

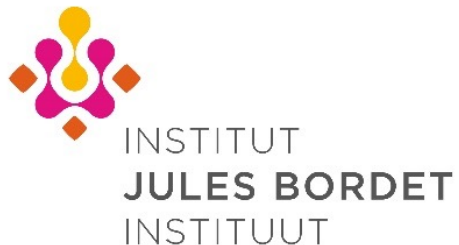


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ACADEMISCH ZIEKENHUIS
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Vanishing hypermetabolic bone lesions : a challenging diagnostic case study.

Hugues Azema , Celine Sergeant , Bruno Couturier ,Carlos Artigas,
Anne-Leen Deleu Patrick Flamen .



Hugues Azema

Who ?

- Female : 53 years old
- Medical history: hypothyroidism and diverticulosis
- Lifestyle : nurse, no smoking, no drinking
- Medication : L-thyroxine, Pantoprazole

Orthopedic consultation :

- Main complaint : **left coxalgia for several months**
- Systemic anamnesis : normal ✓
- Pelvic X-ray : normal ✓
- Initial diagnosis : trochanteritis
- Suggested treatment : physiotherapy + painkiller



NO RESPONSE

PELVIC MRI

Multifocal bone lesions with T2 hypersignal

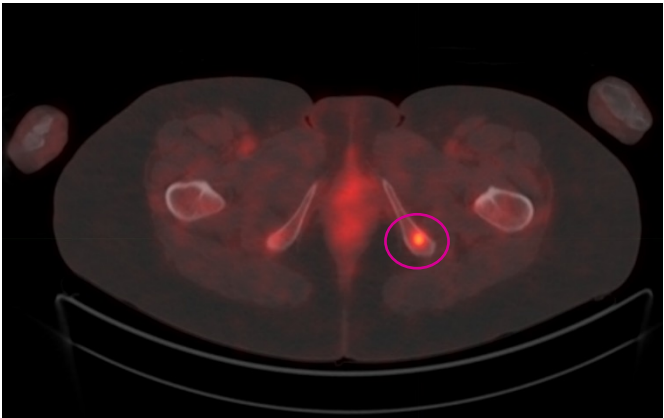
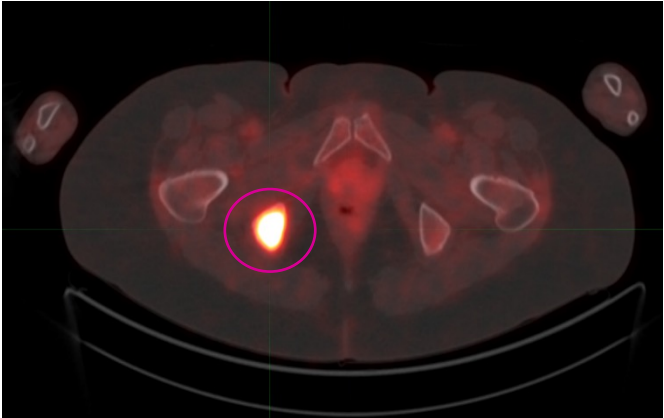


NEOPLASTIC ? OTHER ?

