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A case report of cervicofacial actinomycosis

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ABSTRACT

Actinomycosis is a rare chronic granulomatous disease. Here we reported a 23-year-old male presented with a neck mass in the antero-inferior part for 35 days, with increasing in size and appearing some new masses in various sizes on left side of neck. The masses improved to some extent, with an empirical therapy but relapsed after discontinuation of treatment. After further investigations in second admission, long-term treatment with penicillin was started with a diagnosis of cervicofacial actinomycosis. Bronchial cleft actinomycosis was proved in this patient, which is one of rare cases of actinomyces involvement.

1. Introduction

Actinomycosis is characterized by purulent granulomatosis disease[1,2] accompany by sinus tract, passing from anatomic barrier, relapse after antibiotic therapy[3] chronicity and mass like lesions[4]. Actinomycete is a gram positive bacilli, negative AFB, anaerobic or microphilic that could be like branching[1,2] or beading with staining[5]. Actinomycetes could be normal flora in oral cavity, gastrointestinal tract and female genitourinary tract[2,6-8]. Actinomycosis could be seen more with poor dental hygiene, dental and oral surgery and manipulation[8], oral mucus damage due to radiation, previous abdominal surgery and pulmonary infection due to aspiration[1,5,9]. Interestingly HIV is not a risk factor for Actinomycosis[10].

In up to 15%-20% of the cases it involves throat and in 10%-20% cases it involves the pelvic area[5]. Cervicofacial involvement could be seen with soft tissues swelling, abscess, mass or ulcerative lesion that can be mistake with malignancy[1].

Actinomycosis infection can be seen at any age but it is most prevalent at middle age[1]. In a study during 1972-1999, 1997 cervicofacial actinomycotic patients evaluated with positive culture, the most prevalence age range in female was in 11-40 years old and in male in 21-50 years old group[11]. A study in Germany, reported that actinomycosis infection incidence is one case per 100000 people and in cleveland area is one case per 300000 people [1]. This disease is uncommon but it is not so rare[1]. Here we reported a case of actinomycosis with bronchial cleft involvement.

2. Case report

The patient was a 23-year-old male, hand worker, Isfahan resident since 10 years ago. His admission to our hospital came back to June 2009. The present illness started since 35 days ago before admission in infectious service with a cervical mass in antero-inferior part of the neck which near the mid cervical line. During next 9 days, he had not any sign like, weight loss, night sweats, *etc*. There was no history of oral manipulation or trauma with normal oral and dental hygiene, not any dental decay, not any contact with birds or insect's sting and not any travel out of Isfahan city and not any family history of tuberculosis.

The patient admitted in ENT ward for one week with tuberculosis lymphadenitis as primary diagnosis. Cervical mass excisional biopsy was done and specimen was sent to pathology but there was no evidence of AFB and tuberculosis lymphadenitis. During ENT service admission another red skin lesion was seen upper and in front of the SCM muscle (Figure 1A). Finally, the patient discharged without any diagnosis. The cervical masses were improved partially after one week of oral amoxicillin administration. Twelve days after discontinuation of antibiotic therapy, the lesion was reappeared as more inflammation, larger size and some satellite lesion along SCM muscle and angle of mandible (Figure 1B). He returned to hospital and admitted in infectious service. His scalp skin was normal and had

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not any sign of infection. In oral cavity there was not any lesion while throat and tympanic membrane were normal. Examinations of the chest, heart, abdomen and limbs were all normal. In cervical examination there was not any right lymph adenopathy but in front of the neck there was one surgical scar and there were 5–6 red, painful, hot and fluctuated masses that 3 of them were in front of the SCM muscle and 2 of them were below the angle of mandible angle. The largest mass size was 25 mm×30 mm but without fistula. There was not any lymphadenopathy. The lab data were as follows: CRP negative, BUN, Cr, Na, K, BS, PT, PTT, INR, ESR and U/A were all normal. WBC: 8 100 (58% PMN, 23% Lymph, 19% Mix), Hb: 15.6 mg/dL and PLT: 214 000. After 2 weeks, CBC was checked again that indicated WBC: 6 900 (69% PMN, 28% Lymph, 12% Mix) and Hb: 14.7 mg/dL and PLT: 213 000. Skin tuberculin test was reported negative after 72 hours. Soft tissue sonography of the left side of the neck indicated multiple nodules with septation and heterogenus echogen paranchymal that manifest calcification or granuloma with largest size of 12 mm×26 mm. It showed a region of mixed echo in the mass lesion which could be due to collection and contain a sub cutaneus part with 16 mm×39 mm that was related to deeper soft tissue and intramuscular having a 13 mm diameter sinuse tract that presented bronchial cleft infection. In cervical spiral CT scan parotid and submandibular glands were normal. Parapharyngial and ptrygopalatin cavity were normal. Necrotizing and calcified lymphadenopathy was seen in front of the left carotid sheet and through the left posterior cervircal lymph node circle (Figure 2). After ENT service consultation, FNA was done and there purulent yellow samples were prepared, one of them was sent to microbiology that has gram positive cocci in culture with negative smear. Second specimen was sent to fungal laboratory that had gram positive fine short fiber with negative fungal culture. The third specimen was sent to one governmental center for tuberculosis that had positive AFB. After one week of admission in infectious service, secondary cervical lesion excisional biopsy was done and the result of mass excisional biopsy reported a dense inflammatory cell infiltration with neutrophil predominate in background of granular tissue no evidence of neoplastic cell in this pathology, no positive evidence for TB by PAS and zeil nelson staining.

