(CNO) in short term. Little is known about longterm outcome in CNO after IV-PAM.

Objectives: To describe a consecutive series of pediatric CNO (pCNO) patients who were prospectively followed after treatment with IV-PAM between 2003-2018 at a single center regarding: 1) The effect of IV-PAM on pain and Whole Body Magnetic Resonance Imaging (WBMRI) documented inflammation initially and after flare; 2) Relapse rate and; 3) Spinal CNO and 4) Urine N-telopeptide/creatinine ratio (UNtx/Cr) (a product of collagen 1 breakdown).

Methods:

Patients: All consecutive pCNO patients <18 years at diagnosis, with WBMRI confirmed active disease who required IV-PAM between 2003-2018. **IV-PAM:** First dose 0.5 mg/kg; subsequent doses 1 mg/kg (maximum annual dose 11.5 mg/kg). IV-PAM was administered once monthly x 9. With confirmed flare IV-PAM was repeated (max 9 doses/year). **Pain response:** Visual analogue scale for pain (VAS) with "0" indicating no pain and "10" maximum pain was administered at baseline, at each IV-PAM treatment, and at suspected flare. **Imaging:** WBMRI before 1st IV-PAM, at 6 and 12 months, at suspected flare and after retreatment. CNO resolution was documented as complete (CR) if there was 95% improvement in the WBMRI signal. Spinal x-rays were performed at baseline and annually. **UNtx/Cr** was measured at baseline, after each IV-PAM and with suspected flare.

Results: 17 patients (9F, 8M) were included. The median [range] age at CNO diagnosis was 10.3[4-15] years, and at first IV-PAM 11.6[4-20] years. The median [range] follow-up was 9.2[1-15] years. Six patients had unifocal CNO, 4 had spinal CNO (2 with baseline vertebral fractures) and 6 had multifocal non-spinal CNO. VAS was uniformly 10/10 at baseline and decreased to 0-1/10 first month after IV-PAM. CR was achieved by all at 6 months, which persisted at 12 months. Four patients, 3 unifocal and one multifocal, had no further flares. Twelve patients had WBMRI confirmed flare at previously active sites at 9-36 months and 11 received 1-9 further doses of IV-PAM. With flare, VAS ranged from 4-9/10 and decreased to 0-3/10 within first month after re-initiation of IV-PAM. On final WBMRI, 12/17 (70%) had CR and 5/17 (30%) stable mild increased signal but no clinical symptoms. No further spinal compression fractures occurred. One patient required a third course of IV-PAM. Regarding UNtx/Cr, each patient had appropriate reduction after first IV-PAM. No patients flared while UNtx/Cr remained suppressed. Three patients developed arthritis and one acne. At last follow-up, 10/17 (59%) patients were on no medications, 4/17 (24%) required prn Naproxen for CNO and 3/17 (18%) were on Naproxen and/or disease modifying medications for arthritis.

Conclusion: Long-term follow-up of pCNO patients treated with IV-PAM confirms that while several patients eventually flare, the flares are less painful and have an excellent response to IV-PAM retreatment. No further spinal fractures occurred. No flares occurred while UNtx/Cr remained suppressed, suggesting a role of osteoclasts in CNO. Further prospective multicenter studies are now required to define the long-term clinical and imaging response to IV-PAM.

Disclosure of Interests: None declared

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THU0546 RETROSPECTIVE STUDY ON ANKLE INVOLVEMENT IN CHILDREN WITH JUVENILE IDIOPATHIC ARTHRITIS (JIA) WITH A FOCUS ON HOW TREATMENT IS LINKED TO OUTCOMES

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Background: Ankle and midfoot joints have poorer long-term outcomes in JIA compared to other joints. Esbjörnsson et al found that children with ankle involvement in the first year of disease have higher CHAQ scores and are less likely to have achieved remission by eight years [1]. Adherence to treatment has been shown to improve short term outcomes in children with JIA but there is a lack of studies which look at the long-term outcome of children who experience barriers to treatment [2].

Objectives: The aim of this study was to identify risk factors for severe ankle involvement in children with JIA. This abstract focuses on barriers to treatment; one of the main identified risk factors for adverse ankle outcomes.

Methods: This was a retrospective study which looked at JIA patients who had been treated at the Children's Hospital in Glasgow and had tibiotalar, subtalar or midfoot involvement (TT, ST and MF respectively). 680 patients with JIA were identified from clinic lists from 2002 until 2018, inclusive. The clinic lists from 2008-2010 were inaccessible meaning that anyone who was seen exclusively within those two years has not been identified. Children with systemic arthritis were excluded. Of the 680 who were identified to have JIA; 123 have been found to have TT, ST or MF involvement. Of these; 104 were included in this study and the results of 19 patients were not available for analysis. For each of the 104 patients with ankle involvement their notes were reviewed and information about their JIA disease course was noted; specifically looking at any ankle involvement or potential risk factors for an adverse outcome. Poor ankle outcomes was defined as ankle arthritis having a significant impact on the child's quality of life, persistently limited joint movement for more than 12 months or damage on imaging.

Results: Barriers to treatment was defined as anything which prevented a child from getting their medication as prescribed.

Figure 1 shows the outcomes of children who had barriers to treatment compared to the children who did not. The difference between the two groups is stark with 67% of children who faced barriers to treatment having poor ankle outcomes compared to only 25% in the group which did not experience barriers to treatment. Furthermore, children with poor ankle outcomes are almost three times more likely to need surgery if they have experienced barriers to treatment compared to those who have not. This shows that children who experience barriers to treatment are more likely to have severe long-term damage to their ankle resulting in pain and limited functionality.

Figure 1: Outcomes in Children who experienced Barriers to Treatment

Abstract THU0546 – Figure 1

Five children declined any treatment at one point in their care against the advice of their healthcare team and of these five; four (80%) had severe ankle involvement with one requiring surgery and another's ankle auto-fused.

Conclusion: Experiencing barriers to treatment has been shown to increase the risk of poor ankle outcomes. Identified barriers to treatment ranged from concerns about adherence to needle phobias in addition to side effects and medication being with-held or taken incorrectly. This study demonstrates the need for further work to explore how these barriers can be effectively minimised.

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