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

Jan 2025 Volume 4 Number 1
e-ISSN: 2687-5497

RESEARCH ARTICLES

CASE REPORTS

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RESEARCH ARTICLE

Biomechanical Stress Analysis in Retrodiscal Tissues of Temporomandibular Joint with Unilateral Disc Displacement Without Reduction: A Finite Element Study

Tek Taraflı Temporomandibular Eklem Redüksüyonsuz Disk Deplasmanında Retrodiskal Dokulara Gelen Stresin Biyomekanik Analizi: Sonlu Elemanlar Çalışması

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ABSTRACT

Objective: This study aimed to evaluate stress changes in retrodiscal tissues during mandibular movements in temporomandibular joints (TMJ) with unilateral disc displacement without reduction (DDwoR) using finite element analysis (FEA).

Materials and Methods: Geometric models were created using CT scan data from two patients: one with bilaterally normal disc positioning and another with DDwoR. DICOM files were segmented, and 3D models were reconstructed and converted into mathematical models following a standardized methodology. Two models were analyzed: a control model with bilaterally normal discs and a DDwoR model, with separate evaluations of the left (normal) and right (DDwoR) joints. TMJ movements during mouth opening and closing were simulated, and stress distribution patterns in the retrodiscal tissues were analyzed. Von Mises stress values were measured and compared between the DDwoR and healthy sides.

Results: Both the normal and DDwoR sides demonstrated altered stress patterns compared to the healthy control. The DDwoR side consistently exhibited elevated stress in the superior region during mouth opening, underscoring its mechanical vulnerability. The normal side, while less affected than the DDwoR side, displayed compensatory stress increases, particularly in the intermediate region during mouth opening.

Conclusion: These findings underscore the vulnerability of the retrodiscal tissues, especially the superior region, to mechanical stress in the presence of DDwoR. This study highlights the importance of clinical strategies aimed at preserving retrodiscal tissue integrity to prevent progressive joint damage.

Keywords: Disc displacement without reduction, finite element analysis, retrodiscal tissue, stress distribution, temporomandibular joint

ÖZET

Amaç: Bu çalışmanın amacı, unilateral redüksiyonsuz disk deplasmanı (DDwoR) bulunan temporomandibular eklemlerde (TME) mandibular hareketler sırasında retrodiskal dokulardaki stres değişimlerini sonlu elemanlar analizi (FEA) kullanarak değerlendirmektir.

Gereç ve Yöntemler: Geometrik modeller, biri bilateral olarak normal disk pozisyonuna sahip, diğeri ise unilateral DDwoRlu iki hastanın BT (Bilgisayarlı Tomografi) tarama verileri kullanılarak oluşturulmuştur. DICOM dosyaları segmentlere ayrılmış, 3D modeller yeniden yapılandırılmış ve standart bir metodoloji izlenerek matematiksel modellere dönüştürülmüştür. İki model analiz edilmiştir: bilateral olarak normal disk pozisyonuna sahip kontrol modeli ve unilateral DDwoR modeli. Bu modellerde sol (normal) ve sağ (DDwoR) eklemler ayrı ayrı değerlendirilmiştir. Ağız açma ve kapama hareketleri simüle edilerek retrodiskal dokulardaki stres dağılımı analiz edilmiştir. Von Mises stres değerleri ölçülerek DDwoR tarafı ile sağlıklı taraf karşılaştırılmıştır.

Bulgular: Hem normal hem de DDwoR tarafları, sağlıklı kontrole kıyasla değişen stres paternleri göstermiştir. DDwoR tarafı, ağız açma sırasında üst bölgede artmış stres seviyeleri sergileyerek mekanik açıdan daha hassas olduğunu ortaya koymuştur. Normal taraf, DDwoR tarafına kıyasla daha az etkilenmiş olsa da özellikle ağız açma sırasında orta bölgede telafi edici stres artışları göstermiştir.

Sonuç: Bu bulgular, DDwoR varlığında özellikle retrodiskal dokuların üst bölgesinin mekanik strese karşı savunmasız olduğunu vurgulamaktadır. Çalışma, ilerleyici eklem hasarını önlemek için retrodiskal doku bütünlüğünün korunmasına yönelik klinik stratejilerin önemini ortaya koymaktadır.

Anahtar Kelimeler: Redüksiyonsuz disk deplasmanı, retrodiskal doku, sonlu eleman analizi, stres, temporomandibular eklem

Submission Date: January 4, 2025
Acceptance Date: January 17, 2025
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INTRODUCTION

The temporomandibular joint (TMJ) is one of the most complex joints in the human body, both morphologically and functionally.[1] Mandibular function relies on the synchronized activity of both TMJs, as their movements are inherently interconnected.[2] Temporomandibular disorders (TMD) refer to a group of conditions affecting the TMJ, the muscles of mastication, and associated structures. Among these, disc displacement without reduction (DDwoR) is one of the most frequently encountered internal derangements, characterized by the articular disc's permanent anterior or anteromedial displacement. This displacement disrupts the normal condyle-disc relationship, resulting in mechanical imbalance, joint dysfunction, and often pain.[3] Unilateral DD has been shown to alter chewing patterns compared to healthy controls.[4] It has been demonstrated that the stress distribution on TMJ structures changes following DD. However, no studies have been found that investigate how the loading on the healthy, non-displaced joint is affected.

The retrodiscal tissue, located posterior to the articular disc, serves as a highly vascularized and innervated structure that contributes to joint stability and function.[3,5] It is susceptible to mechanical stress and deformation, especially in cases of DDwoR where the displaced disc fails to return to its normal position, exposing the retrodiscal tissue to abnormal loading conditions.[6,7] Prolonged mechanical stress in this region may lead to inflammation, fibrosis, and structural damage.[8]

Finite Element Analysis (FEA) is a computational method that simplifies complex structures into discrete elements to evaluate their mechanical behavior. This approach, extensively utilized in fields like engineering and biomechanics, facilitates the simulation of stress and strain patterns under varying conditions. In TMJ research, FEA has been instrumental in providing insights into joint mechanics, particularly in estimating stress and strain distributions.[9]

Previous studies have not adequately addressed the retrodiscal tissue mechanics during mouth opening and closing, leaving a gap in understanding the differential stress distribution across various TMJ conditions. This study aims to evaluate the effects of unilateral TMJ DDwoR on the contralateral healthy TMJ retrodiscal tissues using the FEA method. The analysis specifically examines the retrodiscal tissues during both mouth opening and closing movements to provide a comprehensive understanding of the stress distribution in these conditions. The findings of this study are expected to guide clinical strategies for protecting retrodiscal tissues and preventing progressive joint damage.

MATERIALS AND METHODS

This research was conducted at the Department of Oral and Maxillofacial Surgery, Ondokuz Mayıs University, and the Ay Tasarım Ltd. Şti. Laboratory. Ethical approval for this study was obtained from the Ondokuz Mayıs University Clinical Research Ethics Committee (OMU KAEK) under decision number 2019/391.

Modeling Process: Geometric models for this study were created using CT scan data from two patients: one with bilaterally normal disc positioning and another with unilateral DDwoR. Informed consent forms were obtained from participants. Disc positions were verified through magnetic resonance imaging (MRI) conducted on the same individuals. The study protocol received approval from the clinical research ethics committee. DICOM (Digital Imaging and Communications in Medicine) files were processed with 3D Doctor software for segmentation. Model reconstruction and conversion into mathematical models followed a standardized methodology consistent with that employed in our prior research.[10] Two model was created:

Model 1: TMJ with bilaterally normal disc positioning (Healthy control)

Model 2: TMJ with unilateral DDwoR

In Model 2, the two TMJ sides were evaluated separately, including the left TMJ with a normal disc position and the right TMJ with DDwoR.

Software and Tools: Mimics Innovation Suite (Materialise, Belgium) was used for 3D modeling. ANSYS Workbench 2021 R2 (ANSYS Inc., USA) was utilized for mesh creation and finite element analysis. SolidWorks 2021 (Dassault Systèmes, France) was used for geometry adaptation and adjustments.

Material Properties: The material properties of the TMJ components were defined using data from the literature:

-Elastic modulus (Young's modulus) and Poisson's ratio for the retrodiscal tissue were adapted based on prior studies.[8,11]

-Retrodiscal tissues were modeled as isotropic, elastic, and hyperelastic structures to account for their flexible nature.