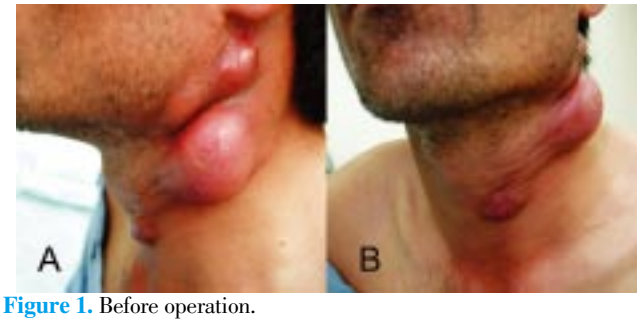

Figure 1. Before operation.

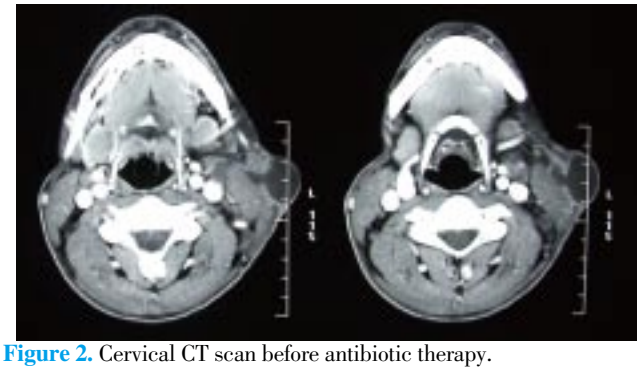
Figure 2. Cervical CT scan before antibiotic therapy.

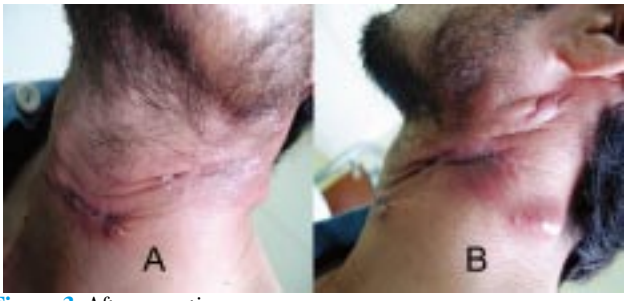

Figure 3. After operation.

By Considering his medical history and paraclinical data and also rapid response to short term antibiotic and early relapse, the cervicofacial actinomycosis and bronchial cleft infection was diagnosed for the patient. Finally, the patient was put under intravenous Penicillin therapy 4 million units, every 4 hours, for one month. The patient was discharged after relative recovery and controlling inflammation with oral Penicillin–V 500 mg, every 6 hours. His lesions became smaller, skin redness on the lesions and masses tenderness was disappeared. In spiral CT scan which had taken in order to assess the lesions, superficial and deep lesions were relatively recovered(Figure 3).

3. Discussion

A case of actinomycosis is reported with multiple cervical masses lymphadenopathy and abscesses, which occasionally is associated with diagnostic challenge. The cervical masses have multiple causes, in one study 84% of cervical mass were benign but 16% were malignant[3]. An infection condition is found in 37% of benign cases[3]. Among infections etiologies that can be presented with cervical masses are tuberculosis, actinomycosis, nocardiosis and some other rare causes like brucellosis, anthrax, toxoplasmosis and infectious mononeucleosis, all of them can also create a persistent lymphadenopathy for several months[12].

Lymphadenitis is the most frequent form of extrapulmonary tuberculosis. It is usually unilateral and cervical in location. It presents as painless, red, firm mass[1,3]. The tuberculin test result is almost always positive. Fine needle aspiration demonstrates cytologic evidence of granuloma, but smears or cultures are usually negative[1]. Localized lymphadenitis is the most common form of non tuberculous mycobacteria (NTM) disease in children, with a peak incidence between the age range of 1–5. NTM–affected lymph nodes are usually in the anterior cervical chain and are unilateral and painless. The tuberculin skin test is often weakly positive(5 to 10 mm), but it may be more than 10 mm[1].

Nocardia is another causes of cervical soft tissue involvement that can cause lung, CNS and systemic involvement, osteomylitis, arteritis, cellulitis. In immune deficient condition, transplantation, cytotoxic and immunosuppressive drugs consumption, nocardial involvement can be fistulated to skin that is more seen in thoracic area. Sulfur granules (bacterial macro colonies) similar to those seen in actinomycosis, may be found in nocardial mycetomas. Nocardia usually is stained by acid fast in tissue sections if a method such as that of Fite–Faraco is use, where as actionomyces spp do not[1].

