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RESEARCH ARTICLE

EROSIVE TEMPOROMANDIBULAR JOINT DISORDER IN A PATIENT WITH ULCERATIVE COLITIS: RARE MANIFESTATION OF ARTHROPATHIES, CASE REPORT

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Abstract

Temporomandibular Joint (TMJ) involvement in patients with ulcerative colitis (UC) is a rare manifestation. To the best of our knowledge there are only two cases in the literature reported the presence of TMJ involvement in a patient with UC. We report a case of 23-year-old female with a two-year history of UC she was complaining of unilateral temporomandibular jaw pain, the pain was severe enough to affect her daily activities, she was not able to speak and there was a limitation of mouth opening. The patient was only able to open her mouth for 2.5 cm. C-reactive protein and erythrocyte sedimentation rate were high, rheumatoid factor was negative. The computed tomography (CT) of the mandible and teeth reveals flattening with erosion of the condylar process of the left mandibular joint without the presence of joint effusion or soft tissue swelling. Our patient was managed by biological treatment Ustekinumab for 6 months, as a result the patient improved and she was able to open her mouth up to 4 cm. Clinicians should be aware of the importance of biological treatment in these circumstances, TMJ arthritis in UC patients is considered a rare and unusual picture of extraintesinal IBD manfistations.

Introduction:-

Temporomandibular joints (TMJs)inflammation can cause pain and limited movement of the jawand occurs in various chronic inflammatory rheumatoid conditions (1). Such as Rheumatoid Arthritis (RA), ankylosing spondylitis (AS) and psoriatic arthritis (PA). Clinical evidence of TMJs involvement in patients with RAis between 19 to 86%and commonly comes with TMJspain, locking and reducesmouthopening (2,3). While AS is involved in 10 to 24% of casesand most clinical features is TMJs pain, stiffness and tenderness (4). However, PA involved TMJs is rare to happen, only 43 cases in 19 articles were reported (5).Up to the best of our knowledge only two cases in the

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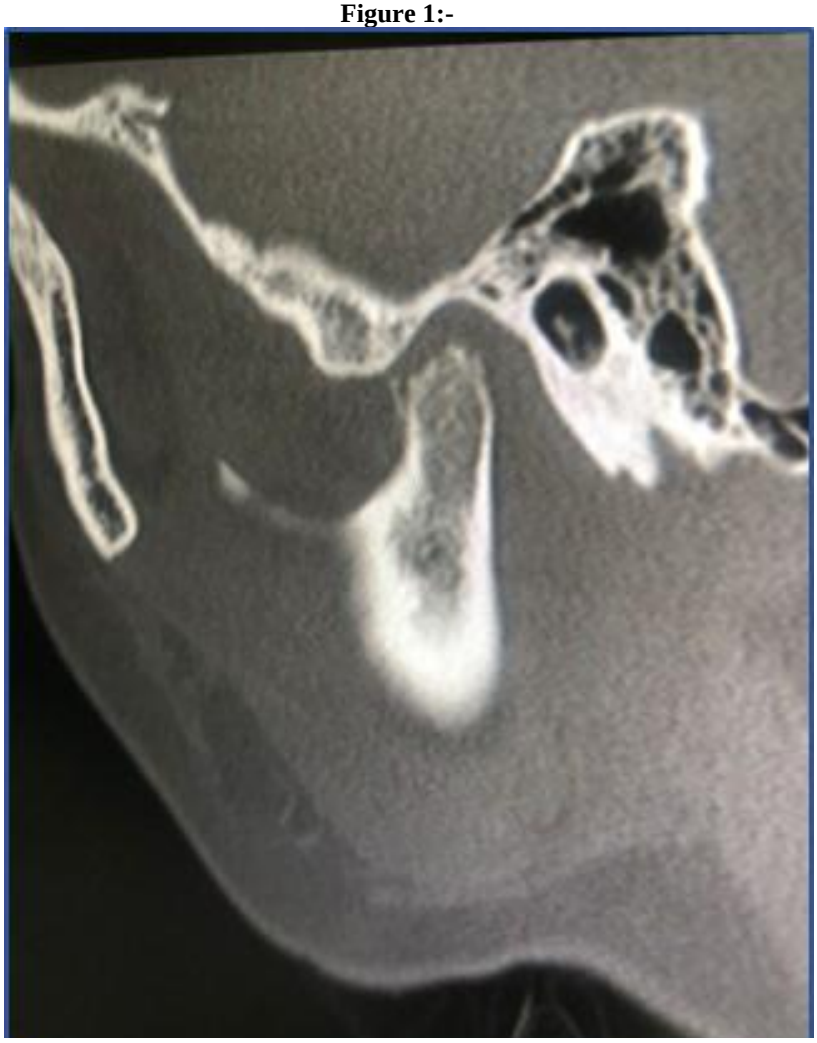
literature reported involvement of TMJs in patients withUlcerative colitis (1,6).In this report we are describing a caseof a patient with Extra-Intestinal Inflammatory Bowel Disease Who developed acute TMJs arthritis.