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Simulation Conditions: After performing the FEA, the maximum and minimum principle streses (MaxPS, MinPS) on condyle and Von Mises (VM) stresses on the disc were evaluated numerically and color-coded during mouth opening and closing.

In mouth closing, joint loading was simulated as a consequence of the resultant force vectors of the three jaw-closing muscles (masseter, temporalis, and medial pterygoid muscles). The insertion points of the muscles, the central point of the anterior teeth, and the rearmost point of occlusal contact were linked as a rigid body to the TMJ FE model. For each muscle, the line of action was defined by the points of insertion and origin. Their maximum forces had been approximated from their physiological cross-sectional areas according to previous studies.[12,13] The total of the resultant muscle force was established as 100N for each side.

In mouth opening, jaw opening muscles were simultaneously activated with the deactivation of the jaw-closing muscles. Mouth opening was simulated differently for each model. In Model 1, the maximum mouth opening is simulated as 45 mm on the straight axis. In the second model, the mouth opening was 35mm and simulated with deflection to the right side (disc displacement side). The amount of deflection was 5mm from the midline to the right side.

Boundary and Loading Conditions: Applied forces were modeled based on average muscle forces during chewing and occlusion, with a load of 200 N (Newton). Von Mises stress values were used to analyze equivalent principal stress (EPS) within the retrodiscal tissues.

Analysis Process: Linear elastic analysis was conducted to simulate stress and deformation. Stress concentrations in the superior, intermediate, and inferior regions of the retrodiscal tissue were evaluated.

RESULTS

The stress values were determined at three points on the retrodiscal tissue during mouth opening and closing (Figures 1 and 2). All von Mises equivalent stresses during mouth opening and closing were measured in megapascals (MPa). (Tables 1 and 2)

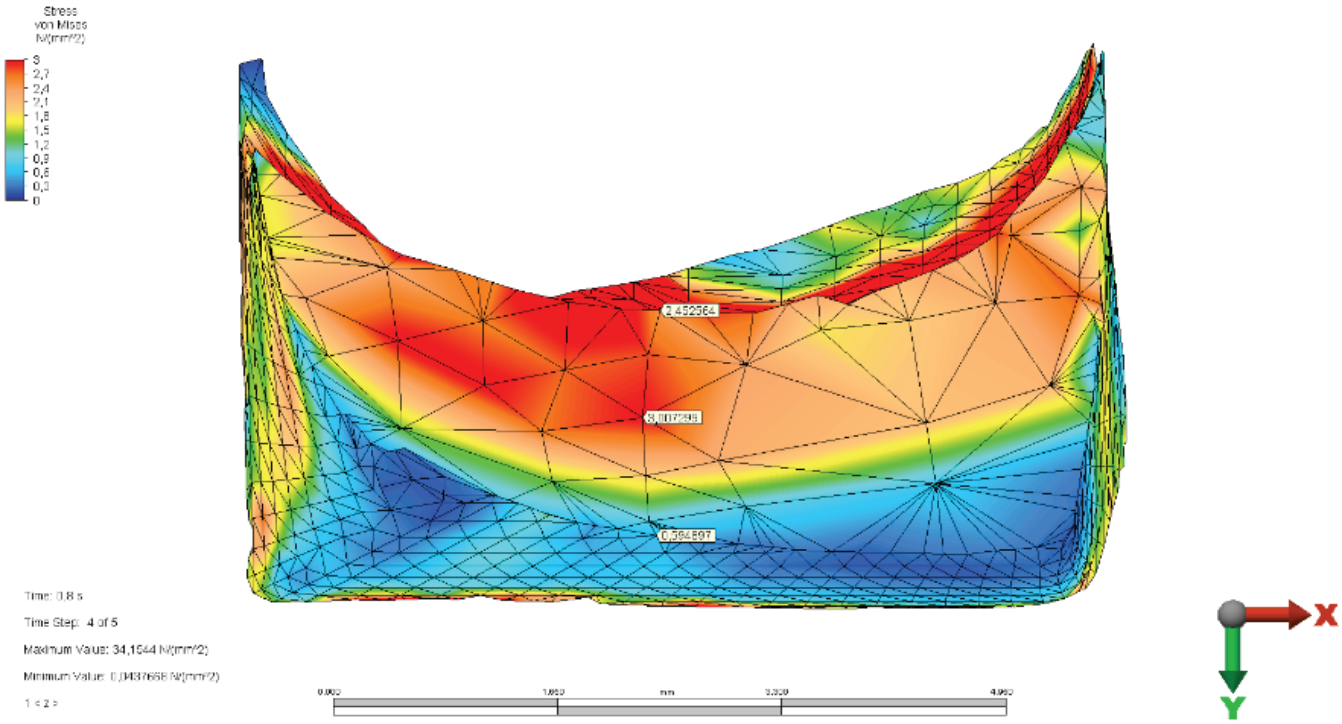

Figure 1. Stress distribution in retrodiscal tissue during mouth opening.

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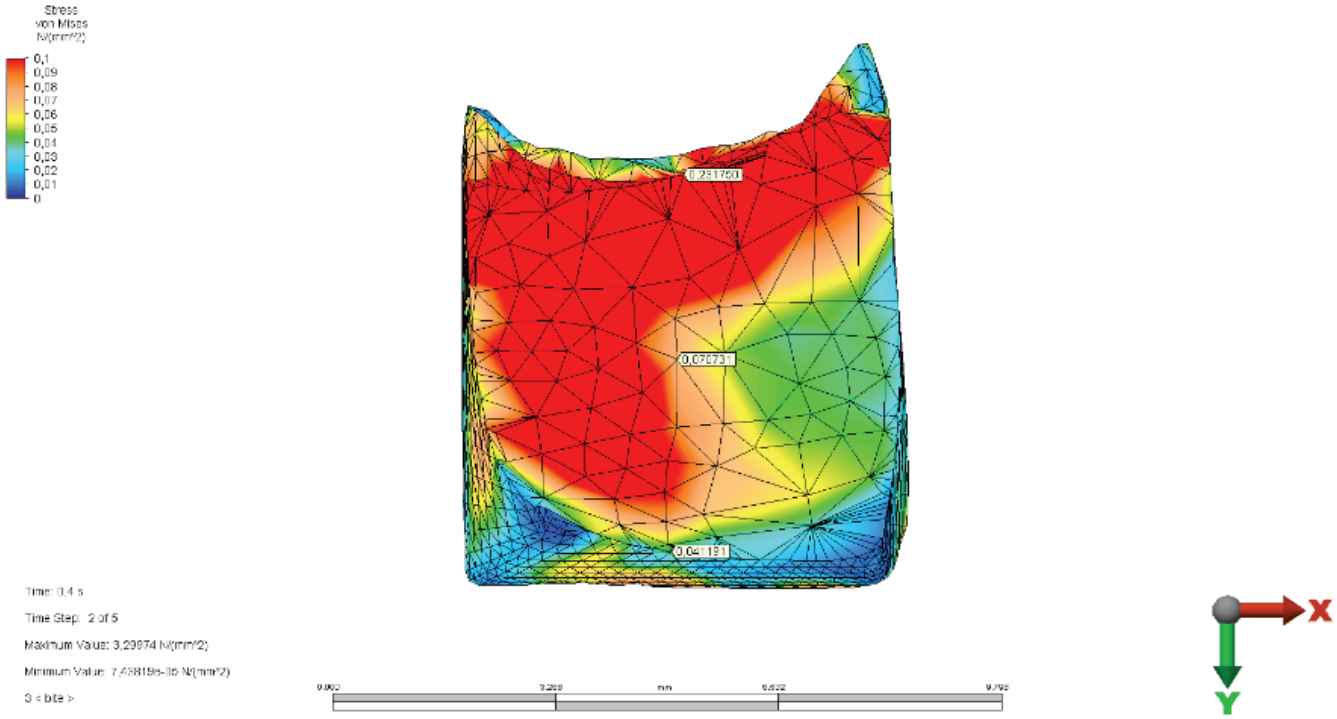

Figure 2. Stress distribution in retrodiscal tissue during mouth closure.

