A rare case of periodontal tissue destruction in a patient with lichen sclerosus et atrophicus

BACKGROUND: Lichen sclerosus et atrophicus (LSA) is an uncommon disorder of unknown aetiology. The disease is clinically characterised by white, macular lesions on the skin and is usually associated with atrophic condition affecting the vulva and peri-anal skin. Involvement of the oral mucosa with or without concurrent genital or skin lesions has been reported only occasionally. In view of the rarity of reported cases, one lesion affecting only the labial mucocutaneous area is presented. A 17-year-old female patient came to us with the chief complaint of an unaesthetic appearance of her maxillary anterior teeth as well as receding gums. A white mucosal lesion was seen affecting the labial mucosa along with severe bone and gingival tissue loss around her maxillary incisors.

CASE REPORT: Clinical examination suggested the diagnosis of LSA, which was confirmed by the histopathological report of the lesion. Intralesional injections of steroids were given twice at an interval of 1 week, accompanied by thorough Phase 1 therapy and instructions for proper maintenance of oral hygiene. The mucosal condition resolved in 4 weeks, the involved teeth were saved and the patient was scheduled for regular recall visits. After 1 year, the patient was satisfied with the treatment outcome.

CONCLUSION: Although rare in the oral cavity, LSA may affect the periodontal issues. The clinician should be aware of clinical and histopathological findings of this condition.

INTRODUCTION

Lichen sclerosus et atrophicus (LSA), originally described by Hallopeau in 1887, is an infrequent benign chronic inflammatory dermatosis affecting both the epidermis and dermis. LSA may affect adults of all races, although the disease is more prevalent in Caucasians. The disease is ten times more common in females than in males and is rare in children. The aetiology of LSA is still not clear, although evidence suggests a multifactorial disease. Studies suggest association of LSA with autoimmune processes, and familial occurrence suggests involvement of genetic factors. However, thyroid function, pernicious anaemia, scarring pemphigoid and diabetes mellitus are also reported. Its pathogenesis is not well known and human papilloma virus has been implied. LSA occurs most frequently in the anogenital area with extragenital lesions in only 15% to 20% of patients. The lesions present in the form of ivory-white papules with follicular hyperkeratosis ultimately forming atrophic white plaques. LSA affecting the oral mucosa is exceedingly rare. Only 13 patients with biopsy-proved isolated oral disease...
have been reported in the literature. We report a case of a young female where LSA resulted in severe loss of periodontal tissue around her maxillary anterior teeth.

Case presentation

A 17-year-old female patient presented with the chief complaint of receding gums and the uneaethetic appearance of her maxillary anterior teeth (Fig 1). The patient had no symptoms except mild discomfort in her maxillary left central and lateral incisors. Her history revealed a white patch over the labial mucosa had been present for the past 3 months, which had slowly spread to involve the gingival and mucosal tissue in relation to her maxillary central and lateral incisors. At the time of clinical examination, a well-delineated white mucosal lesion without any signs of inflammation was noticed. The lesion was 1.5 cm in diameter and affected the periodontal tissues, resulting in severe gingival and bone tissue loss around the left maxillary incisors. The radiographic examination revealed severe bone loss around the affected teeth, whereas the rest of the dentition was normal (Fig 2). A biopsy specimen was sent for pathological examination and a histopathological report revealed hyperkeratosis with follicular plugs, spinous layer atrophy, hydropic degeneration of basal layer and hyalinisation of collagen fibres in the superficial lamina-propria. The work-up (complete blood count, glucose, liver function tests, free T4 and thyroid-stimulating hormone [TSH]) was within normal limits. Based on these clinical and histopathological findings a diagnosis of LSA was made. The patient was also referred to her dermatologist, who ruled out the presence of any associated skin or mucosal lesions.

Fig 1 Pre-operative patient photographs.

Fig 2 Radiographs showing marked localised bone loss.
Management

Treatment was started with peri-lesional corticosteroid injections, with 40 mg triamcinolone acetoneide given twice at an interval of 1 week; this was accompanied by thorough Phase 1 therapy and instructions for proper maintenance of oral hygiene to the patient. The mucosal condition resolved in 4 weeks, but the severe bone and soft tissue loss around the teeth limited the aesthetic reconstruction of the lost tissues, and a gingival prosthesis was fabricated to replace the lost tissues and restore the aesthetics (Fig 4). The patient was not willing to undergo extraction, so was put on a regular recall visit of 2 weeks for the first 2 months, every 4 weeks for another 4 months and then on a 3-month recall programme for 1 year. After 1 year, the patient remained free from any lesions and was satisfied with the treatment.

Discussion

Involvement of oral mucosa in LSA with or without concurrent skin lesions has been reported only occasionally in the literature\(^{10,11}\) and there is only one case report describing the involvement of periodontal tissues in association with LSA\(^{12}\). The cause of LSA remains unknown. According to the literature, there is a 21.5% to 34% rate of association between this entity and autoimmune diseases, and 79% of cases of LSA had autoantibodies\(^{8}\). In the present case there was no such association; therefore, the condition may be best described as ‘idiopathic’.

The differential diagnosis included: lichen planus, candidiasis, scleroderma and leukoplakia. In the present case, the lesion appeared as a localised white elevated patch without any reticular lesions at the periphery or any other region of the oral mucosa. Also, the biopsy report was negative for the presence of fungal hyphae and showed the absence of collagen fibres along with the presence of a sclerotic component in the superficial lamina-propria. These findings ruled out the presence of any other mucosal lesions and the diagnosis of oral LSA was established.

The treatment was based on previous reports describing the condition\(^{11,12}\) and was aimed at controlling the progression of the mucosal lesion and preserving the remaining periodontal tissues and the teeth. Although the severity of the disease process prevented the aesthetic reconstruction of the affected tissues, the limitations were explained to the patient and the decision to fabricate a gingival prostheses was taken.
Both fixed and removable prostheses can be used for the treatment of large gingival defects. However, we chose to fabricate a removable acrylic resin gingival prosthesis as it is easier to fabricate and repair and is cost effective. In addition, missing tissues can be replaced without disturbing the other dental units. After the delivery of the prosthesis, the patient was satisfied with the treatment outcome.

References