Head and Neck Mycetoma

SIRIPORNCHAI SUPANAKORN, M.D.*,
ADISORN PATRADUL, M.D.***,
AMNUAY CUTCHAVAREE, M.D.*
SANUN RUNGRUXSIRIVORN, M.D.**,
WINAI WADWONGTHAM, M.D.*,

Abstract

Two cases of eumycotic mycetomas in the head and neck region are reported. The first case is the localized mycetoma which involved only the soft tissue of the neck. It was completely excised and the patient needed no further treatment. In the second case, the lesion extensively involved the structures in the parapharyngeal space, submandibular space and carotid sheath. The upper limit is the skull base. The patient was treated by combination of wide excision with radial forearm flap and antifungal agents, however, the prognosis is fair.

Mycetoma is a clinical syndrome of localized, indolent, deforming, swollen lesions and sinuses, involving cutaneous and subcutaneous tissues, fascia and bone. It is an exogenous disease in which an organism is implanted into the host tissue through a break in the skin caused by a sharp object, most commonly thorns[1,2].

Etiologic agents may be either bacteria (actinomycotic mycetoma) or fungi (eumycotic mycetoma). With the exception of *Actinomyces israelli* all are soil saprophytes or plant pathogens and gain entrance to the dermis through abrasion or implantation[1].

Endemic areas are the mycetoma belt between latitudes 15°S and 30°N (Sudan, Somalia, Senegal, Mexico, India and Central and South America). Other areas include the Mediterranean region, Greece, Italy, Rumania, the Caribean Islands, Cambodia and Iran. Cases are sporadically seen in Europe and the United States[1,3]. Four cases of mycetoma of the foot with emphasis on roentgenological findings were reported in Thailand, how-

* Department of Otolaryngology,
** Department of Pathology,
*** Department of Orthopedics, Faculty of Medicine, Chulalongkorn University, Bangkok 10330, Thailand.
† Presented at the Thai Physicians Association of America-TPAA meeting, Melia Hotel, Hua-Hin, Thailand, July 4, 1996.

ever, mycetoma in the head and neck has never been reported[4]. There has been no report of the effect of spread of human immunodeficiency virus infection in Africa on the incidence of the disease[3].

We present herein two cases of eumycotic mycetomas in the head and neck region.

REPORT OF CASES

Case 1

A 44-year-old Thai female presented with a slow growing painless mass on the right side of the neck of three months' duration. Physical examination revealed nontender ill defined matted masses at the right supraclavicular region. They were firm and slightly fixed to the overlying skin. Complete excision was performed on Aug.16, 1989. Operative findings were three matted lymph nodes containing black granules, however, the culture disclosed no growth. The patient received no further treatment and recurrence has not been observed.

Case 2

A 45-year-old Thai male farmer from Surin Province in northeastern Thailand was first seen in Mar 1991 having a slow growing mass on the left side of the neck for a month. Past history revealed that he had had a left neck mass removed at Surin provincial hospital four months previously and was treated with an undetermined antifungal drug.

Physical examination revealed a nontender, firm and nodular mass involving the left submandibular region. Wide excision was performed and the lesion was found containing black granules. The section showed diffuse inflammatory cell infiltration with a few foci of basophilic clumps of organisms. The arrangement was similar to sun ray appearence with a homogeneous density in the center and radiating at the periphery. (Fig. 1, 2) Identification of the causative agent was carried out and the report was *Madurella grisea*. Postoperatively ketoconazole at the dose of 400 mg/day was then administered for a month.

He was lost to follow-up and returned a year later (Apr 1992) with a sinus tract. (Fig. 3) He was then treated with fluconazole at the dose of 200 mg/day for 5-6 months then lost to follow-up again. In October 1995 he was treated as an out-patient with itraconazole (Sporal)® at the dose of 400 mg/day for 3-4 months.

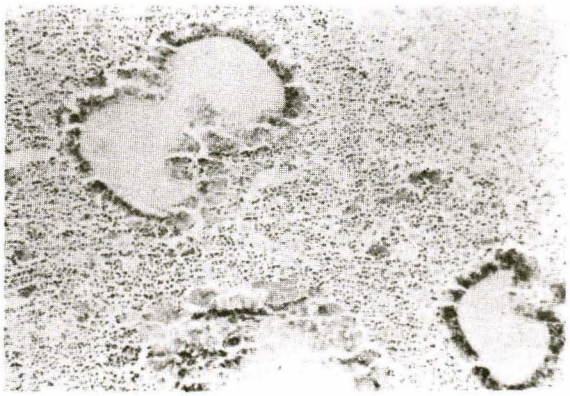

Fig. 1. The section shows diffuse inflammatory cell infiltration with few foci of basophilic clumps of organisms The arrangement is similar to sun ray appearance with a homogeneous density at the center and radiating at the periphery. H&E 100 X

Fig. 2. Special staining of the section with Gommori methenamine silver demonstrates the fungal hyphae and spores. GMS 400 X

The lesion continued growing and deviation of the tongue was noted. M.R.I. on Feb. 1996 revealed the mass surrounded the carotid space, extending to the parapharyngeal and sublingual spaces, and had also infiltrated the left side of the tongue, stylohyoid, sternohyoid and omohyoid muscles. (Fig. 4)

Fig. 3. The patient with nodular infiltration and two sinus tracts at left submandibular region.