Table 1. Equivalent principal stress (von mises) values during mouth opening

Region	Normal Side (MPa)	DDwoR Side (MPa)	Healthy control
Superior	0.650918	1.010919	2,452564
Intermediate	0.950091	0.525255	3,007299
Inferior	0.217510	0.054029	0,594897

Table 2. Equivalent principal stress (von mises) values during mouth closing

Region	Normal Side (MPa)	DDwoR Side (MPa)	Healthy control
Superior	0.231750	0.051345	0,038216
Intermediate	0.070731	0.026345	0,018213
Inferior	0.041191	0.008709	0,005849

During mouth opening, the Von Mises stress distribution varied significantly between the superior, intermediate, and inferior regions of the retrodiscal tissue across the normal, DDwoR, and healthy control groups. The highest stress was observed in the intermediate region of the healthy control group (3.007 MPa), followed by the superior region (2.452 MPa). In the unilateral DDwoR model, the highest stress value was observed in the superior region on the DDwoR side (1.011 MPa), while the normal side exhibited the highest stress in the intermediate region (0.950 MPa).

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During mouth closing, the highest Von Mises stress was recorded in the superior region of the normal side (0.232 MPa) within the unilateral DDwoR model. This stress value was markedly higher than the DDwoR side (0.051 MPa) and the healthy control group (0.038 MPa). Additionally, the inferior and intermediate regions consistently showed lower stress levels across all groups, with the healthy control group exhibiting the lowest overall values.

DISCUSSION

The results of this study provide valuable insights into the biomechanical behavior of the TMJ under different conditions of disc positioning during mouth opening and closing. Studies investigating the biomechanical balance of load distribution within the TMJ reported that stress distributions in the TMJ with a normal disc position were substantially different from those with anterior disc displacement.[13-15] In the literature, while the TMJ condyle and disc are more frequently evaluated, there are only a limited number of studies assessing the forces applied to the retrodiscal tissues.[15] In our study, similarly, it was observed that the load distribution in the retrodiscal tissues changed in both the healthy side and the side with DD in the unilateral DDwoR model compared to the healthy control group. During mouth closing, compared to the healthy joint, the stresses on the retrodiscal region increased in both sides of the unilateral DD model, more prominently on the normal side, while they decreased during mouth opening. This result underscores the compensatory mechanisms that may arise in the normal TMJ to balance the mechanical deficiencies introduced by the DDwoR side. The increased stress in the superior region during mouth closure suggests that this area bears the primary load during occlusal contact and mastication.

In the literature, only one study has evaluated the effect of unilateral DD on the contralateral healthy joint.[14] Hattori-Hara et al.[14] created a unilateral DD model, applied clenching force, and assessed the stress on the contralateral healthy joint using the FEA method. They evaluated the stress on the condyle, disc, and retrodiscal tissue at different time intervals during clenching. A similar model was used in our study; however, instead of clenching movements, biting and mouth opening movements were simulated. Their study concluded that the presence of unilateral DD leads to increased stress in the retrodiscal tissues of the contralateral joint, which has a normally positioned disc. Our study produced similar results in retrodiscal tissue during the biting movement.

Mandibular movement is the result of the combined and simultaneous activities of both TMJs, as the right and left joints cannot function entirely independently.[2] The study by Kakimoto et al.[16], utilizing MRI, demonstrated that contralateral joints are qualitatively affected in patients with unilateral anterior DD. A study on patients with unilateral DD revealed that their chewing patterns differ from those of healthy controls.[4] However, the effects of these changes on joint structures remain unclear. The asymmetrical loading patterns observed in the unilateral DD model provide additional evidence for the biomechanical adaptations that occur in response to joint pathology. The presence of deflection in the DDwoR model may be one of the possible factor contribute to altered stress distributions. The reduced stress levels on the DDwoR side observed in our study could be attributed to the joint's limited mobility and disrupted functional dynamics, resulting in an uneven load-sharing mechanism between the affected and unaffected sides. Based on these findings, a clinical approach to managing patients with TMJ DDwoR should focus on strategies that minimize excessive mechanical loading during mandibular movements, particularly mouth opening. Therapies such as mandibular movement modification, guided exercise programs, and stabilization splints may help redistribute stress and protect retrodiscal tissues. Additionally, educating patients on avoiding wide mouth opening or excessive jaw movements could prevent further joint deterioration and alleviate symptoms.

During mouth opening, the highest Von Mises stress was observed in the healthy control model compared to both the DDwoR side and the normal side of the unilateral DD model. This result can be explained by the fact that in a healthy TMJ, the disc is properly positioned, allowing for the efficient transmission of muscular forces across the joint. This efficient force transmission leads to higher stress levels, particularly in the retrodiscal tissues that are actively engaged in stabilizing the joint during movement. Additionally, changes in muscle dynamics could contribute to this finding. The activation patterns of the jaw-opening muscles may differ between healthy and pathological joints. In a healthy TMJ, muscle forces are evenly distributed, resulting in higher stress concentrations. However, in DDwoR, the altered jaw movements, such as deflection or limited mouth opening, and the asymmetrical distribution of muscle forces, likely lead to lower stress levels on the affected side.

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While this study provides a detailed analysis of stress distribution in retrodiscal tissues, it is limited by its reliance on static modeling of mandibular movements. Dynamic modeling incorporating real-time muscle forces and additional factors, such as tissue viscoelasticity, may offer a more comprehensive understanding. Future research could explore whether the reduced stress on the DDwoR side correlates with increased susceptibility to joint degeneration or altered proprioceptive feedback. Studies also needed to explore the effects of therapeutic interventions, such as splints or surgical disc repositioning, on stress distribution in TMJs with DDwoR.

In conclusion, this study highlights significant differences in Von Mises stress distribution between healthy and DDwoR TMJs, emphasizing the biomechanical adaptations in unilateral DDwoR. These insights contribute to the growing body of knowledge on TMJ function and its implications for the diagnosis and management of TMD

FUNDING

This research was supported by the University Scientific Research Fund (project no: PYO.DIS.1904.20.003)

DISCLOSURE STATEMENT

No potential conflict of interest was reported by the authors.

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CASE REPORT

Osteonecrosis of the Jaw in a Young Patient: Beyond Medication-Related Causes

Genç Bir Hastada Çene Osteonekrozu: İlaçla İlgili Nedenlerin Ötesinde

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ABSTRACT

Medication-Related Osteonecrosis of the Jaw (MRONJ) is commonly associated with pharmacological agents like bisphosphonates. However, osteonecrosis can also develop due to other etiologies, including dental trauma and infections. This case report describes osteonecrosis in a 26-year-old male patient who underwent simultaneous implant placement and surgical tooth extraction. The absence of systemic diseases or medication history highlights the role of dental trauma and impaired healing in osteonecrosis development. Management included surgical debridement and antibiotic therapy, consistent with Stage 2 osteonecrosis guidelines. The case emphasizes the importance of minimizing trauma during dental procedures to prevent osteonecrosis, even in younger patients.

Keywords: dental implant, MRONJ, osteonecrosis, trauma

ÖZET

İlaca Bağlı Çene Osteonekrozu (MRONJ), genellikle bifosfonatlar gibi farmakolojik ajanlarla ilişkilendirilse de, dental travma ve enfeksiyonlar gibi diğer etiyolojik faktörlere bağlı olarak da gelişebilmektedir. Bu vaka raporu, eş zamanlı implant yerleştirilmesi ve cerrahi diş çekimi uygulanan 26 yaşındaki bir erkek hastada gelişen osteonekrozu ele almaktadır. Hastanın sistemik bir hastalık veya ilaç kullanım öyküsünün bulunmaması; osteonekroz gelişiminde dental travma ve bozulmuş iyileşme sürecinin etkisini ön plana çıkarmaktadır. Tedavi, Evre 2 osteonekroz kılavuzları doğrultusunda, cerrahi debridman ve antibiyotik uygulaması ile gerçekleştirilmiştir. Çıkarılan materyalin histopatolojik incelemesi, osteonekroz tanısını doğulamıştır. Bu vaka, genç hastalarda dahi osteonekrozu önlemek için dental işlemler sırasında travmayı en aza indirmenin önemine dikkat çekmektedir.