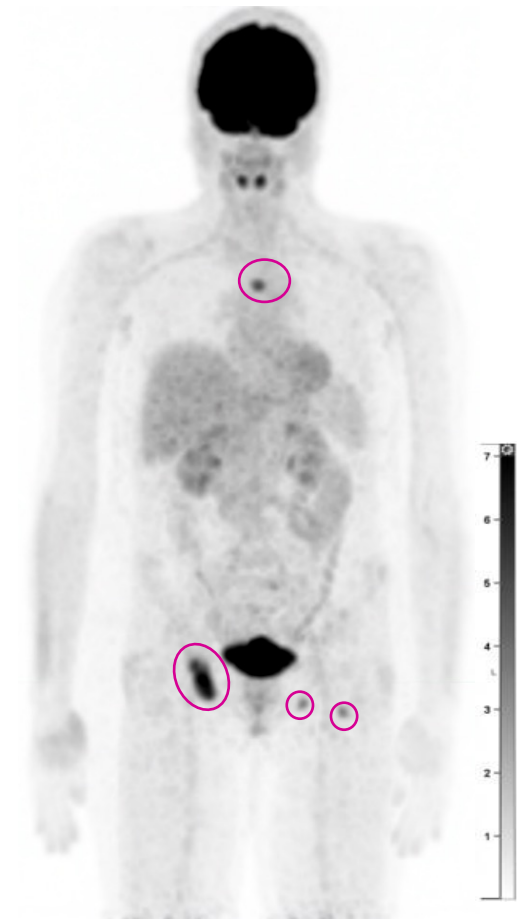
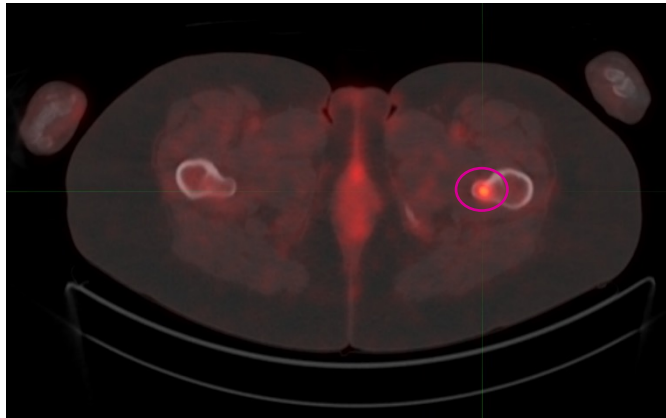
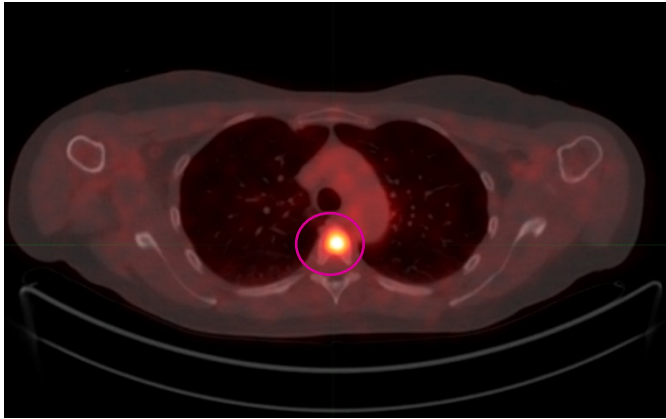
¹⁸FDG-PET/CT

