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Ligneous gingivitis: diagnostic and management challenges in a rare case

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Abstract

Introduction Ligneous gingivitis (LG) is a rare manifestation of plasminogen deficiency characterized by woody fibrin-rich pseudomembranes, recurrent ulceration, and progressive periodontal destruction

Case presentation We report a 16-year-old female with hypothyroidism who presented with persistent gingival lesions and tooth mobility since early childhood after a mumps infection. Clinical and radiographic evaluation revealed generalized alveolar bone destruction. Histopathology confirmed LG with fibrinoid deposits. Hematological analysis showed markedly reduced plasminogen levels, measured using an ELISA-based assay, confirming the systemic deficiency. The patient was referred for ophthalmologic and genetic evaluation, targeting PLG gene mutations (K19E hotspot). She received chlorhexidine therapy with improved ulceration and reduced bleeding. At three-month follow-up, the lesions remained stable, and no ocular involvement was observed.

Conclusions This case illustrates diagnostic challenges, differential considerations with aggressive periodontitis, and emphasizes the need for early recognition, multidisciplinary management, and systemic treatment to prevent severe complications such as blindness, infertility, or progressive periodontal morbidity.

Keywords Fibrinolysis, Ligneous gingivitis, Multidisciplinary management, Periodontal disease, Plasminogen deficiency

Background

Ligneous gingivitis (LG) is a rare condition characterized by nodular gingival overgrowth, ulceration, and periodontal tissue destruction [1]. This disorder is strongly associated with plasminogen deficiency, an autosomal recessive condition that impairs fibrinolysis, leading to fibrin accumulation and pseudomembranous lesions in mucosal tissues [2, 3]. Ligneous gingivitis often manifests in childhood, with affected individuals developing thick, yellowish-white, fibrin-rich pseudomembranes on the oral mucosa, which are referred to as woody plaques that give the disease its name [4].

Globally, the estimated prevalence of plasminogen deficiency is approximately 1 per 1,000,000 individuals, with reported cases from Europe, North America, and Asia [5]. The prevalence of LG is notably higher in Turkish populations, likely due to genetic predisposition [5]. These lesions primarily affect the oral cavity, eyes, respiratory tract, and female genital tract. Clinical features typically include woody pseudomembranes of the gingiva, recurrent ulcerations, and systemic involvement such as conjunctivitis or cervicovaginitis. The hallmark of LG is the deposition of fibrin-rich, amyloid-like material in the lamina propria, which results in nodular growths

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prone to ulceration [2, 4]. The condition is often triggered by minor trauma or infections and has a high recurrence rate.

Diagnostic confirmation typically involves biopsy and histological analysis, revealing characteristic eosinophilic, fibrinoid deposits. Plasminogen deficiency, the primary cause of LG, disrupts fibrinolysis and leads to fibrin accumulation in mucosal tissues. This condition is classified into Type I (hypoplasminogenemia), caused by reduced plasminogen production, and Type II (dysplasminogenemia), involving functional abnormalities of normal plasminogen. LG is predominantly linked to Type I deficiency, with the K19E mutation on chromosome 6q26-27 being the most common genetic defect [3]. The systemic nature of plasminogen deficiency often results in complications such as ligneous conjunctivitis, which can lead to blindness, and ligneous cervicovaginitis, which may cause infertility in homozygous females. Severe deficiency triggers a vicious cycle of inflammation, delayed wound healing, and progressive membrane thickening.

Management of LG remains challenging due to the rarity of the condition and lack of standardized treatments. Options such as systemic anticoagulants, corticosteroids, and plasminogen replacement therapies have been employed with variable success [6]. Chlorhexidine mouthwash is a widely used antiseptic that supports plaque control and reduces gingival inflammation, serving as an adjunctive treatment, though not curative. Heparin mouthwash, on the other hand, is rarely utilized and lacks substantial evidence of efficacy, as reported by Fine et al. (2009) [7]. Surgical excision, while sometimes performed to alleviate symptoms, can worsen the condition by triggering rapid lesion recurrence.

