Case report

Positional Hand Pallor and Isolated Epitrochlear Lymphadenitis Secondary to *Bartonella henselae* Infection.

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Abstract

Cat scratch disease is a communicable disease caused by the *Bartonella henselae* bacteria. Regional lymphadenopathy is the hallmark of cat scratch disease and about 75% of lymphadenopathy cases are localized in the head and neck region. An epitrochlear lymphadenopathy is a rare condition at any age and often misdiagnosed as it is not normally palpable. External compression of an enlarged epitrochlear lymph node compromising vascularity was not mentioned in any literature before. We present a case of a 13-year-old girl with right positional ipsilateral hand pallor and epitrochlear lymphadenitis with serological evidence of *Bartonella henselae* infection.

Keywords: Hand pallor; Epitrochlear lymphadenopathy; *Bartonella henselae*

Bangladesh Journal of Medical Science Vol. 20 No. 04 October'21. Page : 914-917
DOI: https://doi.org/10.3329/bjms.v20i4.54152

Introduction

Cat scratch disease (CSD) is caused by *Bartonella henselae (B. henselae)* which is a fastidious slow growing pleomorphic gram-negative bacillus[1].Cats are the major reservoir for this bacteria[2]. CSD can be transmitted from an infected cat directly via a scratch or bite or indirectly via contact with cat's saliva through broken skin or mucosal surfaces[2]. In the United States, the annual incidence of CSD is 4.5 per 100,000 individuals[3]. To date, there is no epidemiology data available for CSD in Malaysia.

CSD predominantly affects children less than 14 years old. 32.5% cases were reported within this age group[3].CSD presents with a wide range of clinical manifestations. Typical CSD is characterized by regional lymphadenopathy. After a history of exposure to cats, more than 90% of patients were reported to have developed a non-tender papule or vesicle at the primary inoculation site[4]. Regional lymphadenopathy would then develop around two weeks' post-inoculation. About 75% of lymphadenopathy cases are localized and 50% are detected in the head and neck region[5]. Up to 14% of patients had atypical presentations involving the liver, spleen, eye, and central nervous system.

To our knowledge, there have been no published reports on external compression of an enlarged epitrochlear lymph node related to *B. henselae* infection that compromised the vascularity of the upper limb. Pallor and pulselessness of a limb are signs of ischaemia. Most causes of limb ischaemia are due to either embolic phenomenon or thrombosis which results in an arterial occlusion[6]. In rare circumstances, arterial compression by abnormal muscles, muscle hypertrophy or bony spurs could also cause ischaemia[6].

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Case presentation

A 13-year-old girl presented with a one-week history of painful right mid arm swelling near the right elbow joint without overlying skin changes. The swelling rapidly increased in size and restricted mobility of her right arm. She did not have fever, rash or any similar swellings elsewhere in the body. The patient also did not have appetite or weight loss. She had a history of being scratched by a stray kitten around three weeks prior to the development of the mid arm swelling. She developed mild fever for two days at that time, but did not seek any medical attention.

On examination, she was afebrile, not pale and not cachexic. There was a mass at the dorsal aspect of the right elbow, 2 cm superior to the medial epicondyle. It was tender, firm, warm, fluctuant and measured 8 cm x 6 cm in diameter. It was fixed to the skin but not to the underlying musculature. There was no erythema and the mass is not pulsatile. There was restricted flexion of the elbow due to pain. The hands were warm with good capillary return. There was hand pallor with feeble radial pulse noted upon flexion of the elbow of the affected arm (Figure 1). The SPO_2 readings for both hands were normal. There were no other lymph nodes palpable elsewhere and no hepatosplenomegaly.

