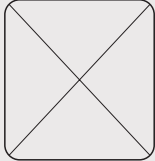
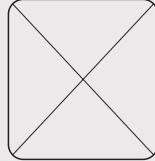


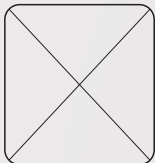
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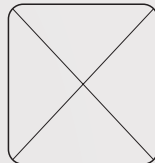
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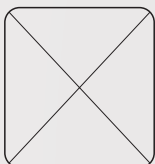
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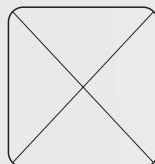
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Scapular Tuberculosis in the Paediatric Population: Suspicion and Early Treatment is the Key

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Abstract

Background: Scapular tuberculosis (TB) in paediatric population is very rare and required clinician suspicion for early diagnosis and treatment.

Methods and Materials: We conducted a retrospective study involving 8 children with a diagnosis of TB of the scapula that was confirmed by histopathological examination. The patients were clinically assessed for signs and symptoms. Pediatric/adolescent shoulder survey (PASS) and numerical rating scale (NRS) scores were used to assess the improvement with treatment.

Results: The mean age of the patients was 10.5 years with a mean duration of symptoms around 4 months. All the patients had raised ESR and CRP values with MRI evidence of inflammation or cold abscess. In all the patients, either FNAC or biopsy was performed. Gene Xpert and histopathological examination confirmed the diagnosis of tuberculosis. All the patients were given (Anti-TB therapy) ATT according to their age/weight for a period of 12 months, as per the RNTCP guidelines. There was significant improvement in ESR, CRP, NRS, and PASS scores with MRI-evidence of disease resolution.

Conclusion: Scapular tuberculosis should be suspected in children presenting with vague shoulder or scapular pain. The patients should be investigated thoroughly and treated with anti-tubercular therapy.

Keywords: Flat bone tuberculosis, Scapular tuberculosis, Paediatric tuberculosis, Ant-tubercular therapy, Biopsy

Introduction

Tuberculosis is one of the leading causes of morbidity and mortality in endemic regions. TB can affect various tissues of the body including the osteoarticular region. TB in children is common in endemic areas but osteoarticular TB constitutes only 2% of all cases [1]. The spine is the most commonly site affected. The occurrence of TB in flat bones such as the scapula without any other foci is very rare [2]. Suspicion and early treatment help in the proper management of the condition We describe a case series of eight children with scapular TB.

Methods and Materials

Data Collection

A retrospective case note review was undertaken on children presenting to four different centres over a 3-year period. Descriptive data including the age of the patient, side involved and duration of symptoms, and relevant history records were retrieved.

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Table 1: Descriptive data of the patients in the study

S.no	Age (Years)/Sex	Side	Duration of symptoms (months)	ESR (mm/hr)		CRP (mg/dL)		Abscess	Biopsy/FNAC	PASS		NRS	
				Pre-treatment	Post-treatment	Post-treatment	Pre-treatment			Post-treatment	Post-treatment	Pre-treatment	Post-treatment
1	6/Male	Right	2	34	12	17.2	1.4	Present	Biopsy	39	89	8	2
2	14/Male	Right	6	58	7	30.6	2.1	Present	FNAC	36	90	7	2
3	13/Female	Left	4	55	6	28.5	2.3	Present	Biopsy	42	94	7	1
4	8/Male	Right	2	43	12	20.2	2.2	Present	Biopsy	48	92	6	1
5	10/Male	Left	5	37	5	18	1.3	Absent	Biopsy	42	85	7	3
6	11/Male	Right	6	62	6	22.6	3.5	Present	Biopsy	29	90	9	3
7	8/Female	Right	4	52	9	23.4	3.2	Present	FNAC	31	93	8	2
8	14/Male	Right	3	51	7	29.8	2.8	Absent	Biopsy	38	99	6	1

Inclusion criteria

1. Patients less than 18 years of age.
2. Confirmed diagnosis of scapular tuberculosis through histopathological examination of the tissue from fine needle aspiration cytology or open biopsy.