Despite advancements in understanding the pathophysiology, further research is needed to establish effective therapeutic protocols and improve patient outcomes. A rare case of lignous gingivitis and the diagnosis of plasminogen deficiency is reported in this article.

Case presentation

A 16-year-old female patient with hypothyroidism, managed with levothyroxine since age eight, was referred to the Oral and Maxillofacial Surgery Department at Istanbul University Faculty of Dentistry due to a diagnosis of aggressive periodontitis. Her primary complaint was a persistent wound on her gums that had not healed since a mumps infection at the age of three, along with tooth mobility and pain during eating (Fig. 1).

The patient reported frequent bleeding during brushing, pain while eating hard foods, halitosis, and aesthetic concerns about her gums. She described her efforts to

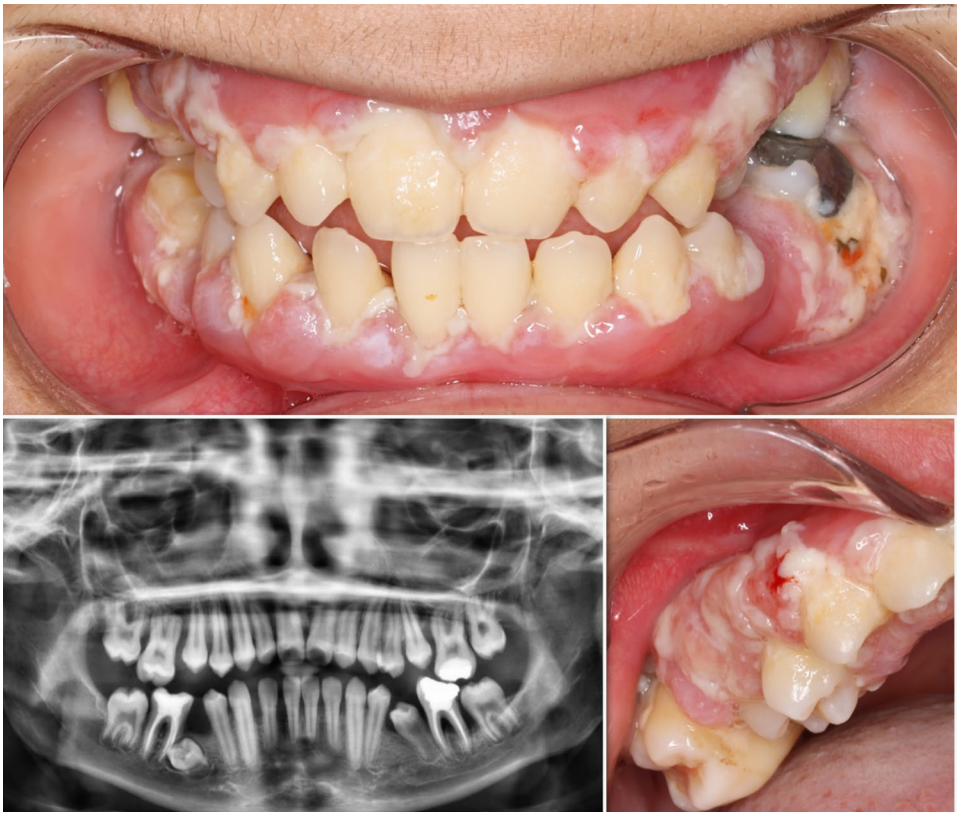

Fig. 1 Clinical appearance and the panoramic radiograph of the patient

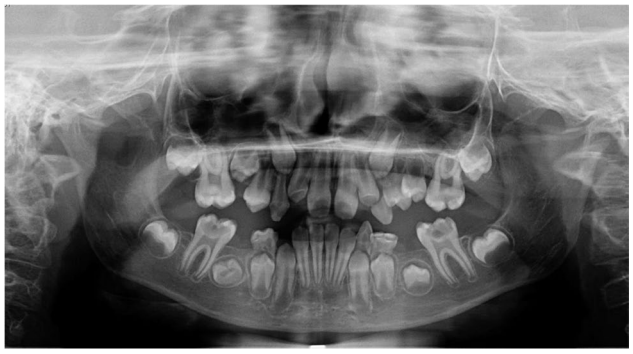

Fig. 2 Panoramic radiograph of the patient taken in 2019

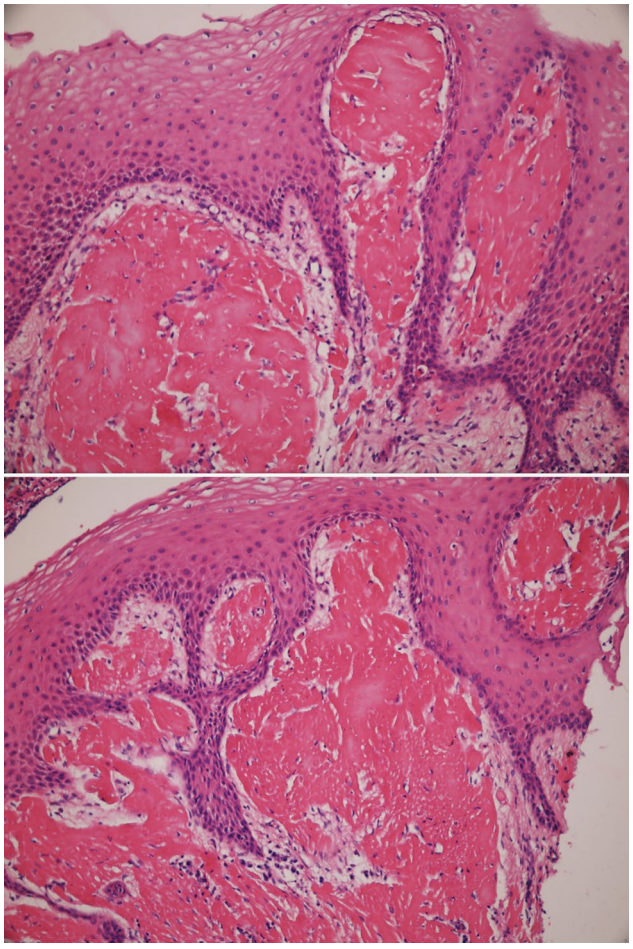

Fig. 3 Structureless, eosinophilic, fibrinoid substance accumulation under the stratified squamous epithelium covering the surface (H&E X 200)

clean the lesions through brushing, which only appeared to exacerbate the condition. Despite undergoing periodontal treatments, including a biopsy at another center, the patient continued to experience persistent symptoms, with no improvement in