Her full blood count showed a normal white cell count. C-reactive protein was 5.1 mg/L (Normal = < 5.0 mg/L). Plain radiograph of the humerus and elbow joint showed no significant finding. The possibility of right mid-arm abscess needed to be ruled out thus an urgent ultrasound of the right arm was performed. The ultrasound demonstrated four hypoechoic lesions suggestive of enlarged lymph nodes with preserved fatty hilum at the medial aspect of the right arm, just above the elbow. The largest node measured 1.4 x 1.6 x 1.9 cm (AP x W x CC), located adjacent to the basilic vein and caused external compression to this vein (Figure 2). The entire length of the basilic vein was fully compressible and there was no echogenic material within it to suggest venous thrombosis. There was no focal collection seen at the affected site. The ultrasound scan in this case was limited only to the medial aspect of the arm as the patient was unable to fully flex the elbow due to pain. Mantoux test and chest X-ray were done and found to be negative. *Bartonella* and *Toxoplasma* serology were also taken.

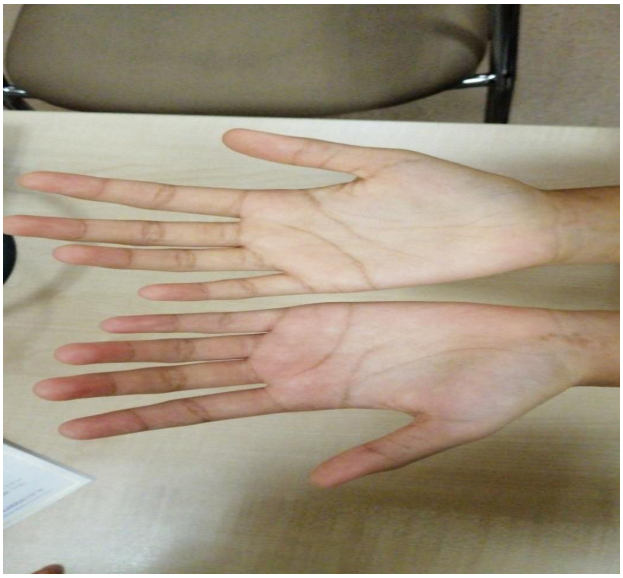

Figure 1: Pallor of the right palm on flexion of the right elbow

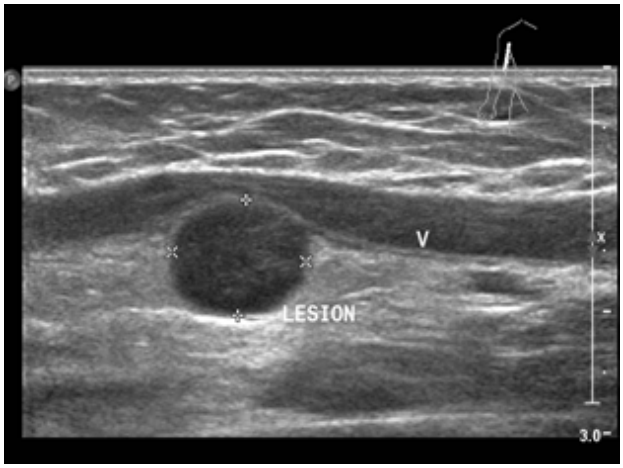

Figure 2: The largest hypoechoic lesion is adjacent to the basilic vein causing compression on the vein

Based on the clinical and radiographic findings, a diagnosis of reactive epitrochlear lymphadenitis was made and oral amoxicillin and clavulanate (Augmentin®) 625 mg BD for five days was prescribed. However, the mid arm swelling did not reduce in size despite Augmentin®, thus diagnosis of reactive epitrochlear lymphadenitis secondary to possible CSD was made. Oral azithromycin 500 mg stat followed by 250 mg for five days was then prescribed. However, the swelling persisted and the option of an excision biopsy due to non-resolving mid arm swelling was discussed.

Serology tests for *B. henselae* were later found to be positive for both IgM and IgG with titres of >1:24 and >1:128 respectively and the diagnosis of right epitrochlear lymphadenitis secondary to CSD was

confirmed. Upon follow up at five weeks of illness, the swelling resolved thus an excision biopsy was not required.

Discussion

An isolated epitrochlear lymphadenopathy is rare and usually manifests as part of a generalized lymphadenopathy[5]. In a study of 1200 cases of cat scratch disease, 610 cases involved the upper limb lymph nodes, of which the location for 586 cases was axillary and only 24 cases were at the epitrochlear region[4]. In children, epitrochlear lymphadenopathy without an obvious cause presents a diagnostic dilemma and an enlargement of more than 0.5 cm is always pathological in nature[5]. It could be the first sign or the only site of involvement in a haematological malignancy, thus detailed history taking, and clinical examination is necessary to determine the underlying cause[5].

