

Cat Scratch Disease in a 1.5-year-old girl – Case report

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Abstract

Introduction. The paper is a case report presenting Cat Scratch Disease (CSD) in a 1.5-year-old girl. Bartonellosis, including CSD, are a group of infectious diseases which are rarely detected, therefore there are no statistical data concerning the aetiology, and the incidence of CSD noted in Poland is low in comparison with other European countries.

Objective. The purpose of the paper is to discuss several problems related to CSD.

Materials and method. A 1.5-year-old girl who was seen in hospital for the sparing use of her left arm when crawling. X-rays showed osteolytic lesions which radiologists described as multi-ocular cyst or infection. As neither clinical examination nor laboratory investigations found pathological signs, the patient was followed-up on an ambulant basis. Repeated x-ray taken 4 weeks later showed increased periosteal proliferation accompanied by pain. The baby was admitted to the Clinic but additional investigations found no pathologies. The baby was consulted by a rheumatologist and haematologist; however, they did not facilitate a definitive diagnosis. As the baby developed, because of a thickening of the soft tissues on the dorsal side of the distal epiphysis in the forearm the doctors decided to inspect the condition operatively. Macroscopic examination found brownish granulated tissue. Suction drainage was inserted and a tissue sample was tested for aerobic and anaerobic bacteria, tuberculosis and borelliosis. The test results were negative. The baby was in good condition, was not pyrexial and suffered from less pain. The diagnostics was further expanded and the baby tested for yersinia, chlamydia, tuberculosis and bartonella, i.e. CSD. The postoperative wound healed soon and radiological bony lesions began to resolve. After a month, we received a positive bartonella test result, the baby tested positively for *Bartonella henselae* IgG class, which confirmed past or active infection of CSD. A repeated test for *B. henselae* taken 6 months later showed a lower level of antibodies.

Conclusions. It should be remembered that CSD, which is an extremely rare infection, can be diagnosed despite mediocre clinical and radiological manifestations. Thus, in the case of infections of unexplained aetiology and mediocre manifestations diagnostics should include testing for *Bartonella henselae*.

Key words

Cat Scratch Diseases, osteolysis, osteomyelitis

OBJECTIVE

The purpose of the paper is to present an atypical case of Cat Scratch Disease (CSD) located at the distal epiphysis of the radius, accompanied by minor local lesions without generalized symptoms. We discuss the problem to establish a protocol for management when certain radiological and clinical changes have developed.

INTRODUCTION

The paper presents a case of a 1.5-year-old girl who was brought to the hospital for sparing her left arm. Her parents reported that for 2 weeks the baby was distinctly avoided loading her left hand when crawling. X-rays taken at that time showed an osteolytic lesion, which radiologists described

as multi-ocular cyst or infection (Fig. 1a, 1b). The baby was admitted to the Clinic. The blood tests taken found WBC: $14.59 \times 10^3/\text{ul}$ 6.00–11.00, ESR after 1h 5 mm/1h (normal range <20 [N]), C-reactive protein – CRP 0.01 mg/dl (<0.50 N). Scintigraphy of the skeletal system found increased bony metabolism in the area of the distal epiphysis of the left radius. A review of epidemiological history revealed no contact with domestic animals. The lymph nodes were not enlarged. Considering the clinical and lab results the baby was followed-up on an ambulant basis.

Four weeks later, the parents brought their baby to the Clinic because of ongoing pain in the left forearm. Repeated x-ray showed increased periosteal layer formation (Fig. 2a, 2b). Additional investigations ordered at that time also found no pathological changes. WBC $12.63 \times 10^3/\text{ul}$ 6.00–11.00; CRP 0.03 mg/dl (<0.50 N). Sonography examination of the abdomen and chest x-rays was negative. Sonographic view of the left forearm found an oval cyst $17 \times 9 \times 19$ mm filled with fluid of on the dorsal side of the distal epiphysis of the radius. The lumen of the lesion was echogenic, in the deeper sections especially. Power Doppler scan found features of increased vascularisation around the lesion and oedematous

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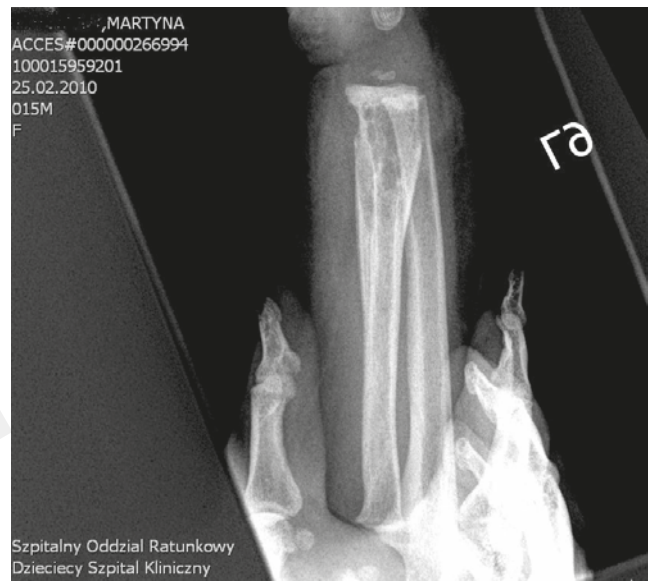


Figure 1a, 1b. X-ray of forearm during first symptoms (28.05.2011)

adjacent soft tissues. There was loss of cortical bone of the humerus, 3mm in diameter, corresponding to the fistula canal. The lesion was connected with the destructive area located within the distal epiphysis of the radius. There were features of oedema affecting the periosteal soft tissues on the palmar side and the pronator quadratus muscle, with distinctly enhanced vascular flow. Sono-scan corresponded to advanced osteomyelitis spreading to the soft tissues, and an abscess formed on the dorsal side of the distal part of the forearm. The opinions of other specialists, a rheumatologist and a haematologist, were of little help with respect to diagnosis.