the lesions. Histopathological findings from the earlier biopsy indicated a benign tissue lined with ulcerated acanthotic squamous epithelium, containing areas of intense chronic active inflammation and inflammatory granulation tissue within the wall.

During the clinical examination, generalized gingival enlargement with hyperemic and nodular appearance was observed, along with advanced gingival recession (Fig. 1). Firm, woody plaques with ulcerations, often yellowish-white, were noted predominantly in the posterior gingiva. Poor oral hygiene, significant bacterial plaque accumulation, tooth mobility, and bad breath were evident. Panoramic radiography revealed generalized alveolar bone destruction, involving both anterior and posterior sites, which had not been observed in imaging taken five years earlier, according to records provided by the patient (Fig. 2).

An incisional biopsy of the lesion, on the buccal side of the right maxillar posterior area including adjacent intact tissue, was performed under local anesthesia using Articaine HCl with adrenaline. The tissue sample was fixed in a 10% formaldehyde solution and sent to the Oral Pathology Department for analysis. Histopathological examination confirmed the diagnosis of ligneous gingivitis, revealing areas where the multilayered epithelium was absent and replaced by exudate and debris. Subepithelial tissue showed prominent eosinophilic, unstructured fibrinoid material deposition and intense inflammatory cell infiltration in deeper regions (Fig. 3).