Intensely hypermetabolic bone lesions

Pelvic lesions



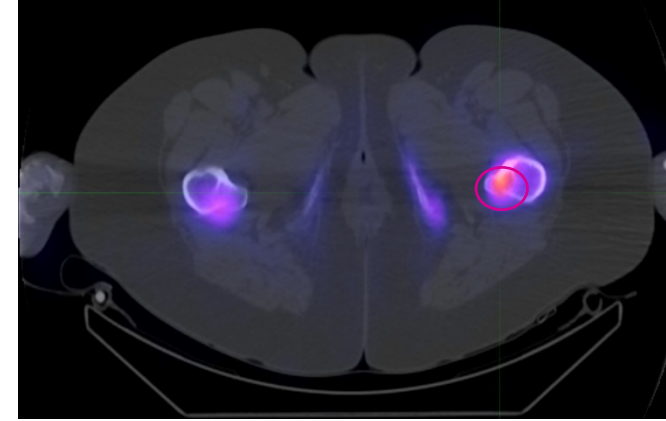
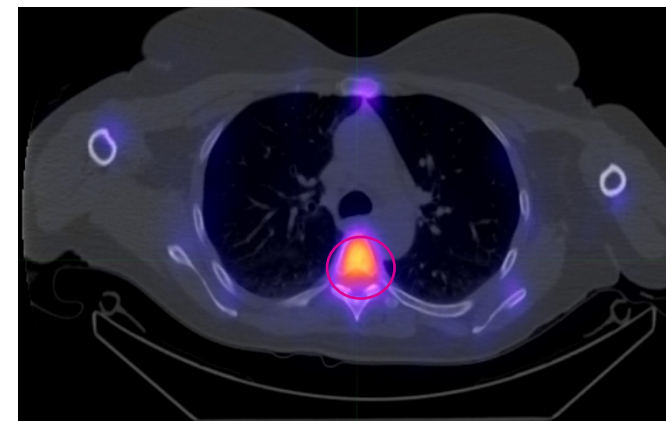
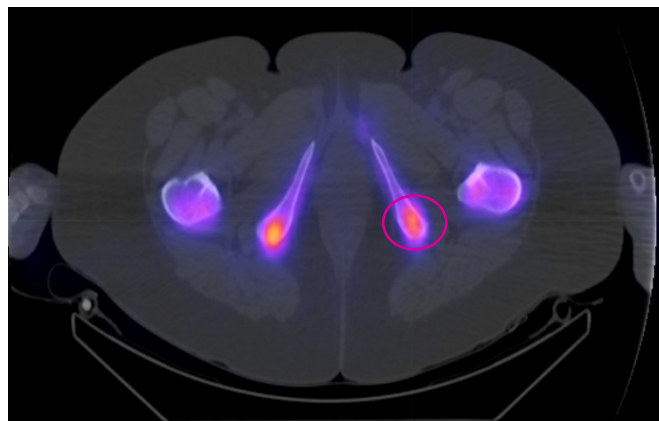
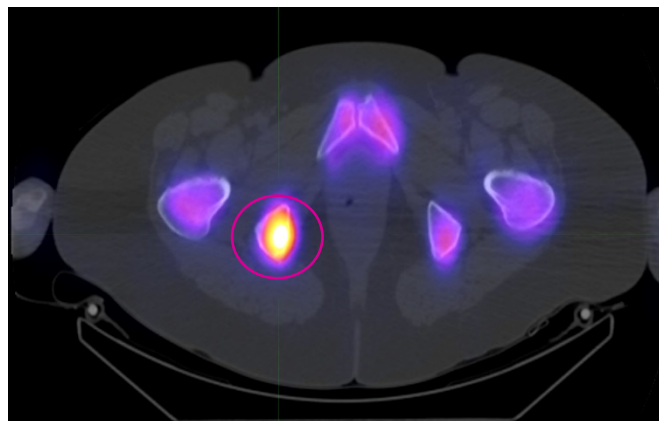
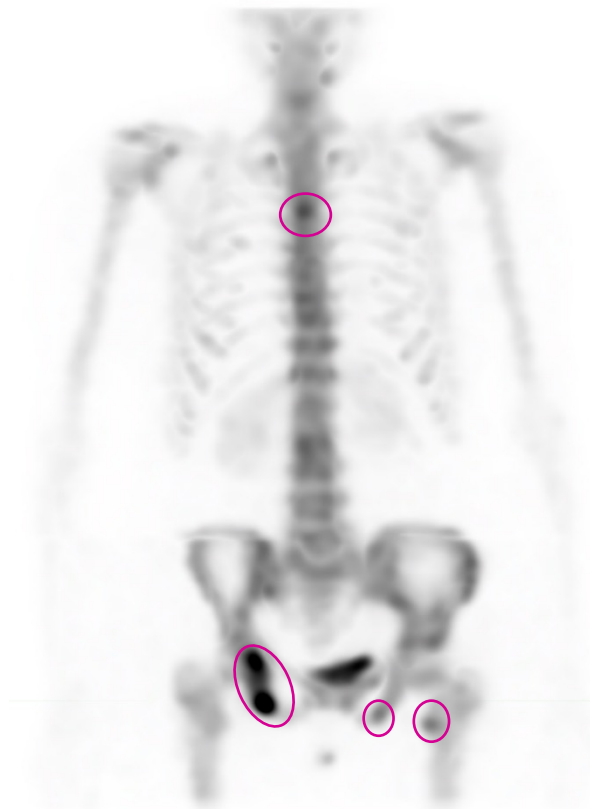
D5 & left trochanter



Positives bones lesions

Pelvic lesions

D5 & left trochanter





- ~~1/ Metastases from solid neoplasia : breast, lung, kidney, melanoma~~
- ~~2/ Hematological diseases : multiple myeloma, lymphoma~~
- ~~3/ Systemic diseases : sarcoidosis, histiocytosis, Paget~~
- ~~4/ Infectious diseases : tuberculosis, infectious osteomyelitis~~

Clinical examination :