Considering the formation of thickening affecting the soft tissues on the dorsal side of the distal epiphysis of the forearm it was decided to undertake surgical inspection which found macroscopic beige-brownish granulated tissue; suction drainage was inserted (Fig. 3a, 3b, 3c, 3d). A sample was histopathologically tested for aerobic and anaerobic bacteria, tuberculosis and borreliosis in The Chair and Department of Clinical Pathomorphology, at the Medical University, in Lublin. The test results were negative. The baby was in good general condition, afebrile and in less pain.



Figure 2a, 2b. Control X-ray of forearm (11.07.2011)





Figure 3a, 3b. Clinical view during operation day, presence of tumor (7.12.2011)



Figure 3c. Intraoperation view, morphological picture corresponding to granulomatous inflammation with abscesses (7.12.2011)



Figure 3d. The wound is sutured. Suction drainage is inserted (7.12.2011)

The results of histopathological tests came 2 weeks later and revealed foci of necrosis partly infiltrated by neutrophils surrounded by macrophages of endothelial cell morphology, single giant cells and unspecific granular tissue located peripherally. The morphological picture corresponded to granulomatous inflammation with abscesses, most likely of bacterial aetiology (yersinia, chlamydia or cat scratch disease). Immunofluorescence test found no *Mycobacterium tuberculosis*.

Diagnostics was expanded to include tests for *Yersinia enterocolitica*, *Chlamydia pneumoniae*, *Listeria monocytogenes*, *Mycobacterium tuberculosis* and *Bartonella henselae* – CSD. After a month it tested positively for bartonella – antibodies against *Bartonella henselae* IgG class 1:320 were found which confirmed past or active infection;

antibodies against *Bartonella henselae* IgM class were not obtained. The results were interpreted with account for the full clinical picture of the patient. The postoperative wound soon healed. Radiological bony lesions began to resolve. Surgical intervention brought improvement of the baby's condition; the markers of inflammation returned to normal.

After 6 months the test for *Bartonella henselae* was repeated: antibodies IgM class were not found, the level of antibodies IgG was 64, which was between positive and negative limit. The last radiographs showed completely healed bone, with only Harris lines presented due to growth arrest at time of infection (Fig. 4).

It was not until diagnosis was made that the baby's parents confirmed the possibility of the baby having had contact with a cat at her grandparents' house. The girl was playing with a cat and must have been scratched by the animal.



Figure 4. AP and lateral view X-rays show no differences between treated [L] and not infected [R] sides (20.06.2013)

DISCUSSION

CSD is a disease of mild bacterial aetiology manifested by regionally enlarged lymph nodes. *Bartonella henselae* is the most common pathogen, other *Bartonella* species are rare. There are 25 *Bartonella* species that are well known, half of which are human pathogens [4,15]. CSD is always related to cat's scratch and is most common among children and teenagers. Several days following contact with a cat an individual develops a primary lesion on arms, legs or face. It resembles papule or follicle and usually heals by itself without a scar, therefore it often goes unnoticed. About 2 weeks later, adjacent lymph nodes enlarge [7]. In the majority of cases, it takes the form of lymphadenopathy, 50% patients can develop proliferative manifestations. In single cases, symptoms from the musculo-skeletal system develop, e.g. bony inflammations, arthropathy, myalgia. In 10% cases, the course is atypical and can take the form of Perinaud's oculoglandular syndrome, hepatitis, inflammation of the spleen, bones or septicaemia. [2]. Musculoskeletal complications are rare. The frequency of osteomyelitis ranges from 0.17% – 0.27% [1, 2, 6, 7, 8, 9].

In western countries, CSD cases are reported and detected considerably more frequently in comparison to Poland. The presenting symptoms include fever, bone infection, and the site of the primary lesion is unrelated to the location of osteomyelitis. The vertebral column and pelvic girdle are the preferred sites of infection. Concomitant lymphadenopathy was a frequent finding, but osteomyelitis also developed after an asymptomatic interval. Only 2 cases involved the hand. The average is 2–62 years, mostly affecting children around 2–5 years old [5, 11, 12].

Managing bartonellosis, including CSD, remains an open issue. In some diseases caused by pathogenic *Bartonella spp.* recovery is spontaneous, in others surgical intervention is required, otherwise it can result in fatal consequences [3, 13, 14, 16, 17].

Single reports on effective antibiotic therapy in CSD suggest the use of ciprofloxacin, rifampicin and cotrimoxazol [5, 8, 10, 14].

CSD is diagnosed on the basis of medical history review, typical clinical picture and serological and/or histopathological studies. In the case presented above, the diagnosis was made postoperatively after the sample had been analysed

and serological results obtained. Histopathological tests and review of the medical history confirmed the diagnosis. According to Sander et al., immunofluorescent assessment (IFA) is the best diagnostic method [4, 14]. Other tests include ELISA or Western Blot analysis. The majority of CSD cases do not require antibiotic treatment, and surgical removal of enlarged lymph nodes or purulent lesions is very rare [8].

CONCLUSIONS

Cat Scratch Disease should always be considered in the case of diagnostically difficult osteolytic lesions, especially those with concomitant enlargement of adjacent lymph nodes. The presented case underlines the importance of the course of the disease, review of epidemiological history and symptoms. It should be remembered that CSD, although it is extremely rare, should be considered despite mediocre clinical and radiological symptoms. In diagnosing the cases of unexplained aetiology, doctors should remember to include specific serology tests for *Bartonella henselae*.

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