Actionmycetes can involve cervical soft tissue and present with cervical masses[1]. Lymphatic spread and associated lymphadenopathyare uncommon[4]. But, cervicofacial actinomycosis is the most common involvement in human and the most common location for diagnosed actinomycosis

is the perimandibular region. The classic lesion located at the angle of the jaw is the most frequent location (submandibular)[1]. Male to female ratio is approximately 3:1 [1]. In actinomycotic lesions, the causative actinomycetes essentially are always associated with other microbes from the indigenous mucosal flora of the infected patient[2], but monomicrobial infections undoubtedly occur [1].

In one study, on 1997 cases of cervicofacial actinomycosis showed, only 4.5% (90 cases) specimens of fermentative actinomycetes were isolated alone in culture. All the other samples (95.5% or specimens or 1907 cases) contained various further aerobic and /or an aerobic bacteria, in addition to the pathogenic actinomycete species[11]. Sulfur granules were never seen in patient that may be related to previous antibiotic consumption. But, other cause may be due to low percentage of this finding (Sulfur granules). Cervicofacial actinomycosis may be associated with draining sinus tract in approximately 40% of cases and grains may be either micro- or macroscopic and be identified grossly from draining sinus tracts[1]. In addition, sulfur granules are characteristic for actinomycosis but grains are seen in mycetoma and butriomycosis[1,3].

Using staining, culture, pathological examination and PCR can be useful for diagnosis of actinomycosis[1,5]. Therefore, it should avoid any antimicrobial therapy prior to obtaining the specimen[1,3]. Mean while, specimens need to be from draining sinuses, deep needle aspiration, or biopsies[5]. The presence of surfur granules are of significant diagnostic relevance but always, are not identified easily[1,13].

Hematoxylin-eosin staining of tissue suffices to demonstrate the grain, but a special stain (*e.g*, Gram, gimsa, silver or 1% methylene-blue solution)[4] is needed to show that the grain is composed of branching bacterial[1]. A gram stain of the specimen is usually seen more sensitive than culture, especially if the patient has received prior antibiotics[1]. In this case, it was reported by requesting for mycology as gram-positive and short filamentous rods. If branching bacteria are seen on staining of the grain and the infection did not originate in subcutaneous tissue, then the diagnosis of actinomycosis is established[1].

Mean while, in our patients spiral CT scan of neck reported deep soft tissue involvement and gram positive branched bacteria in stain. Blood chemistry was normal, CBC may show anemia and mild leukocytosis and ESR often is elevated[5]. Our patient's lab data were normal. Computed tomography (CT) usually reveals an infiltrative, well- or ill-defined mass with inflammatory changes[4], and the patient's cervical spiral CT scan indicated as Parotid glands and submandibular glands are normal. Para pharyngeal and ptyrgopalatin fossa are normal, necrotizing and calcified lymphadenopathy are exist from anterior to left side of carotid sheet and left posterior cervical chain reported.

Antibiotic treatment of actinomycosis is successful in 90% of the cases. The treatment of choice actionmycosis is 12 to 24 million units of PNC-G intravenously for 2 to 6 weeks, followed by oral therapy with PNC-V 500 mg every 6 hours for 6 to 12 months. At first intravenous PNC-G was administered for one month and patient's lesions showed acceptable results. Then the patient was discharged in good condition with oral PNC-V.

In this report we discuss from 5 points of view: Cervicofacial actinomaycosis is more prevalent than other forms of actinomycosis. Secondly, one of rare forms of this disease is involvement of bronchial cleft[1]. Base on sonography the patient has been involved in this site. Thirdly, a sample of aspirated secretions was reported positive for AFB during investigation of TB in a reference laboratory, but our patient's responded perfect to PNC without any anti TB drugs and he had no finding positive for TB in pathologic exam. In addition, it is interesting that although, in our country in patient with cervical masses we should primarily consider TB, (as this patient was AFB positive) it is not always true. Our patient's radiologic lesions had started from deep cervical tissues and these lesions were multiple and not lymphadenopathy and finally no TB granoloma was reported on pathologic exam. It s needed to be mentioned that, tuberculin skin test was negative and patien's lesions improved without anti TB treatment. All these show that patient clinical findings are always more important than just a positive AFB report and we should be aware of this matter. Two request of CBC were sent one week from each other reporting 19% and 12% mixed cell respectively. When sysmex equipments is used for CBC, differentiation of blood cells are reported as neutrophil, or lymphocyt and mixed cell, in which mixed cell include eosinophil, basophil and monocyte. In this case, peripheral blood smear should be requested to verify what the differentiation is.

Fifthly, although in some reports short-course treatment actionmycosis have been reported successful, it is better to apply long-term treatment. As short-course treatment associated with relapse and treatment failure.

Conflict of interest statement

We declare that we have no conflict of interest.

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