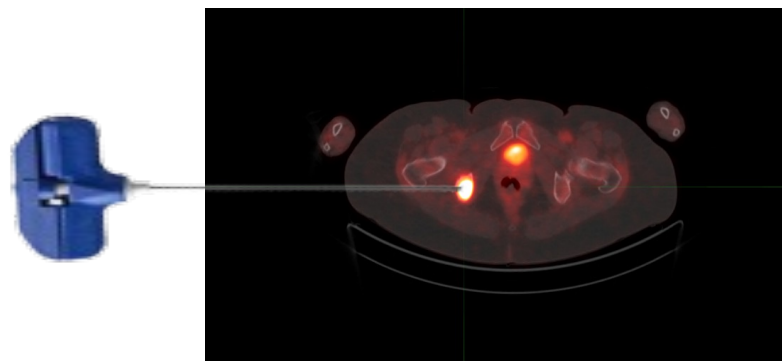
- Parameters : ✓
- C-P : ✓
- Abdo : ✓
- Locomotor : ✓
- Teguments : ✓
- Senological : ✓
- Lymph node areas : ✓

Blood test :

- Tumor markers (CA 15-3, CEA) : ✓
- Hemato (Protein electrophoresis, K/Lambda and urinary Bence Jones) : ✓
- Systemic disease (CRP, ESR, calcium and phosphorus, VIT D, PTH, ALP, ACE) : ✓
- Infectious disease (CRP, IDR) : ✓

Anapath & Microbio :

- **Non-specific inflammatory infiltrate**
- Neoplastic cells : no ✓
- Granuloma : no ✓
- Direct examination : neg ✓
- Cultures : neg ✓
- Ziehl and Grocott staining : neg ✓



Imaging follow-up

Stable T2 hypersignal multifocal bone lesions

3 Month F-U

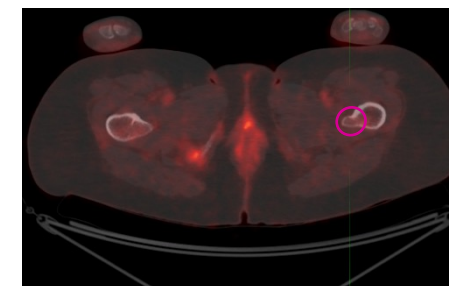
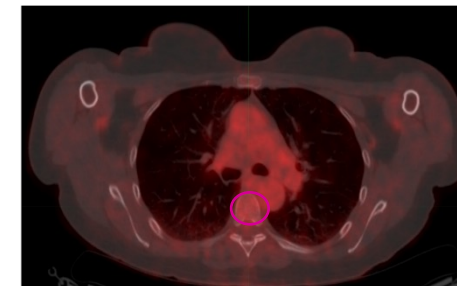
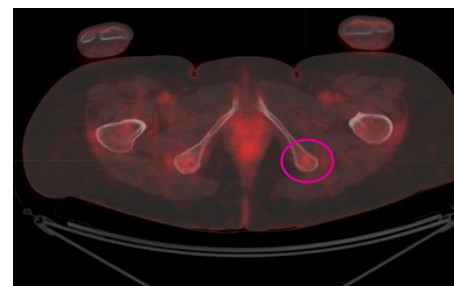
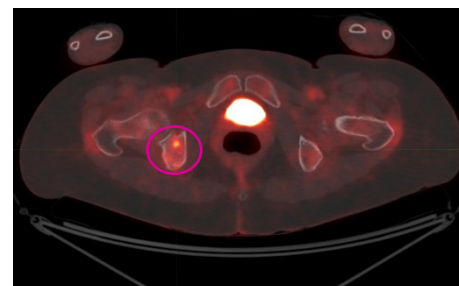


