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Case Report

Dramatic Improvement of Iatrogenic Subcutaneous Atrophy After Steroid Temporomandibular Joint (TMJ) Injections: A Case Report

Chester Tyson[1], Dean Morrell[2*]

[1]UNC School of Medicine, Chapel Hill, NC, USA

[2]Department of Dermatology, UNC School of Medicine, Chapel Hill, NC, USA

***Correspondence author:** Dean Morrell, MD Department of Dermatology, UNC School of Medicine Chapel Hill, NC, USA; Email: morrell@med.unc.edu

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Abstract

Background: Intra-articular steroid injections are a common treatment of Temporomandibular Joint (TMJ) involvement in Juvenile Idiopathic Arthritis (JIA), effectively reducing inflammation and symptoms. However, regional subcutaneous atrophy is a possible complication following these injections. This can produce facial deformities and long-term cosmetic as well as functional consequences.

Case Presentation: We present the case of a pediatric patient with JIA including TMJ involvement who developed significant regional subcutaneous atrophy following bilateral TMJ intra-articular steroid injections. Her JIA was initially treated with methotrexate and then switched to abatacept for improved disease control. Notably, within months of the abatacept initiation, there was complete resolution of prior affected areas with no residual cutaneous or subcutaneous atrophy. Because corticosteroid injection induced cutaneous atrophy can be permanent, this case suggests the possible role of abatacept for the treatment of steroid- induced soft tissue atrophy in the setting of systemic treatment of JIA.

Conclusion: This case is the first to document abatacept's potential involvement in significant and rapid efficacy in resolving iatrogenic-induced subcutaneous atrophy following TMJ intra- articular steroid injections. Abatacept's mechanism of action of targeting T-cell activation and reducing inflammation may extend beyond arthritis treatment and into tissue repair. Further research is needed to explore abatacept's role in reversing steroid-induced atrophy and optimizing treatment strategies for pediatric patients experiencing this complication.

Keywords: Juvenile Idiopathic Arthritis; Temporomandibular Joint; Subcutaneous Atrophy; Intra-Articular Steroid Injection; Abatacept; Pediatric Dermatology

Abbreviations:

JIA: Juvenile Idiopathic Arthritis; TMJ: Temporomandibular Joint; MRI: Magnetic Resonance Imaging; ANA: Antinuclear Antibody; RF: Rheumatoid Factor; HLA-B27: Human Leukocyte Antigen B27; MTX: Methotrexate; MIO: Maximal Incisal Opening; CTLA-4: Cytotoxic T- Lymphocyte–Associated Antigen 4; IV: Intravenous

Introduction

Juvenile Idiopathic Arthritis (JIA) frequently involves the Temporomandibular Joint (TMJ) and can result in significant functional complications if not treated appropriately. Intra-articular corticosteroid injections are commonly used to control TMJ inflammation and help improve the symptoms. However, the injections can be associated with rare but potentially disabling adverse effects, including regional subcutaneous atrophy. This steroid-induced soft tissue atrophy can be permanent and lead to long term cosmetic and functional complications. We report a pediatric case of significant subcutaneous atrophy following bilateral TMJ intra-articular steroid injections with a subsequent complete resolution after the initiation of abatacept therapy, suggesting a potential therapeutic role for abatacept in reversing this complication.

Methodology

This case report was conducted through a retrospective review of the patient's medical record. Clinical data were obtained from pediatric rheumatology and dermatology encounters, including treatment history, physical examinations, laboratory results and imaging studies. Clinical photographs were obtained to document cutaneous findings and treatment response.

Case Report

This case details a female patient who first presented to her pediatrician at 18 months old with a six-week history of left knee swelling. The parents reported that the swelling had progressively worsened over the previous two to three weeks and had become painful with tenderness to palpation. There was no reported injury, warmth or redness and she experienced a single 24-hour febrile episode one week after the onset of symptoms. Her family history included Graves' disease in the maternal grandmother, but no autoimmune disorders such as rheumatoid arthritis, lupus or psoriasis. A referral to a pediatric rheumatologist was made for further evaluation.

