A PAINFUL PUZZLE:

Piecing Together Sacroiliitis and Mucosal Ulcers

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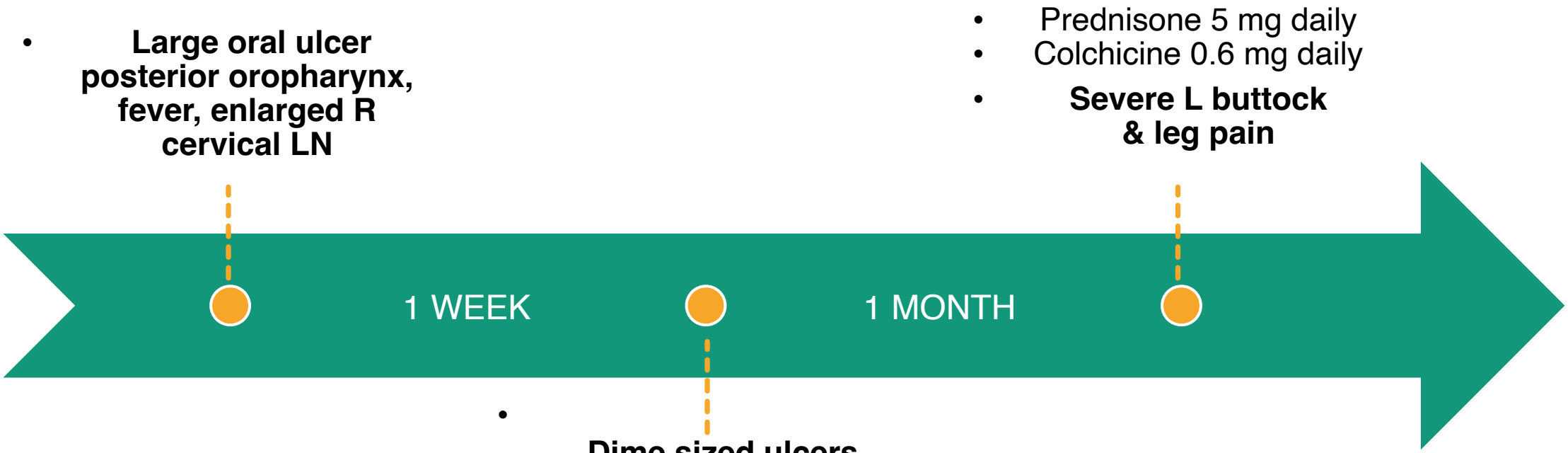
GRAND ROUND 3 TIN SOLDIERS GLOBAL

PATIENT

16-year-old F
with longstanding history of painful non-scarring oral ulcers inside bottom lip, side of cheeks, nearly always present.

- Seen by GI – felt ulcers familial (celiac and IBD ruled out w/ screening labs)
- Other PMHx: asthma, constipation
- Not sexually active
- FHx: Lebanese ethnicity, parents non consanguineous
  - Mother had history of oral ulcers in youth