Baseline

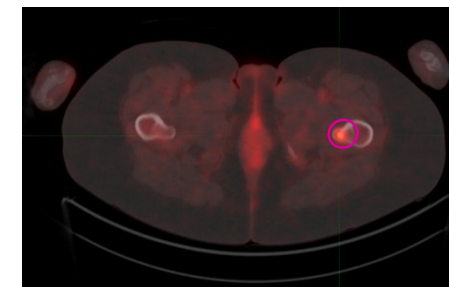
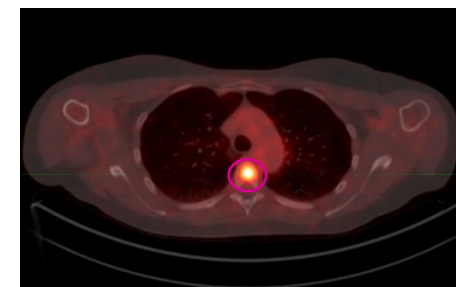
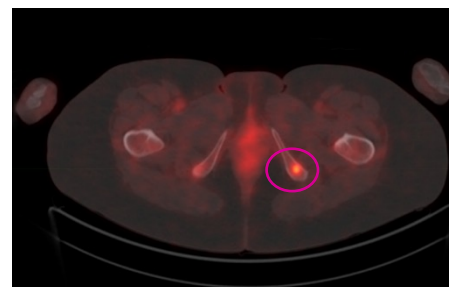
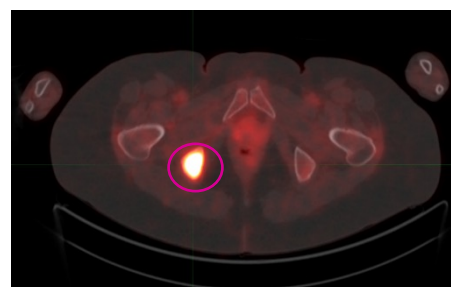
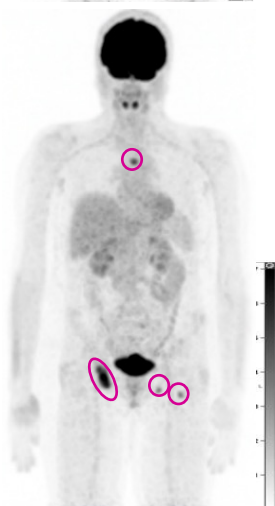


Vanishing hypermetabolic bone lesions

3 Month F-U



Baseline



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Vanishing hypermetabolic bone lesions



INTERNAL MEDICINE CONSULTATION

***CHRONIC NON-BACTERIAL
OSTEOMYELITIS (CNO)***

Definition :

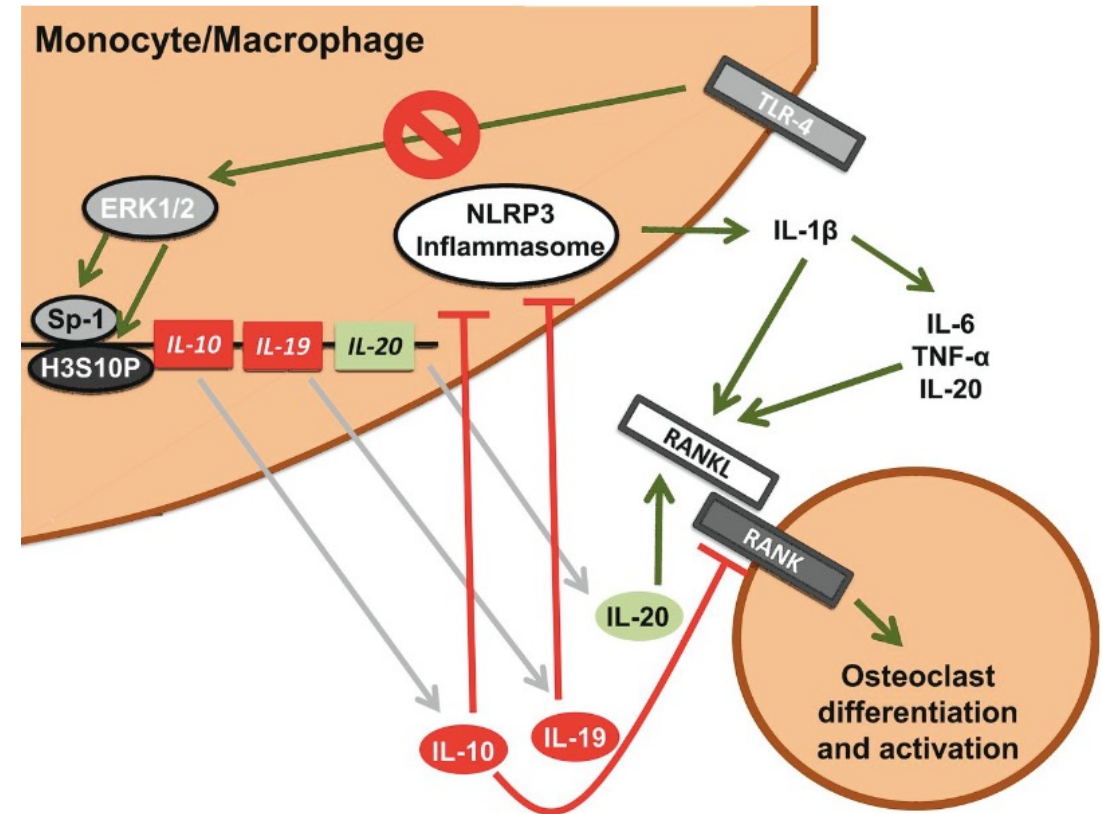
- First described in 1972
- **A chronic, sterile, auto-inflammatory multifocal bone disorder**
- Also known as chronic recurrent multifocal osteomyelitis (CRMO)
- More a pediatric disease