At the rheumatology consultation, she had active arthritis in the bilateral knees, left ankle and left wrist, with restricted range of motion and a limp favoring the left side. Radiographs of the left knee revealed soft tissue swelling consistent with effusion. She was started on ibuprofen at 100 mg three times daily, with plans to consider intra-articular steroid injections. A follow-up visit two months later showed persistent arthritis in four joints, including swelling, effusion, restricted range of motion and a left knee flexion contracture. The family opted for intra-articular steroid injections, which were performed under general anesthesia in the bilateral knees, left wrist and left ankle.

The steroid injections resulted in significant but temporary improvement. At follow-up, she showed increased mobility, reduced swelling and absence of pain or tenderness. Laboratory studies confirmed pauciarticular Juvenile Idiopathic Arthritis (JIA), with positive ANA titers (1:160 speckled and 1:80 homogenous), negative RF and HLA-B27.

Despite initial improvement, arthritis persisted intermittently requiring additional rounds of intra-articular steroid injections into her knees and ankles. Further joint monitoring revealed additional flares over the following 4 years leading to methotrexate initiation at 25 mg subcutaneously weekly to manage persistent inflammation. Around this time, she had also undergone bilateral TMJ washouts and steroid injections due to new complaints of jaw pain. Methotrexate improved arthritis symptoms, but a bilateral facial rash extending into the preauricular scalp appeared 3 months after TMJ injections.

At this point, she was referred to pediatric dermatology for evaluation of the bilateral facial rash. The rash, located on the preauricular face and scalp and more prominent on the right side than the left, was characterized by dermally atrophic plaques with adherence to underlying bone (Fig. 1). There was no cutaneous induration in the affected areas. Magnetic resonance imaging (MRI) of the bilateral temporomandibular joints showed bony changes potentially consistent with morphea but no evidence of underlying joint disease. A diagnosis of localized scleroderma (morphea) involving the bilateral preauricular scalp was considered. Initial treatment included topical calcipotriene 0.005% cream and clobetasol 0.05% cream applied to the involved areas, as well as a three-day course of pulse methylprednisolone (500 mg) administered intravenously was completed. Despite these interventions and with her already receiving methotrexate 25 mg weekly subcutaneously, the lesions persisted, prompting the addition of abatacept at an initial dose of 250 mg intravenously every four weeks, alongside ongoing methotrexate therapy.

At her dermatology follow-up 1 month later, she showed improvement in her lesions following the initiation of abatacept. Examination of the bilateral preauricular scalp revealed atrophic plaques with adherent, bound-down tissue, but with much less erythema compared to her prior dermatology visit. Given the significant reduction in erythema, the use of topical calcipotriene and clobetasol creams was discontinued. She remained on methotrexate and abatacept for her juvenile idiopathic arthritis.

Three months later, her skin atrophy was much improved to resolving, with continued dermal improvement, less adherence and decreased erythema (Fig. 2). An MRI of the bilateral temporomandibular joints revealed no evidence of underlying disease. At a later dermatology visit, her prior cutaneous atrophy and alopecia were completely resolved (Fig. 3) and she remained on abatacept for her juvenile idiopathic arthritis.

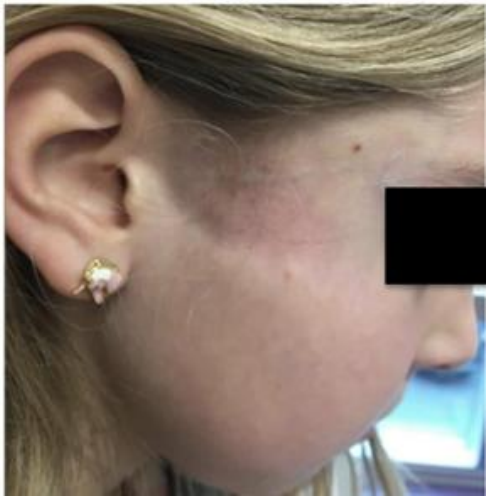

Figure 1: Atrophic plaques with some adherent to bone areas at bilateral pre-auricular scalp/face R>L (right side imaged).