CLINICAL PRESENTATION

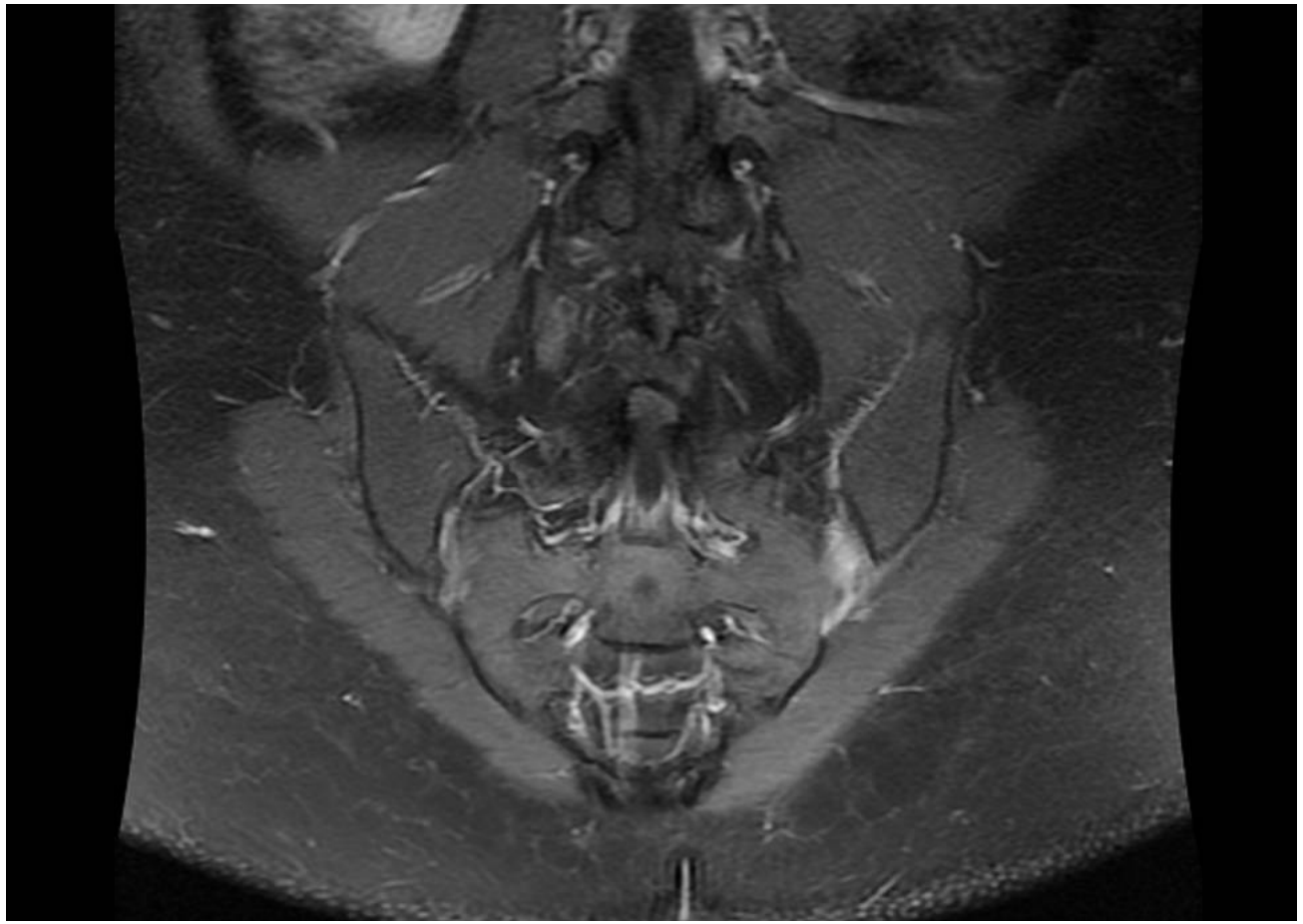


INVESTIGATIONS

- WBC 10.7, **Hgb 99**, plts 305
- **ESR 56, CRP 64.8**
- Ferritin 67, **GGT 73, ALT 66**, AST 18, albumin 37, creat 35
- **Iron 3**, CK/LDH normal, IgGAM normal
- ANA neg, TTG neg, HLAB51 neg, HLAB27 neg
- Infectious work up
  - HSV swab neg, C Trachomatis DNA neg, N Gonorrhea DNA neg, Trichomonas neg, Yersinia serology neg
  - CMV IgG reactive, IgM neg, EBV IgM/G neg, HIV serology neg
  - Resp virus neg, mycoplasma pneumonia PCR neg

INVESTIGATIONS

- **MRI L Spine / SI joints:**
  - Inflammatory changes to anterior and inferior portion of left sacroiliac joint with abnormal enhancement of mild fluid and associated signs of myositis involving the left piriformis muscle
  - No frank abscess, erosive changes or bone marrow edema
  - No sign of sacroiliitis on R side
  - Lumbar spine normal



- Differential diagnosis?
- Additional testing required?
- Treatment considerations?

- **Differential diagnosis?**
  - Lipschutz ulcer, Behcet disease, IBD, spondyloarthropathy, other
- **Additional testing required?**
  - Fecal calpro, Endoscopy, MRE
- **Treatment considerations?**
  - Stay on prednisone 5 mg

COURSE

- Patient admitted for severe pain crisis, requiring opioids, pain continuing to escalate.

DAY 5 of admission

- Fever, mobile CXR poor inspiration, questionable infiltrate vs atelectasis in RUL. Viral panel, blood culture pending. CRP 159.
- Started IV ampicillin
- Upper and lower endoscopy – macroscopically normal appearance.