Anahtar Kelimeler: dental implant, MRONJ, osteonekroz, travma

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Acceptance Date: January 13, 2025
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INTRODUCTION

Osteonecrosis of the jaw (ONJ) is a growing concern in dentistry, commonly associated with medications like bisphosphonates, but can also result from other causes, including dental trauma, infections, and radiation therapy. Medication-Related Osteonecrosis of the Jaw (MRONJ) specifically arises after the use of antiresorptive drugs and antiangiogenic therapies[1], and often following oral surgeries like tooth extractions.[2] Robert E. Marx was the first to disclose ONJ, publishing the study on the adverse effect seen in bone metastases of cancers treated with bisphosphonates in 2003.[3] The American Association of Oral and Maxillofacial Surgeons (AAOMS) redefined the condition in 2014 to include other medications beyond bisphosphonates.[4,5]

Dental implant placement is a common treatment for tooth loss, especially in younger patients.[6] While the surrounding bone is expected to remain healthy, trauma from the procedure can disrupt bone metabolism, potentially leading to complications such as ONJ.[7] This case report discusses a 26-year-old male who developed ONJ following implant placement, without prior medication or systemic disease.

CASE REPORT

A 26-year-old male patient presented with exposed bone and purulent discharge around a dental implant placed at an external clinic (Figure 1). The implant procedure included simultaneous extraction of an impacted tooth13.The patient reported no systemic diseases or medication use. Radiographic examination revealed significant bone loss and sequestrum formation around the implant. In addition, a slightly radiopaque area was observed in the upper region of the implant (Figure 2).

Treatment involved implant removal, surgical debridement, and wound closure. The extracted materials were sent to the Department of Oral Pathology. Histopathological examination revealed dense mixed type inflammation within the connective tissue, microorganism colonies, necrotic debri, along with devitalized bone trabeculae lacking viable osteocytes in the lacunae (Figure 3). In histochemical studies, Brown-Brenn staining revealed the presence of gram-positive bacteria. No actinomyces colonies were identified in the Light Green PAS staining.

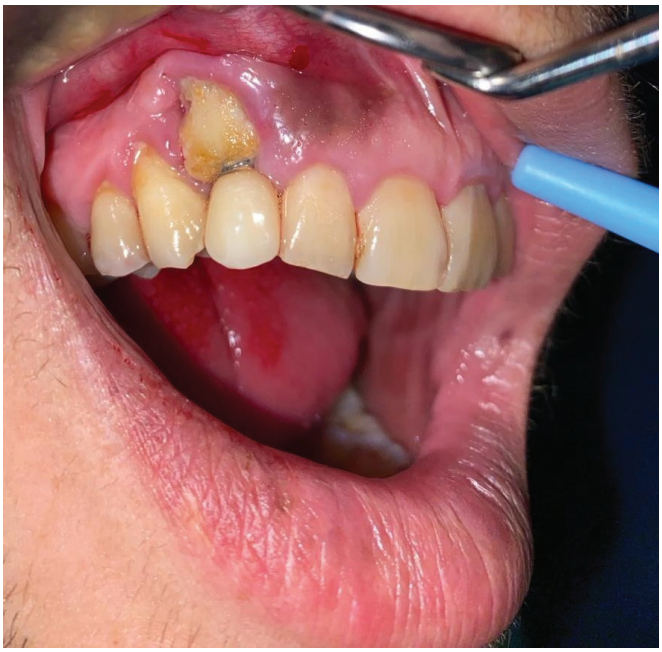

Figure 1. Intraorally, exposed bone and purulent discharge were seen around the implant site, attributed to the absence of tooth 13.

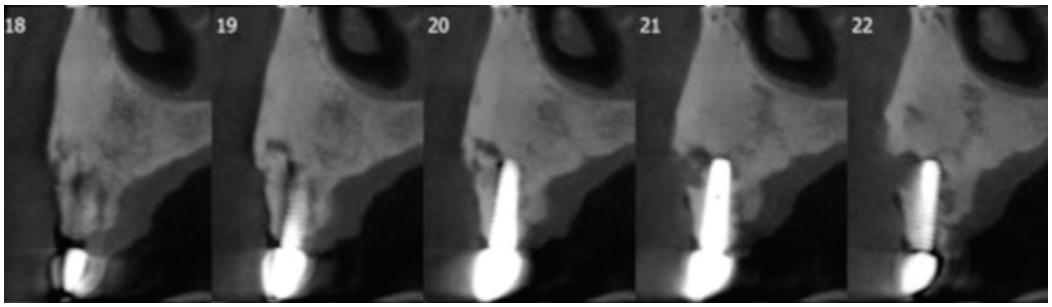

Figure 2. In CBCT, it was noted that significant bone loss and sequestrum formation around the implant.

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The diagnosis of Stage 2 ONJ was established based on bone exposure and discharge. The patient is currently undergoing antibiotic treatment and is in the process of waiting for the affected area to heal.

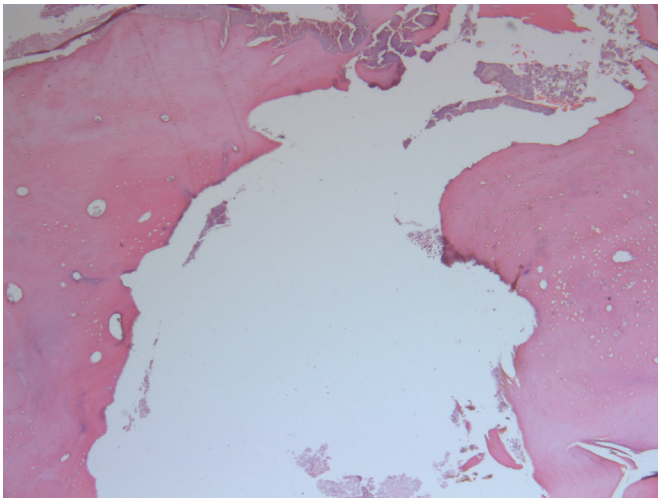

Figure 3. In histopathological examination microorganism colonies, necrotic debri, along with devitalized bone trabeculae lacking viable osteocytes in the lacunae were seen (Hematoxylin&eosin, x40 magnification)

DISCUSSION

Since 2003, osteonecrosis of jaw has been associated with bisphosphonates (BRONJ)[8] was then modified to incorporate other drugs, and in 2014 it was renamed (MRONJ).[5] However, similar clinical presentations can occur without antiresorptive medication use, with reported causes including infections, trauma, smoking, and systemic conditions like diabetes and chemotherapy.[1,5] In our case, dental trauma during implant placement likely triggered ONJ.[9] Studies show ONJ can occur after tooth extraction without medication.[10] The American Association of Oral and Maxillofacial Surgeons has identified dental implant placement as a potential risk factor for osteonecrosis[5], though more research is needed to quantify this risk. The patient's young age contrasts with the typical presentation of MRONJ, which predominantly affects older individuals receiving long-term antiresorptive therapies.[5]

Necrotic areas in ONJs often contain Actinomyces, gram-positive bacteria, and sometimes Candida species.[8] In our case, Brown-Brenn staining revealed gram-positive bacteria, while Light Green PAS staining showed no Actinomyces colonies. Bacterial infection is a key factor in post-traumatic ONJs development and should be considered critically in such cases.[8] It appears that the radiopaque area observed in the upper region of the implant in this case is attributable to a bacterial infection. Furthermore, the fact that the implant was placed in an external centre may be the main cause of bacterial infection, as sterilization rules are not followed in most centres.

Management aligned with AAOMS guidelines for Stage 2 osteonecrosis, focusing on infection control, surgical debridement, and wound closure. This case underscores the importance of minimizing trauma and ensuring optimal healing during dental implant procedures, even in low-risk patients.[5] In the differential diagnosis of osteonecrosis of the jaw, conditions such as chronic osteomyelitis, neoplastic lesions, odontogenic infections, and osteoradionecrosis or Paget's disease should be considered.[1] Osteomyelitis can resemble osteonecrosis with symptoms like bone sequestration, pain, and discharge, but it usually involves more diffuse bone damage, fever, and a history of infection. In our case, the localized bone exposure, absence of systemic symptoms, and histopathological findings of devitalized bone with gram-positive bacterial colonization support the diagnosis of ONJ rather than osteomyelitis. Moreover, there was no history of systemic conditions or medication use that could predispose the patient to other causes of osteonecrosis, such as MRONJ or osteoradionecrosis.[9]

The patient's young age and absence of systemic risk factors emphasize the role of local factors such as osseointegration failure and bacterial colonization, as confirmed by histopathological findings. While osteonecrosis predominantly affects the mandible due to its reduced blood supply, the maxillary occurrence in this case highlights the impact of procedural trauma.[9]

In conclusion, regardless of the patient's age, care should be taken to minimize trauma during dental implant placement, and attention should be paid to ensuring optimal healing of the area.