Exclusion criteria

1. Unconfirmed diagnosis of scapular tuberculosis or given empirical antitubercular treatment.
2. Multi-focal TB or past history of TB

Assessment

All the investigation data were assessed in the study. Radiographs of the scapula/shoulder, contrast MRI and laboratory parameters including complete blood count, erythrocyte sedimentation rate (ESR), C reactive protein (CRP), liver function test (LFT), HIV antibody test, Hepatitis B, Hepatitis C antibody test and peripheral blood smear were recorded. Tissue was analyzed by histopathological examination, Gene Xpert test, AFB and Gram staining, and bacterial and fungal cultures.

Clinical assessment

The patients were clinically assessed for signs and symptoms. Pediatric/adolescent shoulder survey (PASS) and numerical rating scale (NRS) scores were used to assess progress. The parents/guardians helped in this scoring, in case the children were unable to understand the scoring questionnaires.

Treatment

All patients were given anti-tubercular drugs (ATT) according to their weight or age as per the Revised National Tuberculosis Control Program (RNTCP). ATT was given for a total of 12 months (2 months intensive phase and 10 months continuation phase) and a repeat contrast MRI was performed before cessation of treatment. During follow-up visits, ESR, CRP, and LFT were performed.

Statistical Analysis

The descriptive data were tabulated and assessed accordingly. The mean, standard deviation, and range were calculated for the data using SPSS version 23. Student t-test was used to

analyze the comparative data. The p-value was calculated to assess the level of significance of the data.

Results

Nine patients met the inclusion criteria and 1 patient was lost in follow-up, leaving 8 children available for the final analysis (Table 1). The mean age of the patients was 10.5 years with 6 male and 2 female patients. Six patients had right-side scapula involvement and 2 had left-side involvement. All the patients complained about scapular pain and terminal painful shoulder motion of the affected side. There was slight swelling and tenderness in the scapular region. There was no sinus. None of the patients had any constitutional symptoms or TB affecting any other part of the body.

The mean duration of symptoms to diagnosis was 4 + 0.5 months (range: 2 – 6 months). The mean ESR at presentation was 49 + 8.5 and CRP was 23.7 + 6.3. The rest of the laboratory parameters were normal in all the patients except two children who had anaemia. Radiographs of the scapula showed bony changes in 2 patients only (Fig. 1). The contrast MRI T2 weighted images showed hyperintense changes in the scapula region involving the body of the scapula and collection in the infraspinatus muscle with some osteolysis of the scapula body (Fig 2 and 3) or in 2 patients supraspinatus muscle was also involved. There was a cold abscess in 6 patients with collection mostly under the infraspinatus muscle.

Ultrasound-guided FNAC was attempted in all cases and was successful in 3 patients. In the rest of the patients, an open biopsy was performed. The tissue was analysed by histopathological examination and Gene Xpert test. Bacterial culture, Gram stain, and fungal culture were negative in all the patients. AFB staining was positive for mycobacterium tuberculae in two patients. Histopathological examination depicted granuloma with epithelioid macrophages, lymphocytes, and some necrosis and fibrosis (Fig. 4). Gene Xpert was positive in all the patients, sensitive to Rifampicin.

There were no drug-related complications. All patients responded to the treatment and showed a significant decline in the values of ESR and CRP during follow-up. The mean ESR and CRP at 12 months follow-up were 8 and 2.3 respectively with significant improvement ($p < 0.05$). The contrast MRI at the end of 12 months depicted complete resolution of the