Epidemiology :

- **Rare** condition : 4/1M
- Underdiagnosed and underreported

Physiopathology :

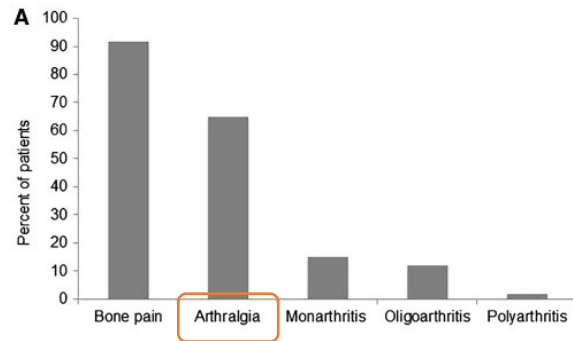
- **Auto-inflammatory** with decreased expression of **anti-inflammatory cytokines** and increased expression of **proinflammatory cytokines**.
- Infectious trigger ?
- Genetic predisposition ?



German incidence surveillance study, 2011

Clinical aspects :

- **100% Musculoskeletal problems**, 19% mucocutaneous manifestations, 8% gastrointestinal manifestations, 3% lymphoid 2% ocular manifestations, 1% cardiac manifestations)



Blood test :

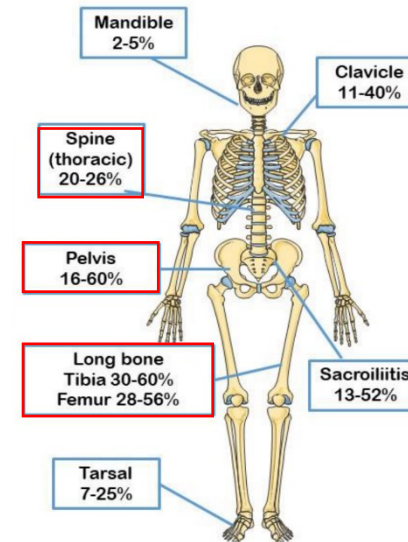
- Inflammatory parameters **normal or elevated** (50%ESR , 60%CRP, 15%WBC)

Histopathology :

- **Non specific** (coexistence of acute & chronic inflammation, fibrosis and bone sclerosis)

Number & Distribution of the lesions:

- **Mean number of 4 lesions** at diagnosis
- Metaphyseal > **Pelvic** > **Vertebral** > Epiphyseal > Clavicle > Diaphyseal > Cranial > Carpal



Girschik et al, 2018

bone inflammation/bone erosion/hyperostosis

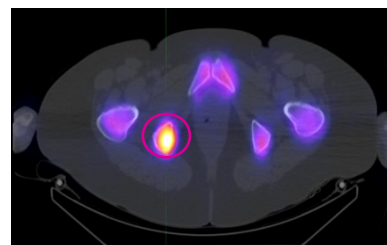
MRI :

- Hypersignal T2 -> Marrow edema



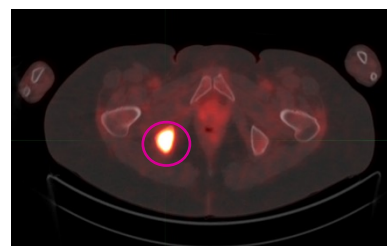
BONE SCAN :