DAY 6 of admission

- Due to severe pain, offered pulse methylprednisolone with plan for anti-TNF therapy
- Low grade fever

COURSE

DAY 8 of admission

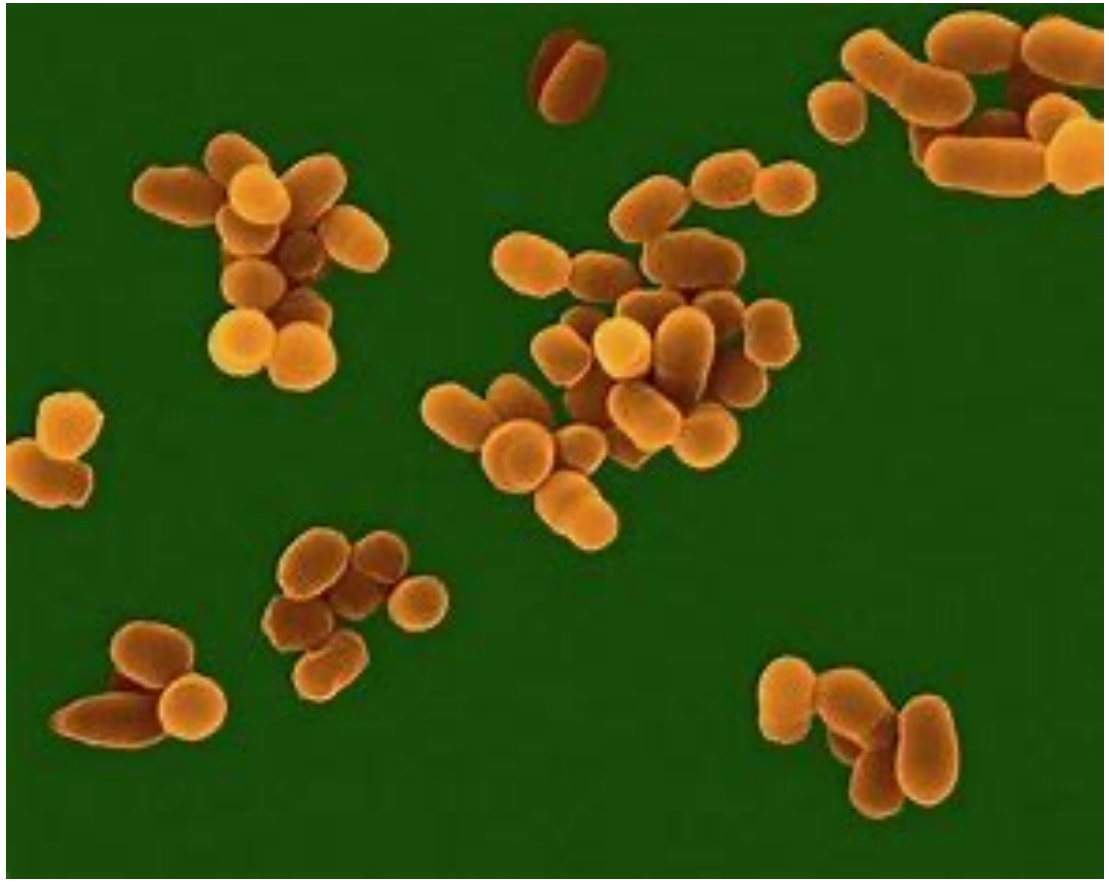
- 3 days of pulse IVMP, some improvement in pain. CRP 45.
- Psych support and rehabilitation
- In evening, report +gram stain (>72 h) for gram positive cocci, vancomycin added. Anti-TNF on hold. Steroid stepped down to pred 50 mg daily.

DAY 10 of admission

- MRE incomplete due to constipation, fecal calpro 377
- Microbiology lab reports gram negative cocci, speciated as Brucella Melitensis (>120 h).
- Started on iv gentamicin x 1week, rifampin + doxycycline x 6 mos
- Rapid prednisone taper, continued colchicine

