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Temporomandibular Joint Ankylosis in A Child: Uncommon Etiology "Buckley's Syndrome"

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Abstract **Case Report**

Temporomandibular joint (TMJ) ankylosis is characterized by bone and/or fibrous union between the condyle and the glenoid fossa, restricting joint movements. The etiology of ankylosis may be related to infectious processes. We present the case of the bilateral temporomandibular joint ankylosis in Buckley's syndrome. It is suggested to be ankylosis secondary to recurrent arthritis of the temporomandibular joint. The treatment was surgical include arthroplasty of the joint cavity, coronoidectomy and reconstruction with autogenous costochondral rib graft.

Keywords: Temporomandibular joint- ankylosis- arthritis- Buckley's syndrome.

INTRODUCTION

Ankylosis of the temporomandibular joint (TMJ) is defined as a bony or fibrous adhesion of the anatomic joint components between the condyle and the cranial base, resulting in loss of function [1]. Ankylosis is most associated with trauma, local or systemic infection, or systemic disease [2]. Buckley's syndrome is characterized by recurrent infections, chronic eczema resembling atopic dermatitis, and markedly elevated serum IgE concentrations [3]. This disease can also affect the skeleton and lead to bone and dental abnormalities.

The management is multidisciplinary which aims primarily at the treatment and prevention of infections associated with surgical treatment.

We present the case of the bilateral temporomandibular joint ankylosis in Buckley's syndrome. It is suggested to be ankylosis secondary to recurrent arthritis of the temporomandibular joint.

CASE REPORT

A 10-year-old girl, followed in the pediatric department for multiple abscesses: pulmonary, pericardial, hepatic, renal and osteitis of the hip joints complicated by fracture of the right greater trochanter and arthritis of the right knee related to hyper IgE Syndrome (HIES). She presents a limited mouth opening evolving for 1 year. The clinical examination objectified a reduced mobility of the two temporomandibular joints, mouth opening was limited to 1 cm without facial asymmetric. A facial computed tomography shows a bilateral TMJ ankylotic mass, the diagnosis of the temporomandibular joint ankylosis was retained. (Fig.1) The patient was admitted to the operating room, under general anesthesia, we performed a resection of the bilateral ankylotic mass with realization of a coronoidectomy, and reconstruction with an autogenous costochondral rib graft (Fig.2).

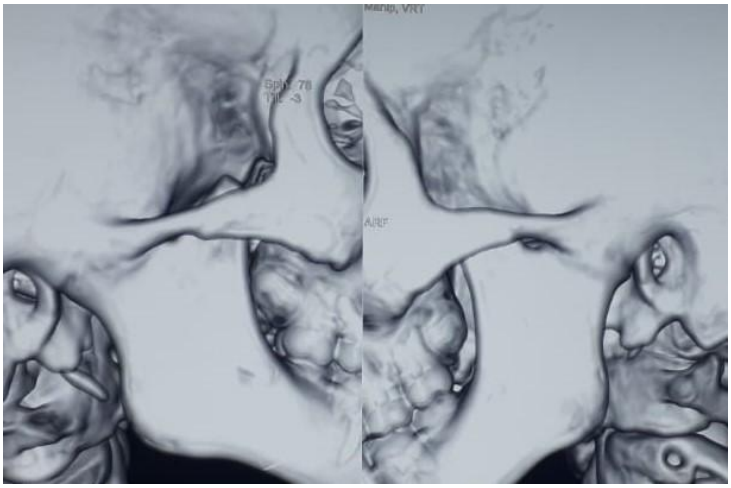

Fig-1: three- dimensional reconstruction of TMJ showing a bilateral fusion of the condylar head to the base of skull

Fig-2: Per operative image after removing the ankylotic mass then reconstruction with autogenous costochondral rib graft

DISCUSSION

Mandibular hypomobility results from a variety of disorders affecting the temporomandibular joint and surrounding structures. It may be classified by a combination of location intra- or extra articular (2). It usually develops before the age of 10 years but could develop at any age (4). Buckley's syndrome is a rare entity that manifests from birth or during infancy, it is defined by a triad: An elevated level of serum IgE, skin abscesses, recurrent pneumonia. The identification of a disease associating cutaneous and pulmonary staphylococci against a background of eczema dermatitis was described in 1966, called Job syndrome. Buckley describes the appearance of disseminated infections in the context of elevated total IgE in patients with facial abnormalities, namely hypertelorism and gross features (5). This disease can also affect the skeleton and lead to bone and dental abnormalities. The therapeutic strategies are essentially aimed at the treatment and prevention of skin and pulmonary infections and their complications. The occurrence of skin or lung abscesses requires prolonged antibiotic treatment and may require surgical management. To our knowledge we have not found a such case in the literature.

In Buckley's syndrome, the Recurrent infections appear from early childhood. All the sites are possible, but your most frequent are the skin and the respiratory tract with pneumonia. We also note bone and joint infections (6).

The etiology of ankylosis may be related to infectious processes, local traumatology, radiotherapy, and TMJ tumors. Among the consequences are the limitation of mouth opening, limited chewing capacity, difficulty in phonation, and oral hygiene [7]. This condition eventually causes aesthetic defects in the face, malocclusion, and facial malformation, particularly during childhood [8]. The characteristic images of computed tomography could be used to indicate the developmental stage of TMJ ankylosis and possibly help in identification of disease severity and treatment options [4].

The treatment of temporomandibular joint ankylosis is based on interpositional arthroplasty and reconstruction of the ramus-condyle unit with autogenous bone or alloplastic material [9].

CONCLUSION

The etiology of temporomandibular joint (TMJ) ankylosis may be related to infectious processes. In our case it is suggested that it is an ankylosis of the temporomandibular joint secondary to arthritis in the context of a Buckley syndrome. The treatment is surgical.

Competing interests

The authors declare no competing interests.

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