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CASE REPORT

Garré's Sclerosing Osteomyelitis – A Rare Case Report

Garré'nin Sklerozan Osteomiyeliti – Nadir Bir Vaka Sunumu

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ABSTRACT

Garre's osteomyelitis (GO) is a rare chronic inflammatory disease characterized by periosteal reactions and new bone formation due to bacterial odontogenic infection. This disease, which usually affects children and young people, occurs in the jaw bones such as the mandible, especially in the premolar and molar regions. Clinically, it is characterized by facial asymmetry and hard swelling in the jaw area. The radiographic finding known as "onion skin" appearance is typical for GO.

In this case report, we describe a 10-year-old male patient with GO arising from the right mandibular first molar. Facial asymmetry and hard swelling in the mandibular region were observed; radiologic examination revealed periosteal reaction and new bone formation. The affected tooth was extracted, antibiotic treatment was administered and the bone contours returned to normal after 9 months of follow-up.

Elimination of the source of infection, antibiotic treatment and promotion of bone healing are essential in the treatment of GO. This case is an important example for clinical approaches in the diagnosis and treatment of GO.

Keywords: Garre's osteomyelitis, Periosteal reaction, Mandible

ÖZET

Garre osteomiyeliti (GO), bakteriyel odontojenik enfeksiyona bağlı periosteal reaksiyonlar ve yeni kemik oluşumu ile karakterize, nadir görülen kronik inflamatuar bir hastalıktır. Genellikle çocukları ve gençleri etkileyen bu hastalık, mandibula gibi çene kemiklerinde, özellikle premolar ve molar bölgelerde görülür. Klinik olarak genellikle yüz asimetrisi ve çene bölgesinde sert şişlik ile kendini gösterir. "Soğan kabuğu" görünümü olarak bilinen radyografik bulgu, GO için tipiktir.

Bu olgu sunumunda, 10 yaşındaki erkek hastada sağ mandibular birinci molar dişe bağlı olarak gelişen GO tanımlanmıştır. Hastada yüz asimetrisi ve mandibular bölgede sert şişlik gözlenmiş; radyolojik incelemede periosteal reaksiyon ve yeni kemik oluşumu saptanmıştır. Tedavi olarak ilgili diş çekilmiş, antibiyotik tedavisi uygulanmış ve 9 aylık takipte kemik konturlarının normale döndüğü görülmüştür.

GO tedavisinde enfeksiyon kaynağının giderilmesi, antibiyotik tedavisi ve kemik iyileşmesinin desteklenmesi esastır. Çocuklarda erken teşhis ve uygun tedavi, büyüme çağındaki kemik sağlığı için kritik öneme sahiptir. Bu vaka, GO'nun tanı ve tedavisindeki klinik yaklaşımlar için önemli bir örnek teşkil etmektedir.

Anahtar Kelimeler: Garre osteomyeliti, Periost reaksiyonu, Mandibula

Submission Date: December 18, 2024
Acceptance Date: January 20, 2025
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INTRODUCTION

GO is a specific type of chronic osteomyelitis characterized by periostal reaction and new bone formation due to bacterial odontogenic infection and was first described by Carl Garre in 1893.[1] It is also referred to as "chronic non-suppurative sclerosing osteomyelitis, proliferative periostitis and periostitis ossificans" in the literature.[2] Predominantly affecting children and adolescents, GO is a rare condition accounting for 2-5% of all cases of osteomyelitis.[3] It occurs at a rate of 13 cases per 100,000 people per year.[4]

Clinically, this reactive process manifests as hard swelling of the jaw and facial asymmetry in patients. This condition primarily affects the premolar and molar regions, one side of the mandibular body. The lesion is usually asymptomatic and not accompanied by general and local signs of inflammation, but the clinical picture can be quite variable.[5]

Radiographically, the disease is characterized by bone thickening on the outer cortical surface of the affected area and new periosteal proliferation of the cortical layer seen in layers. This feature is often referred to as the "onion skin" appearance and is typical radiographic evidence of GO.[6]

CASE REPORT

A 10-year-old male patient presented to our clinic with extraoral swelling in the right mandibular molar region and facial asymmetry.

Extraoral examination revealed a firm and painless swelling, (Figure 1A) and radiologic examination revealed deep dentin caries and periosteal reaction in the right mandibular first molar (Figure 2A). Cone beam computed tomography (CBCT) revealed new bone formation with an "onion skin" appearance (Figure 2B, 2C).

With the consent of the patient's family, the affected tooth was extracted and antibiotic treatment was administered. Following the treatment, complaints decreased, mandibular bone contours returned to normal (Figure 3) and facial asymmetry disappeared in a 9-month follow-up (Figure 1B).

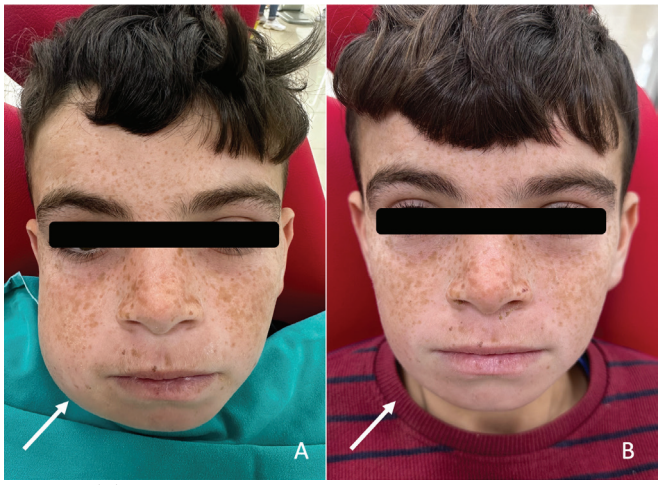

Figure 1: (A) The extraoral photograph of the swelling on the patient's right mandible. (B) Post-operative 9-month extraoral photograph of the patient.

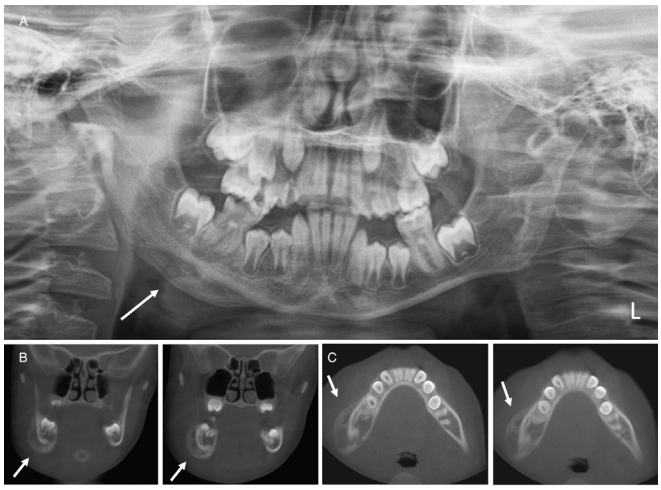

Figure 2: (A) The panoramic radiograph shows an "onion skin" appearance on the outer cortical surface in the right mandibular region. (B) Sagittal section view of bone proliferation on CBCT, (C) Axial section view.