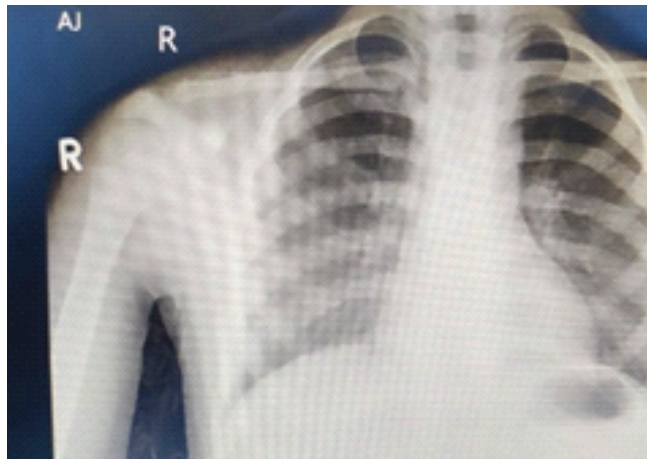


Figure 1: Radiograph of right scapula



Figure 2: Coronal images of contrast MRI depicting hyperintensity in the infra-scapular region.

abscess.

The patients were assessed by PASS and NRS scoring systems. At presentation, the mean PASS score was 38.1 ± 0.7 which significantly improved at the last follow-up at 12 months with a mean value of 91.5 ± 7 . Similarly, the NRS score improved from 7.25 ± 1.4 to 1.8 ± 0.7 at the 12-month follow-up. The improvement in scores was significant ($p < 0.05$).

Discussion

Extrapulmonary TB accounts for 10-35% of all cases. TB of isolated flat bones is a rare diagnosis [3]. Scapula is a triangular flat bone rarely affected by TB. The detection of scapula TB is delayed, essentially due to late diagnosis [4]. Few case reports of scapular TB in paediatric patients are described in the English literature [2, 3, 5-9]. This is the first case series of 8 cases of isolated scapular TB in children. There are 5 published literatures on isolated scapular TB in children (<18 years) (Table 2) [2, 3, 7-9].

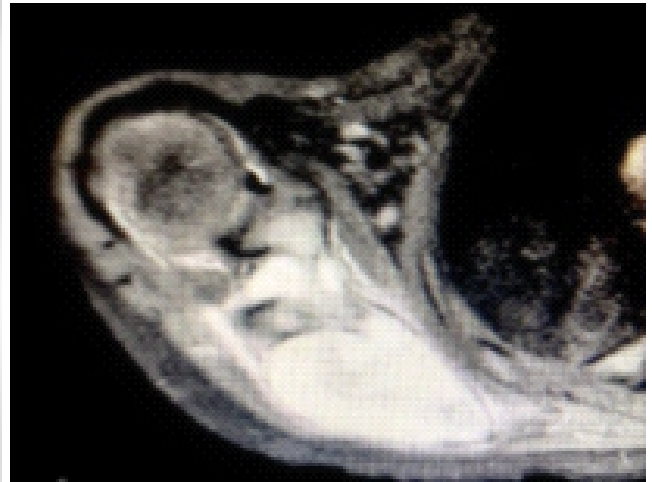


Figure 3: Axial images of contrast MRI depicting hyperintensity in infraspinatus muscle.

