary segmental distraction for correction of maxillary hypoplasia and dental crowding in cleft palate patients: a preliminary report. Int J Oral MaxillofacSurg 2009;38:1237-43.
2. Zemann W, Pichelmayer M. Maxillary segmental distraction in children with unilateral clefts of lip, palate, and alveolus. Oral Surg Oral Med Oral Pathol Oral RadiolEndod. 2011 Jun;111(6):688-92.
3. Ilizarov GA, Lediov VL, Shitin VP. The course of compact bone reparative regeneration in distraction osteosynthesis under different conditions of bone fragment fixation and experimental study. Eksp Khir Anesteziol1969;14:3–12. (in Russian).
4. Polley JW, Figueroa AA. Management of severe maxillary deficiency in childhood and adolescence through distraction osteogenesis with an external adjustable rigid distraction device. J Craniofac Surg 1997; 8:181.
5. Posnick JC, Tompson B. Cleft-orthognathic surgery: complications and long-term results. Plast Reconstr Surg 1997;96: 255.
6. Segmental Maxillary Distraction Osteogenesis With a Hybrid-Type Distractor in the Management of Wide Alveolar Cleft Jianfei Zhang, PhD(c), Wenbin Zhang, PhD, DDS, Steve Guofang Shen, DDS, MD, FICD First Published March 23, 2018
7. Picard, A, Diner, PA, Galliani, E, Tomat, C, Vazquez, Mr, Carls, FP. Five years experience with a new intraoral maxillary distraction device (RID). Br J Oral Maxillofac Surg. 2011;49(7):546–551.
8. Liou, EJ, Chen, PK, Huang, CS, Chen, YR. Interdental distraction osteogenesis and rapid orthodontic tooth movement: a novel approach to approximate a wide alveolar cleft or bony defect. Plast Reconstr Surg. 2000;105(4):1262–1272.
9. Liou EJW, Chen PKT. Intraoral distraction of segmental osteotomies and miniscrews in management of alveolar cleft. Semin Orthod 2009;15:257–67.
10. Liou EJ, Chen PKT. Management of maxillary deformities in growing cleft patients. In: Berkowitz S, editor. Cleft Lip and Palate: Diagnosis and Management. Berlin Heidelberg, Germany: Springer-Verlag; 2006.
11. Erverdi N, Küçükkeleş N, Şener C, Selamet BU. Interdental distraction osteogenesis for the management of alveolar clefts: archwise distraction. Int J Oral Maxillofac Surg. 2012 Jan;41(1):37-41.
12. L. K. Cheung, Q. Zhang, Z. G. Zhang, and M. C. M. Wong, "Reconstruction of maxillectomy defect by transport distraction osteogenesis," Int J Oral Maxillofac Surg, vol. 32, no. 5, pp. 515–522, 2003.
13. N Shahab, DDS, MSc, C Sar, DDS, PhD, M Sarac, DDS, PhD, N Erverdi, DDS, PhD Reconstruction of Premaxilla With Alveolar Distraction Osteogenesis in a Patient With Complete Cleft Lip and Palate: A Case Report. The Cleft Palate-Craniofacial Journal 1-4, 2018.
14. Ding Y, Liu Y, Caob M, Maa Q, Zhoua H, Liu B. Periodontal tissues changes in toothborne distraction osteogenesis: an experimental study of closure of wide alveolar bone defects in dogs. Br J Oral Maxillofac Surg 2009;47:111–5.

EurAsian Journal of Oral and Maxillofacial Surgery
Official publication of AÇBİD (Association of Oral and Maxillofacial Surgery Society)

CASE REPORT

Pleomorphic Adenoma Localized in Buccal Mucosa: Report a Case

Emine ÖRNEK AKDOĞAN, DDS[1], Ferhat AYRANCI, DDS, PhD[1], Kadircan KAHVECİ, DDS[1], Tolunay AVCI, DDS[1]

[1]Ordu University, Faculty of Dentistry, Oral and Maxillofacial Surgery, Ordu

Abstract

Objective

Pleomorphic adenoma is the most common salivary gland tumor with a benign, mixed character. Majority of pleomorphic adenomas originated from minor salivary glands seen in the hard palate. However, rarely it may originate from the other minor salivary glands in buccal mucosa and the tongue. In this case report, excision of the pleomorphic adenoma of the buccal minor salivary gland is presented.

Case

Clinical examination of a 55-year-old female patient who referred to our clinic with swelling in the right buccal region, showed a solid mass in the buccal mucosa and total excision of the mass was performed with a preliminary diagnosis of lipoma. Excisional biopsy revealed the diagnosis as pleomorphic adenoma.

Conclusion

Pleomorphic adenoma originated from the minor salivary glands cause confusions regarding diagnosis with other lesions. Therefore, reaching the definite diagnosis can only be possible by confirming the clinical and radiological examinations with histological examination.