BRUCELLA MELITENSIS

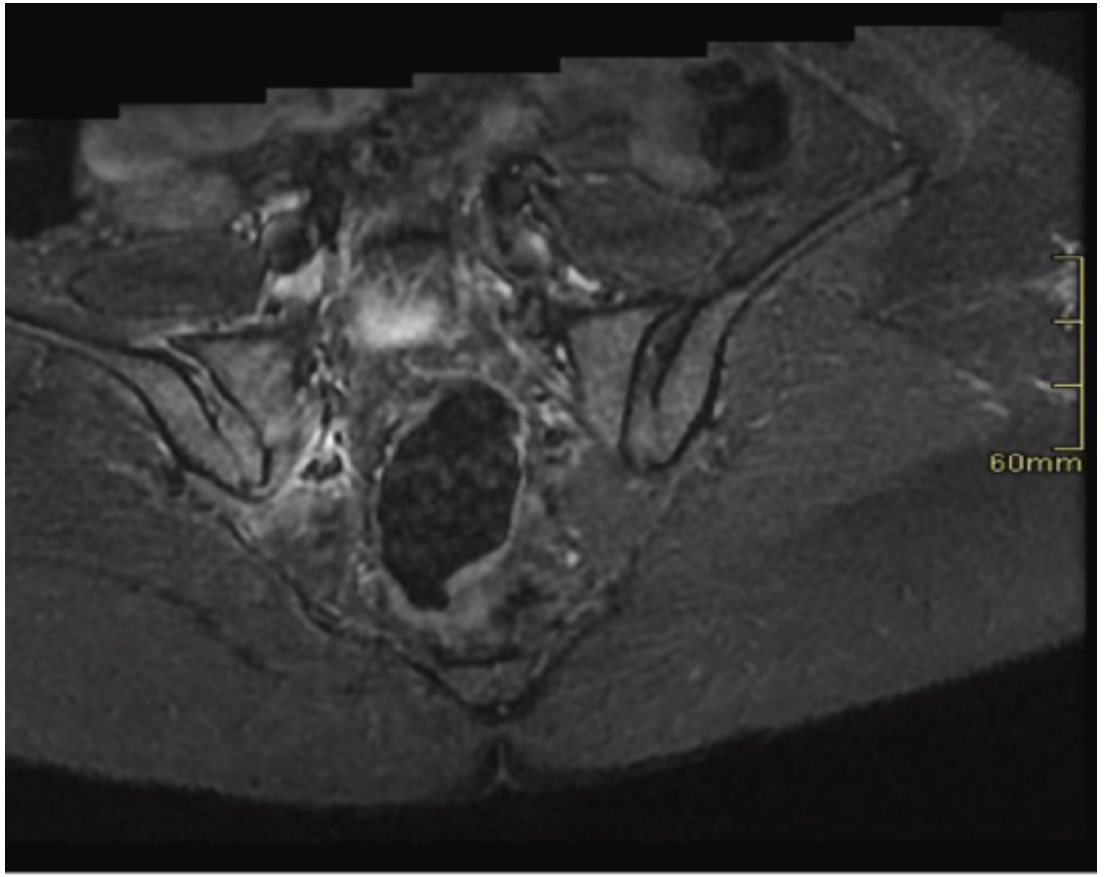
- Gram negative coccobacilli
- Mainly infect cattle, swine, goats, sheep and dogs
- Acquired through direct contact with infected animals, eating or drinking contaminated animal products or inhaling airborne agents. Most cases caused by ingesting **unpasteurized milk/ cheese from infected goats or sheep. Common in Middle East.**
- Typically causes flu-like symptoms (fever, weakness, malaise and weight loss). However, may present in many atypical forms. In many patients the symptoms are mild and, therefore, the diagnosis may not be considered.



WHO.int: Brucellosis

BRUCELLA MELITENSIS

- Prevalence of skeletal complications of brucellosis reported 11-85%
- Sacroiliitis is most common affected site in adults
- Peripheral arthritis more common in children (mono/oligo articular, hip/ knee/ ankle)
- Lumbar spondylitis also reported
- Case of R sacroiliitis & piriformis myositis recently published



Sanaei Dashti A, Karimi A. Skeletal Involvement of Brucella melitensis in Children: A Systematic Review. Iran J Med Sci. 2013 Dec;38(4):286-92.
Sandakly N, El Koubayati G, Ayoub A, Haddad F. Brucellosis complicated by piriformis myositis and sacroiliitis: A case report and a review of the literature. Heliyon. 2024 Mar 27;10(7):e28617.

ULCERS

- **No report** of Brucella melitensis associated **with oral or GU ulcers**
- **Lipschütz ulcers**
  - Non-sexually transmitted genital ulcers on vulva, primarily affect young women and adolescents. May be linked to EBV, CMV, Mycoplasma.
  - Appear suddenly, often accompanied by flu-like symptoms such as fever, fatigue and swollen lymph nodes.
  - Self-resolving. Steroids in severe cases.
- **Genetics**
  - Targeted sequencing of TNFAIP3 and RELA: Negative.
  - Chromosomal microarray: arr(X,1-22)x2 (normal result)

BEHCET?

- Recurrent oral ulcers – **did not improve** with colchicine even at 1.8 mg daily
- GU ulcers did not recur
- Ophtho exam: no intraocular inflammation, mild blepharitis
- No rash
- No pathergy

- Improvement of sacroiliitis on serial imaging.
  Unfortunately still has chronic pain and many non specific symptoms.

TAKE HOME MESSAGES

- There may be more than one process going on
- Consider brucellosis as a cause of infectious arthritis in patients who have been to endemic areas
- Not all cases of oral and genital ulcers are Behcet disease!

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THANK YOU

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