Given the established link between LG and plasminogen deficiency, the patient was referred to the hematology department. Hematological analysis revealed significantly reduced plasminogen levels at 32 mg/dL, while the normal reference range is 65–176 mg/dL, measured using an ELISA-based plasminogen activity assay, confirming plasminogen deficiency as the underlying cause of her condition. Before initiating treatments to increase plasminogen levels, the patient was also referred to the genetics department for further evaluation and consultation. Genetic testing was planned for PLG gene mutations, particularly the K19E mutation on chromosome 6q26-27, which is most frequently reported in association with this condition. She was also referred to an ophthalmologist, where no lesions were found in her eyes.

The patients was advised to use a soft-bristled toothbrush and to brush using her non-dominant hand to reduce pressure. Additionally, the lesions were monitored using a 0.12% chlorhexidine mouthwash twice daily. Chlorhexidine mouthwash, used in combination with a soft-bristled toothbrush for two weeks, resulted in reduced ulceration and improved the overall appearance of the lesions (Fig. 4). The follow-up photograph (Fig. 4) was obtained three weeks after initiating chlorhexidine therapy, and the patient has remained under monthly follow-up at our clinic since diagnosis. The patient has been under regular monthly follow-up for three months.

Fig. 4 Clinical appearance of the lesions after 0.12% chlorhexidine treatment

During this period, the lesions have remained stable with reduced ulceration, less bleeding on brushing, and improvement in oral comfort. No ocular or systemic complications have been detected to date. Future therapeutic planning includes evaluation for systemic plasminogen replacement therapy under the supervision of the hematology department.

Discussion and conclusions

Ligneous gingivitis (LG) was first described by Frimodt-Mollers in 1973 [8]. The term "lignous," derived from the Latin word for "woody," aptly describes the hard, woody appearance of mucosal lesions caused by this condition. LG is a manifestation of plasminogen deficiency, an autosomal recessive disorder that disrupts fibrinolysis and leads to the accumulation of fibrin-rich pseudomembranes in mucosal tissues. The estimated prevalence of plasminogen deficiency is approximately 1 in 1,000,000, making it a rare but clinically significant disorder [2].

Ligneous gingivitis (LG) is notably more common in Turkey, often linked to consanguineous marriages. Studies, including Schuster et al. and Tefs et al., report a high percentage of Turkish patients with plasminogen deficiency, emphasizing genetic factors [3, 9]. A systematic review also found that 53.1% of LG cases were of Turkish origin, highlighting this population's susceptibility. In this case, the patient was also of Turkish origin, further supporting the observed genetic predisposition [10].

Plasminogen plays a critical role in fibrin degradation, wound healing, angiogenesis, and tissue remodeling. Insufficient plasminogen levels result in impaired fibrinolysis and a cascade of chronic inflammation, persistent fibrin deposition, and delayed tissue repair. This vicious cycle leads to the characteristic thickening of mucosal membranes observed in LG [2]. Although LG primarily affects the gingiva, it is often associated with ligneous conjunctivitis, the most common clinical manifestation of plasminogen deficiency. Ligneous conjunctivitis typically involves the upper eyelids and may progress to corneal involvement, potentially resulting in blindness. Similarly, involvement of other mucosal surfaces, such as the respiratory and female genital tracts, highlights the systemic nature of the disease [11].

The triggering events for LG often include minor trauma or infections, as seen in this patient, whose symptoms began following a mumps infection at the age of three. This aligns with existing literature emphasizing infections as a common precipitating factor for the onset of LG [1]. Despite its distinct clinical presentation, LG can be misdiagnosed as aggressive periodontitis due to overlapping features, such as ulceration and gingival inflammation. In fact, this patient initially presented with a diagnosis of aggressive periodontitis, further emphasizing the challenges in distinguishing LG from other periodontal conditions. This highlights the importance of histopathological examination and multidisciplinary collaboration for accurate diagnosis [7].