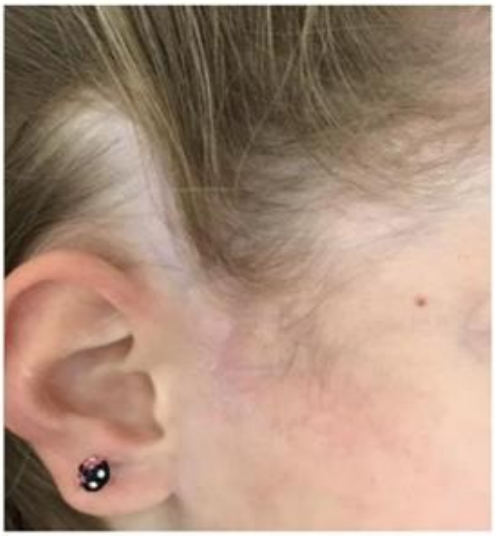

Figure 2: 3 months after abatacept initiation. Much improved and resolving atrophy involving the bilateral preauricular face and scalp (right side imaged).

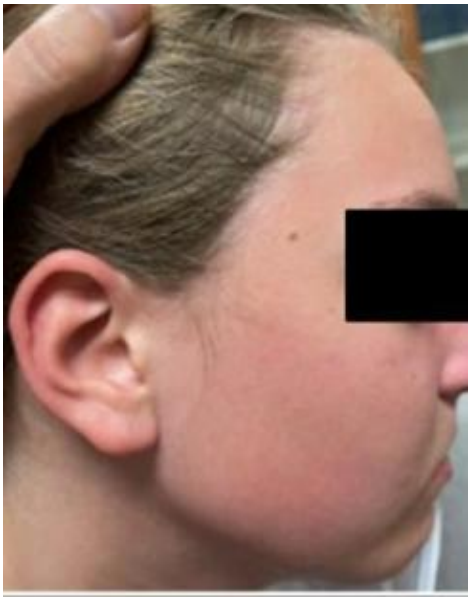

Figure 3: 3 years on continued abatacept use. Atrophy involving the face and preauricular scalp completely resolved (right side imaged).

Discussion

Temporomandibular Joint (TMJ) involvement commonly presents in a significant number of pediatric patients with Juvenile Idiopathic Arthritis (JIA). The prevalence of TMJ involvement differs among studies but is generally high. Based on a population-based cohort study, the cumulative incidence of TMJ involvement from the moment of JIA diagnosis until the transition to adult care is 30.1%. In addition, a retrospective chart review showed TMJ involvement in 34.9% of cases of JIA [1,2].

Involvement of the TMJ in pediatric patients who have JIA results from the inflammatory nature of the disease, which can affect any synovial joint, including the TMJ. The anatomical uniqueness of the TMJ makes it vulnerable to the effect of arthritic changes, which allows it to create possible deformities. It is especially at risk because the main growth center of the mandible is located within the condyle, with just a small fibrocartilage layer separating it from the joint space. There are many negative consequences of TMJ involvement in JIA including facial asymmetry, retrognathia (retreating jaw) and other deformities [3]. These may be due to possible damage of facial growth with the inflammation of the TMJ inhibiting the regular growth of the mandible. It can also lead to functional limitations as TMJ arthritis can cause restricted movement of the jaws, chewing problems, speech problems and chronic pain and discomfort. This can severely affect the quality of life and activities of daily living in pediatric patients [4,5].

Because of the asymptomatic nature regarding the diagnosis of TMJ involvement in JIA, as it is often left untreated from clinical assessment or physical examination alone, contrast-enhanced MRI becomes the diagnostic method of choice to assess and monitor TMJ disorders in those with JIA. This method is capable of demonstrating both soft tissue and osteochondral changes [6]. After diagnosis, intra-articular steroid injections are common treatment options for TMJ involvement in pediatric patients with JIA as they directly reduce intra-articular inflammation and consequently result in relief of symptoms. A retrospective study presented evidence that intra- articular steroid injections can improve the Maximal Incisal Opening (MIO), a component that measures jaw mobility. The MIO increased from 40.8 mm to 43.5 mm in one study after injections. Moreover, 51% of those with TMJ involvement showed MRI evidence of improvement of arthritic changes and of this percentage, 18% achieved complete resolution of TMJ arthritis [7]. Another retrospective chart review reported a mean 6.9 mm increase in MIO with a 3.8 mm increase in MIO following each intra-articular steroid injection [8-10].

