It was also pointed out that the changes in calcium, phosphorus and phosphatase that occur in the generalized form of osteitis fibrosa cystica are not seen in the isolated type. Finally, a brief description of the operative procedure carried out was presented and the microscopic picture of the disease described.

REFERENCES

SYSTEMIC BLASTOMYCOSIS WITH SPINAL CORD INVOLVEMENT

CASE REPORT

Robert C. Greenwood, M.D., and Harold C. Voris, M.D.
*Mercy Hospital and Loyola University Clinics, Chicago, Illinois*

(Received for publication December 27, 1949)

This case is reported because the unusual clinical course and pathological aspects make it important not only in the differential diagnosis and treatment of surgical conditions of the spinal cord but also as a rare manifestation of an already uncommon grave disease. Blastomycosis usually takes either a cutaneous or a systemic course. The cutaneous type tends to heal, whereas the systemic type is characterized by chronicity, and widespread infection in the lungs, subcutaneous tissues, bone, and internal organs. Prognosis in this type is extremely grave.

Blastomycosis itself is relatively rare, since up to 1939 Martin and Smith reviewed 347 proved and presumptive cases. In only 16 of these cases was the central nervous system involved. The usual course of such involvement is that single or multiple abscesses develop in the cerebrum, cerebellum, and brain stem, with a basilar purulent meningitis as a part of the generalized systemic variety. However, in 1940, Craig, Dockerty, and Harrington reported an intravertebral and intrathoracic extradural mass that simulated a dumb-bell tumor. It proved to be a blastomycotic lesion and it is probably the first case report of such a lesion simulating a tumor in the spinal canal. In Friedman and Signorelli’s case the blastomycosis involved only the meninges of the brain and spinal cord. Detailed descriptions of the usual course and pathology of both types of blastomycosis have been described by Montgomery and Ormsby, Wade and Bel, and Stober. With regard to the identification and isolation of Blastomyces dermatitidis and differential features of mycotic lesions, Stoddard and Cutler, Martin and Smith, and Hassin have covered the important points.

REPORT OF CASE

J.M., a 40-year-old white woman, referred by Dr. L. Kratz, was admitted to Mercy Hospital on March 7, 1946, with a diagnosis of myeloma or spinal cord tumor. Her chief complaints were numbness beginning in the calves of the legs and reaching the waist, and progressive
BLASTOMYCOSIS WITH SPINAL CORD INVOLVEMENT

weakness of both legs with difficulty in walking. Both complaints had developed over a 3-week period prior to admission. For 3 years, she had experienced mild mid-dorsal pain in the back that was relieved by ordinary analgesics and was not aggravated by coughing or straining. She had had a fractured right rib 8 years before, and blastomycosis of the skin of the face and upper extremities 4 to 5 years prior to admission. Biopsy of the skin at that time was reported as blastomycosis. The skin lesions healed rapidly and she was apparently well until the onset of the present illness.

Examination. There were several small white scars on the left cheek of the face. Neurological findings were: hypesthesia of the lower limbs and trunk with a level at the 7th thoracic dermatome, absent vibration sense in the left leg, weakness of the trunk muscles and lower limbs, equivocal Babinski signs, and ankle clonus bilaterally.

Roentgenograms of the spine showed erosion of the pedicles of the 6th and 7th dorsal vertebrae with impairment of those of the 8th body. A discrete punched-out area, about 1 cm. in diameter, was found in the head of the 7th rib on the right side (Fig. 1). A routine chest plate was reported to have a light homogeneous density about 6 cm. in diameter, irregularly outlined, but extending out from the right lung root and fading in the parenchyma of the right lung (Fig. 2). Urine analysis was normal. Rbc. 3,850,000; Hb. 74 per cent; Wbc. 7,200 with 54 per cent polys, 40 per cent lymphocytes, 1 per cent eosinophils, and 3 per cent monocytes.

Operation. On March 9 an extensive laminectomy and decompression of the spinal cord with partial removal of an extradural granuloma was carried out. Purulent material exuded from a cavity in the articular process of the 7th rib. The upper and lower limits of the gran-
uloma were uncovered opposite the 3rd and 8th vertebral bodies respectively. The lesion was thickest on the right side and opposite the 6th and 7th bodies, where it was 1 cm. thick on the dorsal surface.

The postoperative course was uneventful, and she was dismissed on April 1, 1946, with moderate improvement in neurological function in that the hypesthesia had receded and she could walk with assistance.