Keywords: Lipoma, minor salivary gland, excision

Introduction

Salivary gland neoplasms are relatively rare and often affect the major glands[1]. Approximately two-thirds of salivary gland tumors that comprise less than 5% of the tumors occurring in the head and neck region, are pleomorphic adenomas (PA)[2]. PA is the most common salivary gland tumor which has benign mixed nature. Only 8% of PAs originate from minor salivary glands and are commonly seen in the hard palate[2]. Less frequently, PA may also originate from the minor salivary glands in the buccal mucosa tongue, lip, and extra-salivary tissues[2, 3]. The PAs of the minor salivary glands are usually cause painless, non-symptomatic submucosal mass approximately 2 to 6 cm in size[2, 4]. PAs are more common in women in the third and fifth decades[4-6]. The accepted treatment protocol of PA is surgical excision[2, 7]. The aim of the surgery is to remove the tumor completely with its capsule to minimize the risk of recurrence[3].

In this case report, a rare case of PA originating from the minor salivary glands of buccal mucosa, is presented.

Case Report

A 55-year-old female patient was admitted to our clinic with a complaint of swelling in the right buccal region. Clinical and radiological examinations revealed a non-invasive, solid mass in the right upper maxillary premolar region near the cheek. Following the clinical and radiological examinations, total excision of the mass was planned with a preliminary diagnosis of lipoma.

After administration of local anesthesia, a horizontal incision was made and the mass was reached by raising a half-thickness flap. Then the mass was removed with blunt dissection from the buccal mucosa (Figure 1). After primary closure was performed, the patient was prescribed antibiotics, analgesics and antimicrobial mouthwashes.

Excised mass was sent for histopathological examination (Figure 2). The histopathology report revealed the diagnosis as PA (Figure 3). One week after the operation, sutures were removed and a dressing was performed at the operation site. The healing was uneventful. No complication was observed at 4 months follow-up.

Corresponding Author: Emine Örnek Akdoğan
Address: Ordu University, Faculty of Dentistry, Cumhuriyet Mah. Mustafa Kemal Bulvarı No: 254, Ordu
Mobile: +90(553).6761032
E-mail: dt.emine93@gmail.com