Case presentation

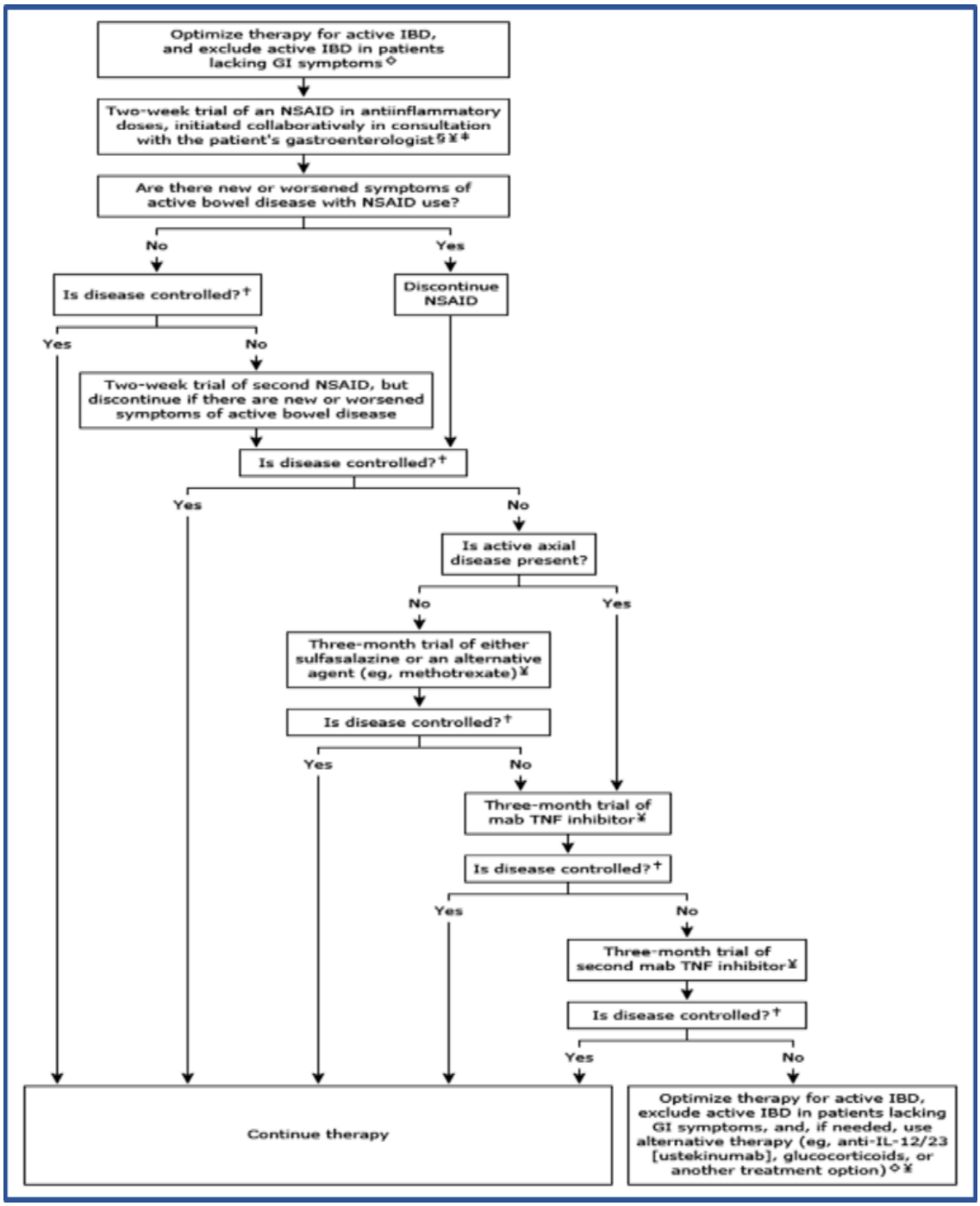
A case of 23 year old Saudi female patient known case of ulcerative colitis (UC) diagnosed by colonoscopy biopsy back to October 2019, presented to the Outpatient rheumatology Clinic with History of 7 days of Temporomandibular jaw pain and asymmetrical oligoarthritis (Right hand and Left knee). The left jaw pain was continues, progressive in nature, stabbing in character, localized in the left side with radiation to temporal area , severe enough to affect her daily activity ,she was not able to speak , eat or even drink due to narrow mouth opening , pain was aggravated by eating relieved partially by non-steroidal anti-inflammatory drugs (NSAIDS) , lasting for more than 30 mins with no history of trauma. She had never experienced similar manifestation like this before. The patient complained of morning stiffness last more than 2 hours on the small joints of her right hand including DIP , PIP and MCP not associated with redness or swelling, no aggravating or relieving factors.HerLeft knee pain was 7 out of 10 in severity with morning stiffness last for more than 1 hour aggravated by rest , relived by NSAIDS and movement , no redness or swelling and no history of trauma. The patient also experienced low back pain which was on and off with morning stiffness for 10 minutes, progressive, aggravated by rest , relived by NSAIDS and daily movement , no history of trauma ,fecal incontinence , urinary