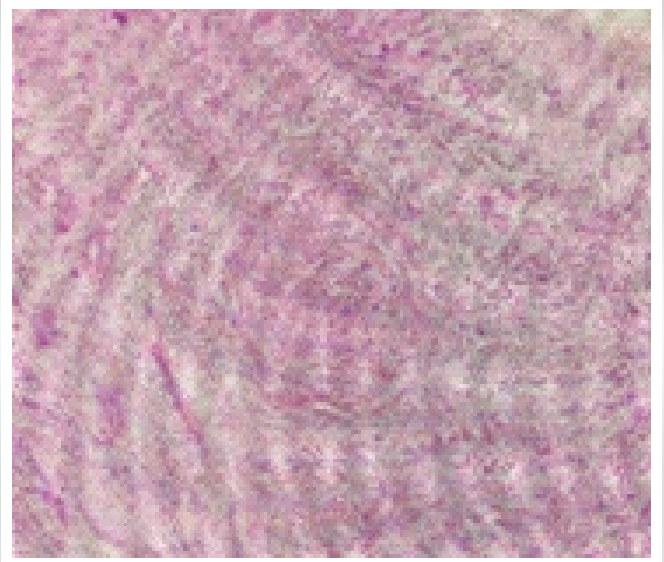


Figure 4: Histopathological examination of the abscess aspirated

TB reaches the bones through contiguous or hematogenous spread. In isolated scapular TB, the primary source of infection may not be detected. Immunocompetent status should be ascertained in scapular TB in paediatric population. Clinically the patient can present with varied duration of the scapular blade or shoulder pain with restriction of overhead abduction. Mild to moderate swelling and tenderness can be seen over the scapular region. Children are generally in good health without significant constitutional symptoms. Haematological parameters were normal in such children with raised ESR and CRP levels. Clinically in our series, no patient had any sinus around the shoulder or scapular blade of the affected side. Few of the case reports described scapular TB as a part of multifocal TB [5]. Osteolytic lesions or cold abscess formation is the most common pathological presentation of scapular TB in children. In early cases osteolytic lesions may not be visible on radiographs. Other changes seen on radiographs include osteopenia, subchondral erosions, varying sclerosis, or cystic

Table 2: Review of literature

S no.	Study	No. of cases	Age(years)/gender	Site of tuberculosis	Presenting complaints	Radiological investigation	Treatment
1	Jain et al [9]	1	14/Male	Body of Scapula involving glenoid margin	Pain and discharging sinus right upper scapula	Radiographs and MRI	ATT for 18 months
2	Hosalkar et al [2]	1	8/Female	Scapula	Pain and swelling from 3 months	Radiographs, MRI,	ATT for 12 to 15 months
3	Chandane et al [3]	1	7/Male	Scapula	Pain and swelling in the right shoulder	Radiographs, Mantoux test, MRI	Incision and drainage with ATT for 12 months
4	Husen et al [7]	1	18/Male	Spine of scapula near glenoid cavity	Pain and swelling in the left shoulder	Radiographs, CT scan	ATT for 9 months
5	Balaji et al [8]	2	17/Male 17/Female	Body and spinous process Inferior angle	Pain and swelling left suprascapular region Swelling over her right upper back	Radiographs, CT scans, MRI	ATT for 9 months Incision and drainage of the abscess along with debridement, sequestrectomy, and curettage of the right scapula. ATT for 9 months

changes. Only 2 patients in our series had radiological changes affecting the scapular body. The body of the scapula remains the most common site of affection in scapular TB. Contrast MRI helped in the early detection of scapular TB. Contrast MRI even in the early stages of the disease can delineate the cold abscess, inflammation, and bone erosions. They also guide the site best suited for biopsy or FNAC. Scapular TB in children being a rare disorder requires a histopathological diagnosis. There are a few differential diagnoses of scapular TB in children including eosinophilic granuloma, sarcoidosis, cystic neoplasm, chordoma, fungal infection, and other pyogenic infections [10]. In all the patients, tissue was obtained for diagnosis either by biopsy or FNAC. The histopathological changes and GeneXpert test helped in the diagnosis of TB in each case.

All children were treated with ATT as per the national guidelines. All responded well to medical management and none required surgical intervention. This is similar to the published literature, treating isolated scapular TB by ATT [four drugs (isoniazid, rifampicin, pyrazinamide, and ethambutol) for 2 months followed by two drugs regimen (isoniazid and rifampicin) for 10months]. At the final follow-up of 1 year, all the children had no clinical symptoms and signs of TB with normal ESR values and MRI.

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Conclusion

Scapular tuberculosis is a rare entity, especially in the pediatric age group. The clinician must be aware of the possibility of scapular TB in case of persistent scapular pain. There should be a low threshold to suspect TB in scapular pain. Contrast MRI should be performed early in the course of the disease since radiographs frequently show no changes. Early diagnosis and proper management help in alleviating the pain and restriction of shoulder movement. Histopathological examination is required for diagnosis as the scapula is a rare site for TB in pediatric patients. ATT is sufficient for the treatment of scapular TB.

Clinical Relevance

Scapular tuberculosis in paediatric population is a rare entity. Clinician suspicion is required to further investigate the patients. MRI should be performed early in such patients and histopathological diagnosis should be ascertained in such patients. Early initiation of anti-tubercular therapy leads to a better outcome.

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Declaration of patient consent : The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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