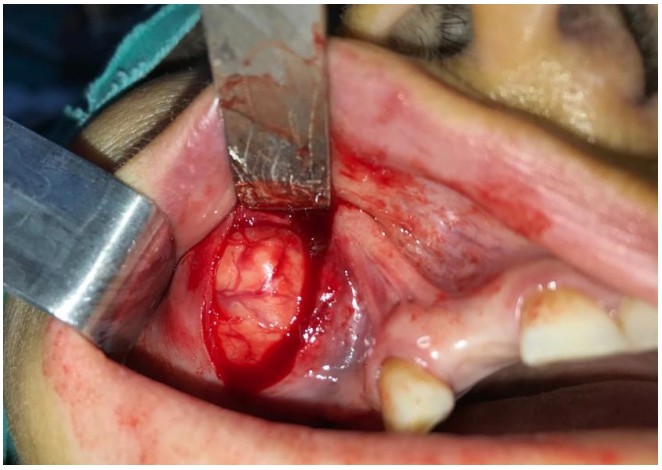


Figure 1: Intraoperative view of removing the mass with blunt dissection

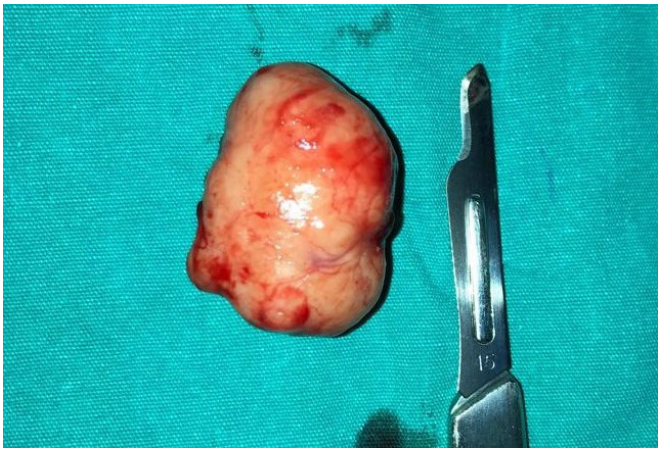


Figure 2: Excised specimen

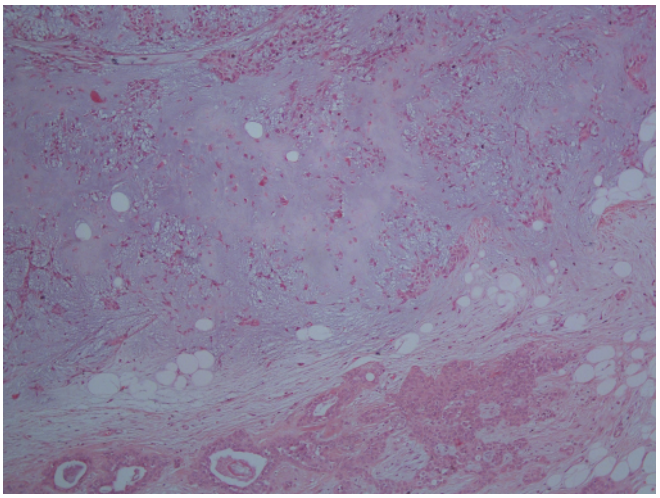


Figure 3: Histopathological view of PA

Discussion

PA is a benign, mixed salivary gland neoplasm originating mostly from the major salivary glands. 63% of PAs are seen in the parotid gland, 10% in the submandibular gland and 0.1% in the sublingual gland[3]. Minor salivary gland tumors are rare and constitute 9-23% of all salivary gland tumors[1]. PA is most commonly originating from the minor salivary glands of the palate however, it may also originate from the minor salivary glands in the areas such as lip, tongue, floor of the mouth, buccal mucosa, sinuses, epiglottis and trachea[3, 6, 7].

Nardone et al.8 presented a case of PA originating from minor salivary glands in the external ear canal, nose and throat. Unlike the major salivary gland PA, minor salivary gland PA is not encapsulated by connective tissue[2]. In contrast to what is generally reported, in our case PA was originated from the minor salivary glands of the buccal mucosa and was also encapsulated with connective tissue.

The clinical features of PA usually a gradually increasing painless swelling. Pain is not a common symptom, but local discomfort is the main problem for patients. Even though these lesions reach a very large size, they rarely develop ulcers on the surface of the lesion[2, 4]. Chaturvedi et al.7 reported a PA with an unusual size of 3 cm x 4 cm in the palatal region, which cause difficulties in chewing, swallowing and speech. Also, in this report ulceration and bleeding have been reported due to persistent friction on the mucosa on the swelling. In our case, only a painless swelling was present.

Malignant transformation of PA can be seen and called carcinoma ex pleomorphic adenoma[9]. Thus, during the excision of the PA, care must be taken not to disrupt the continuity of the capsule of PA. The rupture of the capsule and incomplete excision of microscopic pseudopod-like extensions may result in recurrence of PA[3, 5]. Spiro reported recurrence in 7% of and 6% of 1342 patients with benign parotid gland neoplasia and benign minor salivary gland tumors, respectively[4, 10]. The ideal treatment protocol of PA is total excision with safety margins[4, 7].

Conclusion

As a conclusion, PA can be found in the buccal mucosa and must be considered in the differential diagnosis of the lesions that affecting this area. In addition to clinical and radiological examination, histological examination is also required for definitive diagnosis.

References

1. Pires FR, Pringle GA, de Almeida OP, Chen SY. Intra-oral minor salivary gland tumors: a clinicopathological study of 546 cases. Oral Oncol 2007; 43: 463-470.
2. Khandekar S, Dive A, Munde P, Wankhede ND. Pleomorphic adenoma of the buccal salivary gland. J Oral Maxillofac Pathol 2015; 19: 111.
3. Lingam RK, Daghir AA, Nigar E, Abbas SA, Kumar M. Pleomorphic adenoma (benign mixed tumour) of the salivary glands: its diverse clinical, radiological, and histopathological presentation. Br J Oral Maxillofac Surg 2011; 49: 14-20.
4. Debnath SC, Saikia AK, Debnath A. Pleomorphic adenoma of the palate. J Maxillofac Oral Surg 2010; 9: 420-423.
5. Sharma N. Pleomorphic adenoma of the buccal salivary gland: magnetic resonance imaging findings with differential diagnoses. J Investig Clin Dent 2012; 3: 228-231.
6. Ramesh M, Krishnan R, Paul G. Intraoral minor salivary gland tumours: a retrospective study from a dental and maxillofacial surgery centre in salem, Tamil Nadu. J Maxillofac Oral Surg 2014; 13: 104-108.
7. Chaturvedi M, Jaidev A, Thaddanee R, Khilnani AK. Large Pleomorphic Adenoma of Hard Palate. Ann Maxillofac Surg 2018; 8: 124-126.
8. Nardone M, Ferrara G, Nappi O, Di Maria D, Villari G. [Pleomorphic adenoma in unusual anatomic sites: case reports and review of literature]. Acta Otorhinolaryngol Ital 2002; 22: 158-163.
9. Goyal P, Sehgal S, Ghosh S, Agrawal D, Singh S. Rare Carcinoma Ex-pleomorphic Adenoma of Buccal Mucosa: Case Report and Review of Literature. Rare Tumors 2016; 8: 6138.
10. Spiro RH. Salivary neoplasms: overview of a 35-year experience with 2,807 patients. Head Neck Surg 1986; 8: 177-184.