incontinence , weakness or sensory loss. Her symptoms also accompanied with erythematous painful rash on her extensor aspects of her left leg, associated with swelling, no ulcer, not oozing any blood or discharge (Erythema nodosum). There was insignificant history of weight loss less than 10% in the last 6 months from 52 kg to 47 Kg. The patient denies fever, anorexia, night sweating, lymph node enlargement, dark or pale urine, yellowish skin discoloration. Her past medical history is remarkable for two previous admissions, the first one was in October 2019 when she presented with bloody chronic diarrhea, after workup and colonoscopy she was diagnosed with UC confirmed by Biopsy , second admission was in September 2020 due to UC acute flare when she received course of 3 weeks of oral steroids. Family history is positive for autoimmune Hashimoto hypothyroidism and Diabetes mellitus and negative for rheumatoid arthritis, cancer or cardiac diseases. She has a history of blood transfusion of 2 packed RBC in here first admission back in 2019 due to Iron Deficiency anemia caused by UC , not followed by any acute complication of transfusion. Her current medications are oral Tablets Mesalamine 2 g twice daily, infliximab subcutaneous injection 40 mg once per 4 weeks, iron tablets of ferrous sulfate 190 mg po once a day, with good compliance to her medication. Her systemic review, past surgical history, allergic and travelling history were unremarkable.