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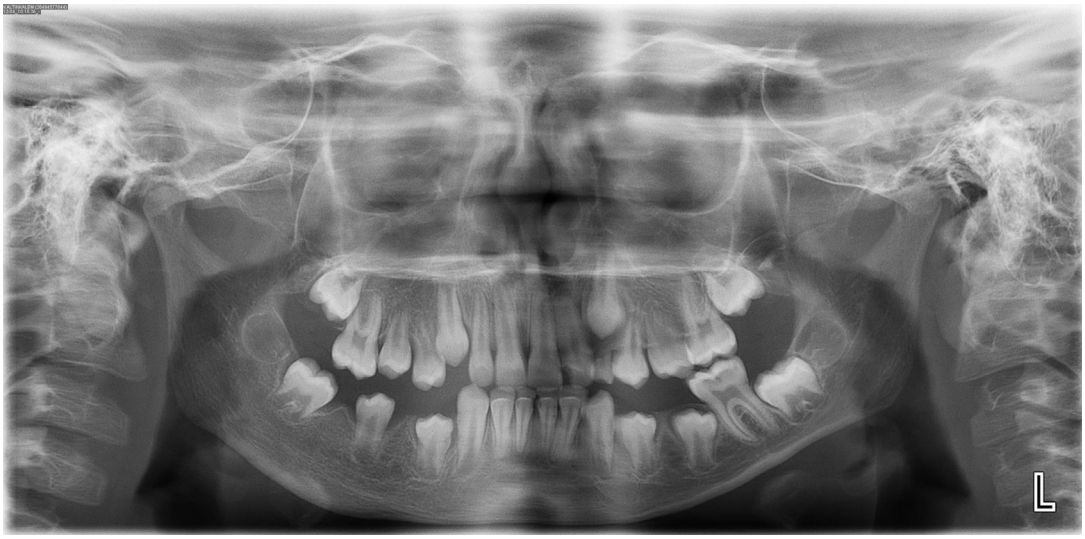

Figure 3: The post-operative panoramic radiograph taken at the 9th month after tooth extraction shows healing in the trabecular pattern and a reduction in cortical bone thickness in the right mandibular region.

DISCUSSION

GO is a rare chronic inflammatory disease characterized by new bone formation as a result of periosteal reactions. It usually occurs in young individuals and leads to intense osteoblastic activity in the periosteum. The mean age of affected individuals is reported to be 13 years.[7]

GO is usually caused by odontogenic infections. The most common cause of these infections is periapical infections due to dental caries.[8] The most commonly accepted method of treatment is the administration of antibiotics and extraction of the infected tooth. Antibiotic treatment causes the odontogenic infection to disappear.[9] When the source of infection is addressed or removed, the enlarged bone gradually returns to its original shape due to the pressure exerted by the surrounding muscles. This process highlights the chronic nature of the condition and helps to understand the balance that exists during the bone healing process. With the extraction of the causative tooth, the periosteal reaction is restored over time. However, in some cases, no obvious cause can be found and these cases are considered idiopathic.[8]

Patients with GO often have facial asymmetry, while pain is rarely reported.[10] Biopsy is usually not required when the cause of infection is identified. Conventional radiography or CT imaging is usually sufficient for diagnosis.[11]

Nortjé et al. analyzed 81 cases of jaw osteomyelitis with periosteal reaction and reported that periosteal bone proliferation occurred in 51.2% of mandibular first molars, 13.2% of second molars and 8.5% of second primary molar.[12]

Differential diagnoses of GO include Fibrous dysplasia, Ewing's sarcoma, Caffey disease, and İnfantile cortical hyperostosis.[11] However, GO can be distinguished by characteristic radiographic findings, such as the "onion skin" appearance, which sets it apart from other conditions.

CONCLUSION

GO is a chronic inflammatory condition commonly found in children. This case, where the bone contours returned to their normal state with proper treatment, highlights the significance of early diagnosis and appropriate intervention. Treating osteomyelitis in pediatric patients is crucial for preserving bone health during the growth phase.

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CASE REPORT

Zimmermann-Laband Syndrome: A Case Report

Zimmermann-Laband Sendromu: Bir Vaka Raporu

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ABSTRACT

Zimmerman-Laband Syndrome (ZLS) is a rare genetic disorder characterized by gingival fibromatosis, craniofacial abnormalities, and limb deformities. This case report describes a 21-year-old male presenting with classical and unique features of ZLS. A male patient was referred for failure of anterior teeth eruption. Clinical examination revealed gingival fibromatosis, hypoplastic toenails, and a broad, flat nose. Additional findings included congenital curly toes and an anterior open bite (AOB). Radiographic evaluation identified multiple impacted and supernumerary teeth. The patient underwent extraction of carious and supernumerary teeth. Orthodontic and surgical interventions were planned for managing unerupted teeth and AOB. This report expands the phenotypic spectrum of ZLS, describing previously unreported findings such as curly toes and hammer toes. Comprehensive evaluation and interdisciplinary management are essential for optimizing outcomes in ZLS patients.

Keywords: Anterior open bite, congenital abnormalities, gingival fibromatosis, impacted teeth, Zimmerman-Laband syndrome.

ÖZET

Zimmerman-Laband Sendromu (ZLS), gingival fibromatozis, kraniofasiyal anormaliler ve uzuv deformiteleri ile karakterize edilen nadir bir genetik hastalıktır. Bu vaka raporu, ZLS'nin klasik ve özgün özellikleriyle başvuran 21 yaşında bir erkek hastayı tanımlamaktadır. Hasta, anterior dişlerin sürmemesi nedeniyle sevk edilmiştir. Klinik muayene, gingival fibromatozis, hipoplastik tırnaklar ve geniş, düz bir burun ile uyumluydu. Ek bulgular arasında doğuştan gelen kıvrımlı parmaklar ve anterior open-bite yer alıyordu. Radyografik değerlendirme, birden fazla gömülü ve süpernümerer dişi ortaya koymuştur. Hastanın çürük ve süpernümerer dişleri çekilmiştir. Sürmeyen dişler ve anterior open-bite için ortodontik ve cerrahi müdahaleler planlanmıştır. Bu rapor, ZLS'nin fenotipik spektrumunu genişleterek, daha önce bildirilmeyen kıvrımlı parmaklar ve çekiç parmaklar gibi bulguları tanımlamaktadır. ZLS hastalarında sonuçların iyileştirilmesi için kapsamlı bir değerlendirme ve multidisipliner yaklaşım gereklidir.

Anahtar Kelimeler: Anterior open-bite, gingival fibromatozis, gömülü diş, konjenital anomaliler, Zimmermann-Laband Sendromu

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INTRODUCTION

Zimmerman-Laband Syndrome (ZLS) is a rare genetic disorder characterized by gingival fibromatosis, craniofacial abnormalities, and other systemic manifestations. The syndrome typically includes a coarse facial appearance, hyperextensibility of small joints, hypoplasia or aplasia of toenails and terminal phalanges, hepatosplenomegaly, and intellectual deficits of varying severity. Additional features such as hirsutism and "dystrophic" finger and toenails have also been documented. The first case of ZLS was described by Zimmerman in 1928, with Laband et al. later reporting familial occurrences and suggesting autosomal dominant inheritance.[1,2]

Although ZLS is generally considered to follow an autosomal dominant inheritance pattern, recessive inheritance has been suggested in certain cases, particularly in families without parental manifestations.[3,4] To date, approximately 36 cases of ZLS have been documented in the literature, highlighting its rarity. Despite its distinctive phenotype, variability in clinical presentation underscores the importance of comprehensive case evaluations.

This report describes a new case of ZLS in a young male patient. Along with the classical features of the syndrome, unique findings such as congenital curly toes and hammer toes are presented, contributing to the expanding phenotypic spectrum of the disorder.

CASE REPORT

A 21-year-old male was referred to our clinic with concerns regarding the failure of eruption of his upper and lower anterior teeth. His medical history indicated that he was born at 39 weeks of gestation to healthy, non-consanguineous parents (29-year-old mother and 33-year-old father). Pregnancy and delivery were uneventful, and his birth weight was 3,150 g.

The patient's physical examination revealed a height of 152 cm and a weight of 55 kg, both within normal limits for his age. His head circumference measured 57 cm. Facial features included a broad, flat, fleshy nose and thick, floppy ears (Figures 1 and 2). Examination of his hands showed no abnormalities; however, the toenails were bilaterally hypoplastic, and the third toes were rudimentary (Figure 3). Additional findings included congenital curly toes involving the fourth and fifth toes of the left foot and the fifth toe of the right foot, as well as hammer toes affecting all toes except the halluces. The patient's skin was dry, soft, and velvety, with thick and bushy eyebrows. Hair growth on the scalp and body was unremarkable.

