Clinical profile and management of craniocerebral *Madurella* mycetoma

To The Editor: We read with great interest the case report published by Goel et al.[4] (Goel RS, Kataria R, Sinha VD, et al: Craniocerebral maduromycosis. Case report. *J Neurosurg Pediatr 10:*67–70, July 2012). These authors reported the case of a 17-year-old girl with a craniocerebral mycetoma caused by *Madurella mycetomatis* (*M. mycetomatis*). The authors are to be congratulated for the excellent result achieved in managing this case.

We would like to take the opportunity to highlight several key points in the clinicopathology and management of cranial *Madurella* mycetoma. Cranial involvement in mycetoma is distinctly uncommon and occurs in less than 4% of cases.[5] According to previous reports, the causative organisms of cranial mycetoma are predominantly actinomycetes: *Streptomyces somaliensis*, *Actinomadura madurae*, and *A. pelleterii*.[2,5] *Madurella mycetomatis*, the major etiologic agent of human eumycetoma, has rarely been implicated, with only 9 cases reported in the English literature (Table 1).

Early in the clinical course of cranial eumycetoma, the patients typically present with painless scalp swellings discharging black grains, or ear and nasal discharge.[7,10] Neurological complications of cranial eumycetoma infection include epilepsy, cranial nerve palsies, brain abscess, and meningitis.[2] The incidence of these complications in cranial actinomycetoma is variable, ranging from 0% to 62%.[5,2] Interestingly, in the 9 cranial *Madurella* mycetoma cases reviewed, 7 patients (78%) had clinical evidence of neurological complications. The diagnosis of cranial mycetoma is often based on mycological studies, cytology, histological examination, and, more recently, molecular techniques. Scans using CT and MRI remain the gold standard for assessing the extent and pattern of bone involvement and intracranial extension and planning management; CT is superior to MRI in detection of early bone changes. A general consensus holds that cranial mycetoma infection almost always involves more than one bone and produces mainly osteosclerotic lesions, with loss of the trabecular pattern and dense bone formation.[5] In contrast, 4 of the 5 cases of cranial *Madurella* mycetoma for which this information is available were localized to one bone. Moreover, 63% of the patients (5 of 8) with cranial *Madurella* mycetoma showed predominantly osteolytic changes, such as cortical erosion, cavity formation, or complete bone lysis, a pattern similar to that described in the context of *Madurella* mycetoma of other sites.[6] It seems, therefore, that the bone tissue response to *M. mycetomatis* infection is generally the same regardless of the site of involvement.

Treatment of cranial eumycetoma is challenging, and early surgical excision with wide margins offers the only chance of cure. Nevertheless, this is often precluded by the advanced stage of the disease at presentation and the deep anatomical location and multiplicity of the lesions. In the current review, 2 of the cases were considered inoperable, leaving an overall cure rate of 50%. We recommend that a meticulous operative technique be used during exposure and resection of cerebral lesions to prevent spillage and seeding of grains into the adjacent operative field. This should be supported with appropriate antibiotic prophylaxis and intraoperative antibiotic wash, given the high incidence of concomitant bacterial infection, particularly in cases of eumycetoma.[1] Additionally, perioperative chemotherapy with azole-class antifungals is essential, as it helps to stabilize the lesion prior to surgery and reduce the risk of recurrence.

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Disclosure

The authors report no conflict of interest.

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TABLE 1: Reported cases of craniocerebral *Madurella* mycetoma*

Authors & Year	Age (yrs), Sex	Duration (yrs)	Site/Pattern of Bone Involvement	CNS Involvement	Imaging	Medical Treatment	Surgical Treatment	Outcome & Follow-Up
Muyunga-Kasengulu et al., 1971	4.5, M	1	rt maxillary & sphenoid; PL	CN III & VI palsies, meningitis	skull x-ray	parahydroxyphenyl salicylamide	cystic mass excision	died
Natarajan et al., 1975	25, M	2.5	rt parietal; PL	lt hemiparesis & focal Sz	skull x-ray & carotid angiogram	none	abscess drainage followed by craniotomy & excision	persistent sinus, residual weakness, no recurrence; 18 mos
Gumaa et al., 1986	49, M	15	NR; PS	NR†	skull x-ray	griseofulvin & procaine penicillin	none‡	poor response; 1 mos
	26, M	1	NR; PS	NR†	skull x-ray	griseofulvin & procaine penicillin	none‡	poor response; 3 mos
Yagi et al., 1998	22, F	"several years"	lt mastoid; PL	CN VII palsy	skull x-ray	griseofulvin & procaine penicillin	radical mastoidectomy	cured; 6 mos
Arbab et al., 1998	26, M	3	NR; PS	headache	CT	NR	NR	NR
Beeram et al., 2008	18, M	1.5	lt parietal; PL	generalized Sz	CT (lt parietal cerebral mass)	itraconazole	excision of involved bone & mass	persistent sinus, no recurrence; 5 mos
Maheshwari et al., 2010	31, M	2	NR	lt hemifacial pain & CN VI palsy	MRI (lt paranasal & cavernous sinus mass)	liposomal amphotericin B	craniotomy & subtotal mass excision	recurrence; 18 mos
Goel et al., 2012	17, F	2	rt parietal; PL	generalized Sz	CT (rt parietal cerebral mass)	voriconazole & terbinafine	excision of involved bone & mass	cured; 6 mos

* CN = cranial nerve; NR = not reported; PL = predominantly osteolytic; PS = predominantly osteosclerotic; Sz = seizure.
† One patient had epilepsy and right-sided hemiparesis.
‡ These patients had advanced lesions, and no definitive surgery was attempted.