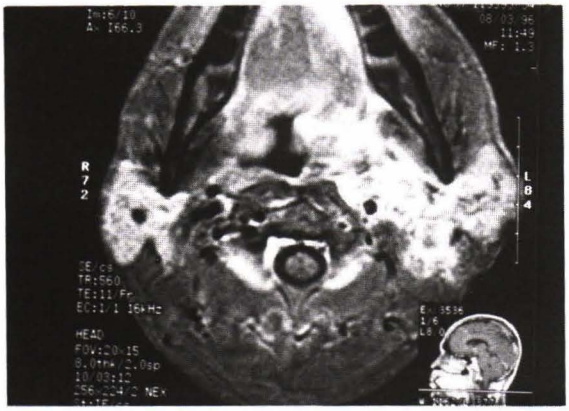

Fig. 4. MRI showing the extent of the lesion.

The patient underwent wide surgical excision with radial forearm flap. Operative findings revealed the lesion extensively involved the structures in the parapharyngeal space, submandibular space and carotid sheath. The upper limit is the skull base. Postoperatively additional amphotericin-B was administered.

DISCUSSION

Clinically, mycetoma is characterized by the triad of tumefaction, draining sinuses and grains. The most common site is the foot; however, in endemic areas other rarer sites may be affected. In the endemic areas, e.g. Sudan, Senegal and Mexico 2-3.75 per cent involved the head and neck. In the head and neck region they must be differentiated from actinomycosis, botryomycosis, chronic osteomyelitis and tuberculosis[1,2].

This lesion may involve connective tissue and bone, but tendons, muscles and nerves are usually spared[5]. In our second case the hypoglossal nerve as well as other soft tissues were involved.

Grossly, actinomycotic agents are more likely to disseminate and produce extensive involvement of bone and muscle than the eumycotic agents. Actinomycotic mycetomas also have a more persistent pyogenic reaction and more commonly involve the head and neck region; most commonly, the mandible[5].

Diagnosis can be made by histological examination of the surgical specimen using hematoxylin and eosin or periodic acid-Schiff stains to identify the causal organism and demonstrate the type of local tissue reaction against it. Confirmation is possible by demonstrating significant levels of antibodies against the different types of causal organisms by counter-immunoelectrophoresis, immunodiffusion or enzyme-linked immunoabsorbent assay[3].

The majority of head and neck mycetoma are caused by actinomycetes, particularly *S. somaliensis* followed by *A. madurae, A. palletieri* and *M. mycetomatis*[2]. The causative organism in our first case could not be identified and the second case revealed *Madurella grisea*, however, the number of our patients is small.

In regard to prognosis; actinomycotic disease is more amenable to treatment than are eumycotic mycetomas. Treatment consists of specific medical treatment and conservative surgery; radical surgery is the last resort. Specific medical treatment for actinomycotic mycetoma includes antibiotics and some antimicrobial agents. Amphotericin-B is ineffective in controlling eumycotic mycetoma. Two new medications in use are fluconazole and itraconazole[1,5].

In the second case, the causative fungus was *Madurella grisea*. The recommended treatment for localized eumycetoma due to *M. grisea* is surgical excision supplemented by antifungal agents[2]. A team approach is required, with detailed discussion among the surgeon, infectious disease physi-

cian, pathologist, and microbiologist(5). Our second patient received antifungal medications which included amphotericin-B, fluconazole and itraconazole as well as conservative surgery but did not respond well. Extensive surgery was then performed; however prognosis is still guarded.

(Received for publication on April 29, 1997)

REFERENCES

1. Rippon JW. Medical mycology. Chicago 2 nd. ed. W.B.Saunders, Co. 1982: 79-114.
2. Gumaa SA, Mahgoub ES, El Sid MA. Mycetoma of the head and neck. Am J Trop Med Hyg 1986; 35: 594-600.
3. Fahal AH, Hassan MA. Mycetoma. Br J Surg 1992; 79: 1138-41.
4. Kaewjinda C, Suvansuthi R, Prabhasawat D. Mycetoma : roentgenological findings. J Med Assoc Thai 1974; 57: 379-82.
5. Altman DT, Lubahn JD, Kuhn PJ. A case report and review of mycetoma of the head : a diagnostic and therapeutic challenge. J Hand Surg 1994; 19A: 998-1002.

มัยเซโตมาของศีรษะและคอ†

ศิริพรชัย ศุภนคร, พ.บ., สนั่น รังรักษ์ศิริวร, พ.บ.***
*อดิศร ภัทราดูลย์, พ.บ.***, วินัย แวดวงธรรม, พ.บ.*, อำนวย คัจฉวารี, พ.บ.**

ผู้ป่วยด้วยมัยเซโตมาบริเวณศีรษะและคอ ไม่เคยมีการรายงานในประเทศไทยมาก่อน ผู้รายงานขอนำเสนอผู้ป่วย 2 ราย รายแรกมีก้อนมัยเซโตมาที่เกิดกับเนื้อเยื่ออ่อนบริเวณคอ ผู้ป่วยได้รับการรักษาโดยการตัดเอาก้อนเนื้อออก และไม่กลับเป็นอีก รายที่สองมัยเซโตมาเกิดกับโครงสร้างต่าง ๆ ในบริเวณส่วนลึกของคอ การรักษาประกอบด้วยการผ่าตัดเสริมสร้าง และยารักษาเชื้อรา อย่างไรก็ตามการรักษายังรอการติดตามผล

* ภาควิชาโสต นาสิก ลาริงซ์วิทยา,
** ภาควิชาพยาธิวิทยา,
*** ภาควิชาออร์โธปิดิกส์, คณะแพทยศาสตร์ จุฬาลงกรณ์มหาวิทยาลัย, กรุงเทพฯ 10330
† รายงานในที่ประชุม Thai Physicians Association of America หัวหิน วันที่ 4 กรกฎาคม 2539