On general examination the patient looks ill , mobilized , walking and talking , average body built , not cyanosed , pale or jaundiced , not attached to anything with no obvious deformity noted. Vitaly she was stable with Temp of 37.3 C , BP : 124/71 , HR : 81 B/M , O2 saturation RA 99% , RR : 13/m , random Blood Glucose was 127 mg/dl , GCS of total 15/15 , alert conscious oriented to time, place and persons, cooperative and responsive. On local examination her mouth can not be examined properly because the patient was unable to open her mouth fully, While examining her mouth, patient had moderate tenderness over the left Temporomandibular Joint. there was swelling but no redness with decreased range of motion. Her limb examination was remarkable for 2^{nd} and 3^{rd}MCP , PIP , DIP tenderness without effusion in her right hand , and left knee tenderness without effusion. Other examinations were normal. We ordered labs for the patient such as CBC, iron studies, inflammatory markers , and chemistry. The lab results were normal except for the CBC, iron studies and inflammatory markers which showed iron deficiency anemia and elevated inflammatory markers. Additionally rheumatological work up were done and it was negative RF , ANA and Anti-DS DNA . For the imaging we performed a CT scan of the mandible and teeth and it showed flattening with erosion of the condylar process of the left mandibular joint, and there was No obvious joint effusion or soft tissue swelling. Likely represent arthropathy, infectious process or trauma (figure 1).

Figure 1:-



The recommended treatment for this patient is early physiotherapy because it would be beneficial, and Control of the underlying IBD should also be optimized in patients with arthritis in collaboration with the patient's gastroenterologist, NSAIDS continuous administration may modify disease course but may exacerbate IBD , NSAIDS should be administered together with proton pump inhibitor . Intraarticular corticosteroid in mono or oligoarthritis after excluding septic arthritis. Anti-TNFs (adalimumab or infliximab) they are Effective for both axial and peripheral manifestations, improves function and may slow progression of structural changes, Sowe startedourpatientonUstekinumabasshedevelopedsecondaryfaluire fromInfleximab. Note that Anti-IL12/23(Ustekinumab): works effisiontly for peripheral arthritis in IBD patient with less effectiveness on axial arthropathies .



Our patient at the beginning she was only able to open her mouth for only 2.5 cm(figure 2A), after 3months of Ustekinumab biological treatment she got improved and started to open her mouth up to 3 cm (figure 2B) , after 6 months of treatment she started to open her mouth up to 4cm (figure 2C).



(Figure 2A):- **(Figure 2B):-** **(Figure 2C):-**

Discussion:-

The diagnosis of ulcerative colitis associated with spondyloarthropathy in our case was based on clinical features, Colonoscopy biopsy, and radiographic findings. Although there is an increased rate of family history among UC patients (7), our patient reported no family history of the same diagnosis. However, there was positive family history of Hashimoto's disease. due to the affect of spondylitis on the tempo-mandibular joint, patient was unable to fully open her mouth. On imaging, there was flattening with erosion of the condylar process of the left mandibular joint. Consistently, a previously reported case of psoriatic arthritis complained of the inability to open his mouth completely due to the involvement of TMJ which displayed bilateral condyle ankylosis on CT scan (5).

It is estimated that 46% of IBD patients develop musculoskeletal manifestations (8). Peripheral arthritis representing 5 to 20% while spondyloarthropathies occur among up to 25% (9). Multiple cases reported TMJ involvement with numerous types of arthritis (1,5,10). Our case is one of the few cases that report the axial involvement of TMJ in spondylarthritis associated with UC.

Since the first aim of management is to relieve pain (3), non-steroidal anti-inflammatory drugs (NSAIDs) was used by the patient which was partially effective in relieving her pain. Also, the patient was started on Ustekinumab as she developed secondary failure from infliximab. An improvement was noticed as the opening of her mouth increased from 2.5 cm to 4 cm after 6 months. Futher follow up of disease activity and Extraintenialmanfistation is needed.

Conclusion:-

We reported in this study a case of TMJ Spondyloartheritis diagnosed in 23 year old female who is a known case for ulcerative colitis. The patient improved after 3 months of Ustekinumab biological treatment as she previously developed a secondary failure to infliximab.

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Informed consent:
A written informed consent was obtained from the patient for this case report publication.

Declaration of interest:
Authors have no conflict of interest to be declared.

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Data availability statement:
All the data related to this study is available upon request.

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