However, subcutaneous atrophy remains a potential complication that accompanies these injections in pediatric patients with JIA. The thinned or lost subcutaneous fat tissue at the injection site creates an appearance of a depressed or indented part of the skin. The incidence of this complication can vary anywhere from one study reporting subcutaneous atrophy in <2% of patients to another reporting up to 38.6% of patients with injection site subcutaneous atrophy complications [8,9]. In a retrospective study investigating TMJ involvement with patients diagnosed with JIA, 12% of those identified were found to have adverse effects with either subcutaneous atrophy or intra-articular calcifications [10]. Although not pediatric, a unique presentation of a related case report revealed development of skin depigmentation and subcutaneous lipoatrophy post 3-week injection of methylprednisolone into the TMJ [11]. These complications can be quite burdensome with further facial deformity production and functional disabilities. These can become permanent or even require surgery for facial asymmetry as one did in the previously mentioned retrospective study [10].

Here we presented an additional case of a patient with JIA and TMJ involvement with an intra-articular steroid injection complication. Our patient, initially treated with methotrexate that partially improved arthritis symptoms, also underwent bilateral TMJ steroid injections due to new complaints of jaw pain. Three months later, a bilateral facial rash and subcutaneous atrophy appeared on the preauricular scalp due to injection complications (Fig. 1). However, transitioning to abatacept for improved arthritis control led to significant clinical improvement of both her JIA and cutaneous atrophy. Within three months of initiating abatacept therapy, the patient experienced a marked resolution of active skin lesions (Fig. 2). With continued abatacept use, her clinical outcomes included not only resolution of atrophy, but also complete regrowth of previously lost hair (Fig. 3). This is particularly remarkable as tissue atrophy as a complication from intra-articular steroid injections can be permanent.

This sustained response suggests that abatacept has a unique role in the immune mechanism as it works by targeting the T-cell costimulatory pathway, essential for activating T-cells and triggering inflammation. As a recombinant fusion protein, abatacept mimics the natural CTLA-4 molecule, blocking the interaction between CD80/CD86 on antigen-presenting cells and the CD28 receptor on T-cells. By preventing the full activation of T-cells, which play a crucial role in the inflammatory process, abatacept

reduces inflammation and limits collagen deposition in the skin and subcutaneous tissues. This mechanism has also proven effective in children with refractory pediatric localized scleroderma who have not responded to standard treatments like methotrexate, helping to reduce symptoms and improve previously thought-to-be permanent skin atrophy and musculoskeletal involvement [12,13].

To the best of our knowledge, this is the first report that supports abatacept's efficacy in reversing iatrogenically-induced subcutaneous atrophy after intra-articular steroid TMJ injections. The resolution of longstanding skin lesions and reversal of atrophy seen in this patient underscores abatacept's potential to not only halt disease activity but also repair damage previously thought irreversible. Additional research is needed to investigate the role of abatacept in treating iatrogenically-induced subcutaneous atrophy, with a focus on its potential to reverse atrophy and maintain sustained remission. Further retrospective studies and prospective trials should be done to investigate other possible intra-articular steroid injection complication resolutions with abatacept treatment and to help provide an understanding of the optimal dosing strategies and long-term efficacy and safety for this pediatric patient population.

Conclusion

This case highlights a rare, but clinically significant complication of intra-articular TMJ corticosteroid injections in a pediatric patient with JIA. It demontrates a complete resolution of this steroid-induced subcutaneous atrophy following the initiation of abatacept therapy. Because this corticosteroid-induced atrophy can be permanent, the rapid and sustained improvement observed in this patient suggests abatacept’s novel role beyond just disease control, but potentially contributing to soft tissue recovery. To our knowledge, this is the first report supporting abatacept’s efficacy in reversing injection-induced subcutaneous atrophy.

Further investigation is needed to clarify abatacept’s role in the treatment of iatrogenic subcutaneous atrophy and to guide management strategies for pediatric patients experiencing this complication.

Conflict of Interest

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

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Acknowledgement

None.

Data Availability Statement

Not applicable.

Ethical Statement

The project did not meet the definition of human subject research under the purview of the IRB according to federal regulations and therefore, was exempt.

Informed Consent Statement

Informed consent was taken for this study.

Authors’ Contributions

All authors have contributed equally to this work and have reviewed and approved the final manuscript for publication.

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