- Bone remodeling

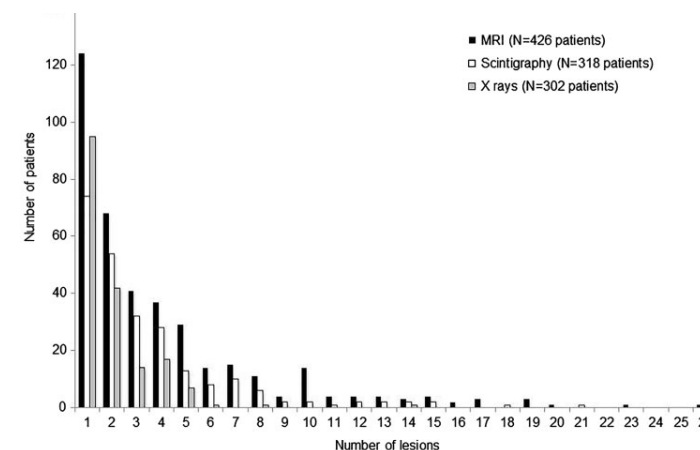


¹⁸FDG-PET/CT :

- Hypermetabolism in active phase



Study N = 486 cases (Euro fever international registry)



Girschik et al, 2018

- ¹⁸FDG-PET/CT was **not included in the study**
- Only a **few cases** reported with ¹⁸FDG-PET/CT
- **First case with 2 concomitant MRI & PET-CT during two different phases**

Challenging diagnosis

- Non specific symptoms/biomarker/imaging features/histopathology data.
- Mean time **from symptoms onset to diagnosis : 1 year**
- A **diagnosis of exclusion** which associates clinical/imaging/histopathological parameters

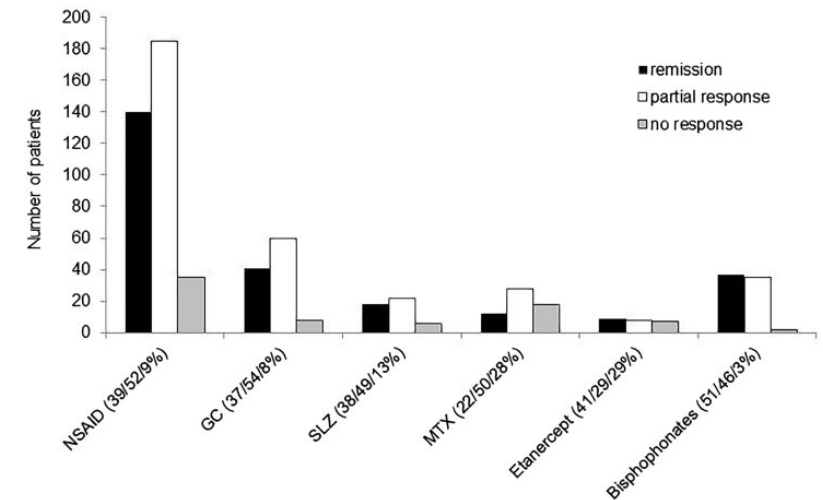


Fig. 4 Response categories by different therapeutic modalities

Treatments :

- **First line** : NSAIDs
- **Second line** : Corticosteroid, DMARDs (methotrexate, sulfasalazine), TNF α inhibitor, biphosphonates

In our case : NSAIDs was **not tolerated** → Biphosphonates



Girschik et al, 2018

CNO : Chronic Non Bacterial osteomyelitis

- Rare auto-inflammatory disease affecting bones
- Children more than adults
- Can be very debilitating without treatment

This case

- Underscores the fluctuating nature of the disease and the value of molecular imaging for demonstrating the inflammatory activity

In the end

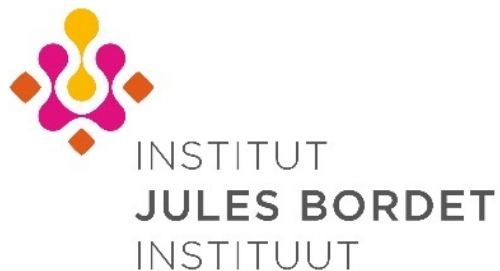
- We may have a role to play using bone scan and ^{18}F FDG-PET/CT
- To make this hard and challenging diagnosis faster



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THANK YOU



Hugues Azema