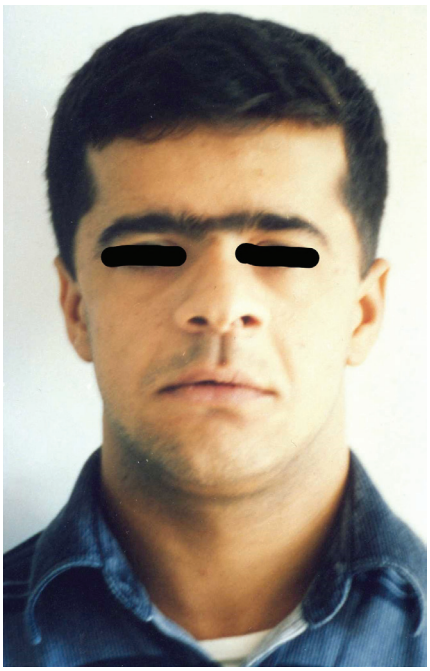

Figure 1. Front view of patient showing broad, flat and fleshy nose.

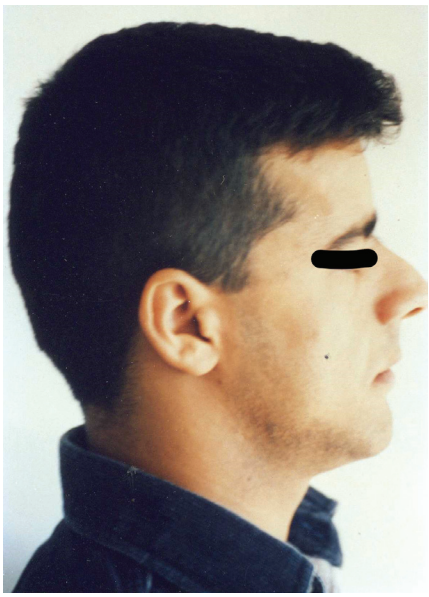

Figure 2. Lateral view of patient showing thick and floopy ears.

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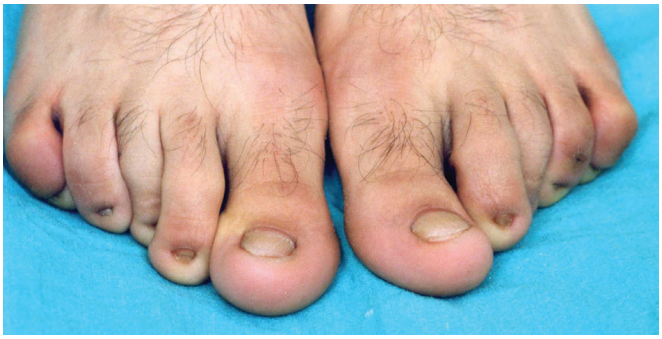

Figure 3. Toes view of patient showing bilaterally hypoplastic nails and other features of toes.

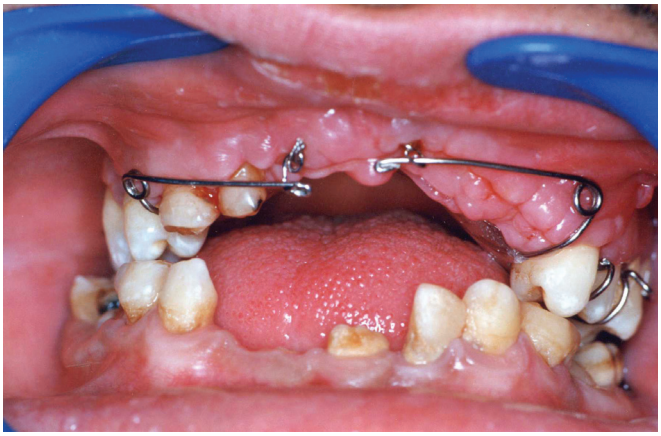

Figure 4. Intraoral view showing an anterior openbite and the condition of teeth.

Neurological and ophthalmological evaluations revealed no pathological findings. Intelligence appeared normal upon subjective assessment. Joint hypermobility was noted in the metatarsophalangeal and knee joints. Liver and spleen were non-palpable, and routine hematological and karyotyping analyses (46, XY) were normal. Since there is no specific gene associated with ZLS yet and it was not possible to establish contact with the patient's relatives, further genetic testing could not be performed.

Oral examination revealed pronounced gingival fibromatosis, which contributed to the failure of tooth eruption. The patient was in the permanent dentition phase, but most permanent teeth were unerupted. Teeth present in the oral cavity included: maxillary deciduous lateral incisors, canines, and primary molars, mandibular deciduous canines, primary molars, and partially erupted permanent central incisors and second premolars. Several unerupted teeth, including maxillary central and lateral incisors, canines, and mandibular lateral incisors and canines, were confirmed via radiography (Table I).

The patient also exhibited an anterior open bite (AOB) (Figure 4), despite no history of thumbsucking or tongue thrusting. There was no evidence of macroglossia.

A panoramic radiograph revealed multiple unerupted permanent teeth and five supernumerary teeth, one located in the maxillary right posterior region and four in the maxillary anterior region (Figure 5). No abnormalities were observed in the vertebral skeleton, skull, or chest.

The patient was admitted for extraction of carious and supernumerary teeth and for the management of unerupted teeth. The informed consent of the patient was obtained regarding the planned procedures. Further orthodontic and surgical interventions were planned to address the AOB and facilitate the eruption of impacted teeth. Since the patient was performing military service during the treatment, long-term follow-up could not be conducted after the patient was discharged.

Table I. Conditions of the teeth of the patient

Fully Erupted Teeth	52,53,14,15,17,62,63,24,25,26,27,83,85,46,72,73,75,36
Partially Erupted Teeth	41,31,45,35
Unerupted Teeth	11,12,13,18,21,22,23,28,32,33,34,35,42,43,44,48
Extracted Teeth	16,47,37,38

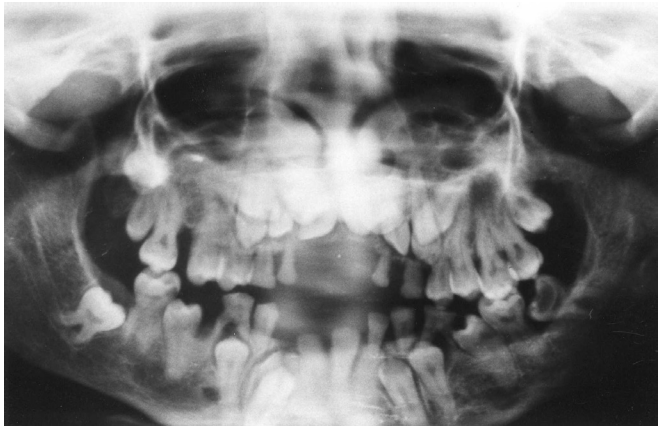

Figure 5. Panoramic radiograph showing unerupted teeth and supernumerary teeth.