Pathological Report. The surgical specimen was a granuloma with single and budding forms of a pathological yeast rather sparsely distributed. Culture of the fresh material on Sabouraud’s media yielded a growth of Blastomyces dermatitidis.

Further Course. Soon after dismissal from the hospital, the neurological complaints recurred, with loss of visceral sphincter control. She was readmitted on May 10, 1946. She had an analgesic level at the umbilicus, spastic paraplegia with mass reflexes of both legs, and an atonic, incontinent bladder.

Lumbar puncture revealed a complete block. Only 5 cc. of CSF could be obtained; this contained 8 cells/c.mm., 300 mg. per cent total protein, and numerous round to oval bodies with a doubly refractile ring found on direct smear.

The patient received nine deep X-ray treatments while in the hospital, but she became very depressed and desired to return home. She was dismissed on June 10, 1946.

Subsequently a small draining sinus developed in the dorsal region of the back. During June 1946, she was examined at another institution where cultures from the sinus were reported to be those of Blastomyces. The course of her illness was progressively downward, and still later she was hospitalized in her home city, where Blastomyces were again cultured from the sinus. She died about October 1946.
Autopsy. There were chronic inflammatory granulomatous lesions in the lungs, and thickened dura along the spinal cord, with elevated patches of grayish tissue on the dura for about 14 cm. of the thoracic region. About 20 cm. from the conus, this thick gray tissue entirely encircled the cord. The dura was about 1 mm. thick in this region.

Microscopic Examination. The tissue at the mid-dorsal region in the extradural space contained numerous foci of polymorphonuclear neutrophils surrounded by a zone of macrophages and fibroblasts. Multinucleated cells were numerous in some areas as well as new capillaries. Rather sparsely distributed were round to oval doubly refractile bodies, some single and others with a single bud in these foci of neutrophils. The dura was adherent to this tissue and very thick, with numerous capillaries surrounded by small round cells and polymorphonuclear neutrophils.

The spinal cord itself showed no changes other than softening and degeneration of its columns following vascular changes from the cuff of granuloma about it. There were no significant findings in the brain.

Diagnosis. It was believed that the microscopic abscesses, the predominance of polymorphonuclear neutrophils and chronic inflammation were features that differentiated the lesions from tuberculosis. A pathological yeast was considered more likely to be the causative agent since different laboratories isolated the organism and reported it to be Blastomyces dermatitidis. This organism was further differentiated from Torula histolytica by staining the sections with toluidine blue, which brings out the typical capsule and its numerous spikes, spines, or spicules of the Torulace. This morphology was not demonstrated in the present case; the organisms remained oval or round with a doubly refractile ring (Figs. 3 and 4). The presence
of single budding forms differentiated this yeast from Coccidioides immitis; the latter organism never exhibits budding. A peripachymeningitis with extradural abscesses simulating spinal cord tumor, due to Blastomyces dermatitidis, was probably the primary lesion.

COMMENT

This patient was apparently cured of her skin lesions of blastomycosis. After a history of mild non-progressive pain in the mid-dorsal region for about 3 years, there rapidly developed a neurological deficit that caused her to seek attention. In the absence of signs and symptoms of systemic involvement, a diagnosis of a spinal cord tumor was very probable. The clinical manifestations and roentgenograms seemed to confirm this diagnosis. Also, the lack of systemic involvement by the Blastomyces made it difficult to interpret the discrete punched-out area in the head of the 7th rib on the right side. Dr. H. E. Potter in 1914 emphasized the sharp line of demarcation between bone apparently intact and areas of total necrosis. He felt that such a marked localizing destruction was so constantly present in ordinary blastomyecotic lesions, that a search should be made for Blastomyces. However, the lesions are most distinct and numerous in the long bones. The localized nature of the lesions seemed, therefore, to justify surgical exploration.

SUMMARY

An unusual case of systemic blastomycosis is reported. A difficult problem in differential diagnosis was presented by the clinical manifestations of a localized lesion in the spinal canal on the one hand, and the healed cutaneous blastomycosis without apparent evidence of systemic involvement on the other hand. The organism isolated by different laboratories was reported to be Blastomyces dermatitidis and its morphology remained constant when stained with toluidine blue. The previously described roentgenographic characteristics of the bone lesions in blastomycosis are emphasized.

REFERENCES