The differential diagnosis of LG includes aggressive periodontitis, chronic gingivitis, and mucocutaneous disorders such as lichen planus, which may mimic LG due to their ulcerative gingival manifestations [12]. These conditions should be carefully excluded through histopathology and systemic evaluation. Furthermore, distinguishing LG from other pediatric oral pathologies, such as recurrent aphthous stomatitis or even systemic conditions manifesting with oral ulcers, requires a thorough clinical assessment, given that the prevalence of oral ulcers in children can be as high as 9% and often presents diagnostic challenges due to their non-specific nature [12].

Management of ligneous gingivitis remains a major clinical challenge. Local measures such as chlorhexidine rinses and meticulous plaque control may provide temporary symptomatic relief but are insufficient as stand-alone therapy. Systemic interventions have been explored with varying degrees of success. Plasminogen replacement therapy, using fresh frozen plasma or purified plasminogen concentrates, has demonstrated lesion regression in several case reports [13, 14]. Antifibrinolytic agents, corticosteroids, and immunosuppressants have also been trialed, though outcomes are inconsistent [7]. Surgical excision is generally discouraged due to high recurrence rates and potential exacerbation of lesions. Supportive periodontal therapy—including atraumatic oral hygiene, soft-bristled toothbrushes, and regular follow-up—is essential for stabilizing gingival tissues. In this case, chlorhexidine therapy and modified brushing technique achieved partial improvement, and the patient's condition has remained stable during three months of follow-up. Longer-term outcomes will depend on systemic correction of plasminogen deficiency, highlighting the need for coordinated care between dentistry, hematology, and genetics.

Surgical excision in the treatment of ligneous gingivitis (LG) is generally considered a short-term solution for alleviating symptoms and improving aesthetics. However, sources highlight the high recurrence rate of lesions and the potential risks associated with such interventions. Schuster et al. reported that surgically removed membranes quickly reformed, recommending surgery only in emergency cases [3]. Similarly, Sartori et al. observed the development of firm fibrin plaques following root extraction during dental treatment [14]. Fine et al. found limited benefits from excising fibrinous masses and using topical treatments [7]. Additionally, Günhan et al. documented rapid recurrence of lesions after subgingival curettage and gingivectomy [15]. Therefore, in this case, surgical excision was not performed due to the worsening of lesions and increased tooth mobility. Chlorhexidine has been shown to slow the progression of ligneous gingivitis (LG) by reducing bacterial plaque and alleviating gingival inflammation [12]. In this case, the use of chlorhexidine mouthwash twice daily, combined with improvements in brushing technique, resulted in reduced ulceration and a noticeable improvement in lesion appearance after three weeks.

Plasminogen replacement therapy, particularly using fresh frozen plasma (FFP) and purified plasminogen concentrates, has shown significant improvement in LG cases. Studies such as those by Schuster et al. and Kızılocak et al. have reported symptom resolution and lesion regression with increased plasminogen levels [3, 13]. In this case, we anticipate clinical improvement with similar plasminogen augmentation methods. The patient has been referred to the hematology and genetics departments for advanced evaluation, with treatment protocols being determined based on the findings. Additionally, the patient is under regular follow-up with both an ophthalmologist and our clinic to monitor the condition.

In conclusion, this case emphasizes the diagnostic difficulties and systemic implications of ligneous gingivitis. Early recognition, detailed histopathological and genetic evaluation, and a multidisciplinary management strategy are crucial for improving patient outcomes. The combination of chlorhexidine mouthwash and oral hygiene optimization provided partial local control, but systemic treatment remains essential. Awareness of differential diagnoses and timely systemic evaluation can prevent severe complications such as blindness, infertility, and extensive periodontal destruction, ultimately improving the quality of life in affected individuals. This case not only illustrates diagnostic clarity but also adds value to current literature by providing structured insights into management strategies and follow-up outcomes.

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Authors' contributions
M.B.K. and B.G. were the surgeons; N.V.O. was the oral pathologist of the case. A.T. prepared manuscript text/figures and M.B.K revised the paper.

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Data availability
Data sharing is not applicable to this article as no datasets were generated or analyzed during the current study.

Declarations

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Consent for publication
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Competing interests
The authors declare no competing interests.

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