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Table II Laband syndrome:Summary of reported findings (Adapted from data in Chadwick et al)

Author and case#	Country of origin	Sex	a	b	c	d	e	f	g	h	i	j	k
Zimmerman 1	Germany	M	+	N	+	N	?	Spina Bifida	+/+	?	+	?/?	-
Zimmerman 2	Germany	F	+	(M.retard)	+	+	?	N	+/+	?	+	?/?	MG
Jacoby et al 1	U.K.	F	+	N	N	N	+	Spina Bifida	+/+	?	N	N/N	-
Laband et al 1	Trinidad	F	+	IQ70	N	+	+	N	+/+	+	N	+/+	-
Laband et al 2	Trinidad	M	+	IQ70	N	+	+	N	+/+	+	N	+/+	-
Laband et al 3	Trinidad	F	+	IQ70	N	+	+	N	+/+	+	N	+/N	-
Laband et al 4	Trinidad	M	+	IQ70	N	+	+	Kyphosis	+/+	+	N	N/N	-
Laband et al 5	Trinidad	F	(+)	IQ70	N	+	+	N	+/+	+	N	+/+	-
Laband et al 6	Trinidad	F	+	IQ70	N	+	+	N	+/+	+	N	N/N	-
Alavandar 1	S. India	F	+	N	N	+	+	N	+/+	+	?	N/N	-
Alavandar 2	S. India	M	+	N	N	+	+	N	+/+	N	?	+/N	-
Alavandar 3	S. India	M	+	N	N	+	+	N	+/+	+	?	+/+	-
Alavandar 4	S. India	M	(+)	N	N	+	+	N	+/+	+	?	N/N	-
Alavandar 5	S. India	M	(+)	N	N	N	N	N	+/+	+	?	N/N	-
Anatasov 1	Bulgaria	F	+	N	+	?	?	?	?/+	?	?	?/?	-
Anatasov 2	Bulgaria	M	+	(M.retard)	?	?	?	?	+/+	?	?	?/?	-
Oikava et al 1	U.S.A.	F	+	(M.retard)	+	+	+	Kyphosis	+/+	N	Thick eyelashes	+/N	-
Chodirker et al 1	Canada	M	+	M.retard	+	+	+	Scoliosis	+/+	+	N	N/N	MG
Pino Neto et al 1	Brazil	F	+	(M.retard)	N	+	+	N	+/+	+	+	+/+	MG
Beemer 1	Holland	F	+	N	+	+	N	N	+/+	N	?	N/N	MG/ AOB
Ili˙na et al1	Ukraine	F	+	(M.retard)	+	+	+	Scoliosis	+/?	N	N	N/N	AOB
Bazoupoulou et al 1	Greece/ Albania	F	+	(M.retard)	+	+	+	N	+/+	+	Synophrys	N/N	AOB
Bakaeen and Scully 1	Jordan	F	+	N	N	+	N	N	+/?	+	N	N/N	-
Bakaeen and Scully 1	Jordan	M	+	N	N	+	N	N	+/+	+	N	N/N	-
Pfeiffer et al 1	Germany	M	+	M.retard	+	+	N	Spondylodyplasia	+/?	?	+	+/N	-
Pfeiffer et al 1	Germany	M	+	N	N	+	+	N	+/+	?	+	+/+	-
Chadwick et al	U.K.	F	+	N	+	+	N	N	+/+	N	+	+/N	AOB
Chadwick et al 2	Pakistan	M	+	(M.retard)	+	+	+	N	+/+	N	+	N/N	AOB
Koch et al 1	Germany	F	+	N	+	+	?	N	+/+	N	N	N/N	ARP
Lacombe et al 1	S. India	+	+	? Slightly delayed motor development	+	+	+	?	+/+	(+)	+	+/N	-
Van Buggenhout et al 1	Holland	M	+	M. retard	N	+	+	Scoliosis	+/+	+	Bushy eyebrows	N/N	RI
Robertson et al 1	Australia	M	+	M. retard	+	+	+	?	+/+	+	Thick eyebrows, synophrys, hirsutism	N/N	CVC
Dumic et al 1	Croatia	F	+	?	?	+	+	?	+/+	+	+	+/?	MG
Katz et al 1	USA	F	+										
Stefanova et al 1	Bulgaria	F											
Stefanova et al 1	Bulgaria	F											
Holzhausen 1	Brasil												
Davalos et al 1	Mexico	F											
Davalos et al 1	Mexico	M											
Atabek et al 1	Turkey	M											
Kim et al 1	USA	M											
Kissi et al 1	France	F											
Douzgou et al 1	Italy												
Ortakoglu et al 1	Turkiye	M	+	N	N	+	+	N	N/+	+	Thick eyebrows	N/N	AOB
Shrian et al 1	Iran	F	+	N					+/N		+	+/+	VSD, telecanthus
Kshirsagar et al 1	India	F	+	Impaired intellectual and adaptive function	+	+							AOB, MG

List of Abbreviations
a: Gingival fibromatosis
b: Mental development
c: Thick lips
d: Broad nose
e: Large Ears
f: Spine
g: Aplasia or hypoplasia of nails or terminal phalanges of hands / feet
h: Hyperextensibility of joints
i: Hypertrichosis
j: Hepatomegaly / splenomegaly
k: Other
M: Male
F: Female
N: Normal
?: Not reported
MG: Macroglossia
AOB: Anterior open-bite
(...):Mild manifestation
IQ Intelligence quotient
ARP: Atypical retinitis pigmentosa
CVC: Cardiac and vascular complications
RI: Retrognathia inferior
VSD: Ventricular septal defect

EurAsian Journal of Oral and Maxillofacial Surgery
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DISCUSSION

A total of 36 cases of ZLS have been reported in the literature. Laband et al. described six individuals from a single family, Alavandar documented five cases within a pedigree, and Bakeen et al., along with Shirian et al., reported two affected siblings.[2-4,22] Additionally, 21 isolated cases without familial involvement have been identified.[1,5-19,23] This article presents a further isolated case of ZLS, where no other family members exhibited symptoms of the syndrome.

ZLS is a rare genetic disorder characterized by gingival fibromatosis, nasal and/or ear abnormalities, and hypoplasia or absence of the nails or terminal phalanges of the hands and feet. Other clinical manifestations may include joint hyperextensibility, hepatomegaly, splenomegaly, hypertrichosis, mental retardation, thick lips, macroglossia, AOB, and occasional spinal abnormalities. The features of reported cases, summarized in Table II, show overlapping characteristics but also variability. Despite phenotypic similarities to known storage disorders, no definitive biochemical defect has been identified

The diagnosis of ZLS in our patient was based on hallmark clinical findings, including a coarse facial appearance with a broad nose, thick and floppy ears, gingival fibromatosis, and hypoplasia or absence of toenails.

AOB has been widely reported in ZLS cases, including those by Ili'na et al., Beemer, Bazoupoulou-Kyrkanidou et al., and Chadwick et al.[5-8] These authors proposed that AOB is caused by the combined effects of a macroglossia and gingival fibromatosis. In our patient, AOB was also observed; however, macroglossia and digit-sucking habits, often associated with AOB, were absent.

Supernumerary teeth have been previously reported by Chadwick et al.[5] In our case, this finding was also noted, suggesting that supernumerary teeth may represent a secondary but less common feature of ZLS.

Congenital curly (varus) toes and hammer toes have not been described in the literature to date and may represent a novel clinical manifestation of ZLS.

Laband et al. reported a mother with seven children, five of whom were affected, while Alavandar described an affected mother, three affected sons, and an affected grandson.[2,3] These cases support an autosomal dominant inheritance pattern. Although a complete family pedigree was unavailable for our case, the observed data are consistent with autosomal dominant transmission

Genetic investigations into ZLS remain inconclusive. Stefanova et al. identified a candidate locus in the 3p14.3 region of the chromosome, while Hoogendijk et al. found an insertion in the 8(10) chromosomal region. Shirian et al. and Kshirsagar et al. conducted genetic analyses that failed to detect any DNA sequences associated with known genetic diseases or nonsyndromic conditions.[20-23] These findings underscore the need for further research to elucidate the genetic basis of ZLS and its underlying pathophysiology.

This case contributes to the expanding phenotypic spectrum of ZLS and highlights the necessity for ongoing genetic and clinical studies to better understand this complex syndrome.

CONCLUSION

ZLS is a rare genetic disorder with a wide spectrum of clinical manifestations. This case report documents a 21-year-old male presenting with both classical features of ZLS, such as gingival fibromatosis, impacted teeth, and craniofacial abnormalities, and novel findings, including congenital curly toes and hammer toes. These observations expand the phenotypic spectrum of the syndrome and highlight the variability in its presentation.

The dental and systemic challenges associated with ZLS require a multidisciplinary approach involving oral surgeons, orthodontists, geneticists, and other specialists to optimize patient outcomes. Comprehensive evaluation and early intervention are essential for addressing dental and skeletal abnormalities, improving quality of life, and preventing complications

Further research and genetic studies are necessary to confirm the inheritance patterns and underlying genetic mutations associated with ZLS. This case underscores the importance of documenting rare syndromes to enhance understanding and guide the development of tailored management strategies. As the existing studies to date have not identified a specific treatment for the disease, the current treatment approach focuses on managing the abnormalities associated with the condition. In the case presented here, the aim was not the treatment of the disease itself but rather its diagnosis and a comparison of the symptoms reported in other studies.

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

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