Our patient was also found to have a unique presentation of transient hand pallor upon flexion of the affected elbow with a weak palpable radial pulse. Theoretically, this symptom can be explained by an arterial occlusion as a result of thromboembolic phenomenon[6]. Brachial artery compression syndrome is a rare cause of hand ischaemia[6] in which the artery is compressed externally by surrounding structures such as abnormal muscles, muscle hypertrophy or bony spurs[6]. Anatomically, the epitrochlear lymph nodes lie alongside the basilic vein with other contents of cubital fossa including the brachial artery, brachial vein, median and radial nerve. Because of its anatomical location, an enlarged epitrochlear lymph nodes could cause compression of the brachial artery that could explain the unique presentation of our patient. In this case, the ultrasound scan was limited only to the medial aspect of the arm as the patient's arm was in a semi-flexed position due to pain. Visualization of the brachial artery was not possible at this time, but the scan revealed the right basilic vein compression by one of the enlarged nodes. Even without direct imaging evidence of arterial compression, we strongly feel that this is a possible explanation of the patient's unique presentation of unilateral hand pallor, mimicking a thromboembolic phenomenon. To the best of our knowledge, this finding has never been reported before in literature.

The diagnosis of CSD was previously made clinically[4]. Currently, the best approach of diagnosing CSD is by serological test as the *Bartonella* skin test is no longer used and a positive culture is difficult to be obtained because of its slow growing nature[7]. There are two serological tests available, Indirect fluorescence assay (IFA) testing and Enzyme-linked immunoassay (ELISA). For our patient, IFA testing for *B. henselae* was sent after three weeks of symptoms onset and were positive for both IgM and IgG with titres of >1:24 and >1:128 respectively. The presence of immunoglobulin M is usually difficult to obtain because of its subtleness[7]. A positive IgM test therefore strongly suggests a recent infection[7]. The sensitivity of the combined IgM and IgG test is 98% for detection of *B. henselae*[7].

The classical CSD is self-limiting, but current recommendations suggest treatment even in immunocompetent patients as recent data shows that *B. henselae* infection can disseminate in up to 14% of patients [4]. The recommended dosage for children less than 18 years old is tablet azithromycin 10 mg/kg PO on day 1, followed by 5 mg/kg PO for four days; for children less than 45.5kg, the dose is 500 mg PO on day 1, followed by 250 mg for four days for children more than 45.5 kg[8]. Ciprofloxacin is an alternative to macrolide[9]. A repeat treatment is also suggested in a patient with lymphadenitis that does not respond to therapy after three to four weeks, which is similar to our case.

Lymph node biopsy is indicated in those with persistent lymphadenopathy despite treatment or those with high risk of malignancy or immunocompromised[2,4]. In our case, an excision biopsy was planned but was not carried out due to resolution of symptoms at week 5 of illness, after a total 15 days of antibiotics treatment.

Conclusion

In conclusion, it is important to consider epitrochlear lymphadenitis secondary to CSD as a differential diagnosis of mid arm swelling, especially in the paediatric age group. A thorough history taking and complete physical examination is important to recognize this condition, ensuring correct treatment initiation, as well as eliminating the need for any invasive procedures. Hand pallor is an interesting atypical sign found in this patient with epitrochlear lymphadenitis.

Funding

None

Conflict of interest

The authors declare that they have no competing interest.

Ethical clearance

A verbal consent was taken from the patient and a written consent was taken from the patient's parent.

Authors's contribution

NA Jamil and **IA Ismail** were the primary care doctors that attended to the case and **ND Mohamad Ali** was the radiologist on call who did the ultrasound. **NA Jamil** drafted the manuscript. **ND Mohamad Ali** provided the radiology input for the manuscript. **IA Ismail** and **SF Badlishah-Sham** revised it critically for important intellectual content. All authors have read and given approval of the final manuscript. Each author has participated sufficiently in the work to take public responsibility for appropriate portions of the content as described above.

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