Actinomycotic osteomyelitis of the skull and epidural space

Case report

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Actinomycosis, a chronic granulomatous disease secondary to a microorganism (Actinomyces israeli) with properties intermediate between those of bacteria and molds, may on occasion involve the central nervous system. The following case represents an unusual neurological manifestation of the disease, with lesions evident in both the cranial epidural space and calvarium.

Case Report

The patient, a 40-year-old Spanish-American farmhand, was admitted to the Colorado General Hospital in July, 1967, because of recurrent generalized seizures of 6 years' duration, usually occurring a day or two after weekend drinking "spree." On the day prior to admission, he had experienced a typical seizure which progressed to status epilepticus.

The past medical history was significant. In 1945, at age 18, he had been hospitalized because of purulent drainage from the socket of a recently extracted left mandibular molar, and treated with intramuscular injections of penicillin, 20,000 units every 3 hours for 8 days. He was rehospitalized 1 year later because of purulent drainage from a cutaneous sinus over the vertical ramus of the left mandible and radiological evidence of osteomyelitis in this area (by report). Cultures of pus from the cutaneous sinus grew staphylococcus aureus, coagulase positive. A bony sequestrum was resected from the left mandibular ramus, but no description of this specimen is available. He was again treated with 20,000 units of penicillin given intramuscularly every 3 hours for 15 days; the mandibular wounds subsequently healed, with no further evidence of purulent drainage from any part of his head.

Examination. On admission in July, 1967, the patient was in an afebrile postictal state, lethargic but easily roused, with a mild left hemiparesis. Neurological deficits cleared rapidly after admission. Examination of the cranial nerves, including funduscopic examination, was within normal limits. His neck was supple. There was a well-healed scar over the left mandibular area and a slight deformity of the mandibular ramus. The skull was slightly asymmetrical, the left parieto-temporal area being more prominent than the right, but there was no local scalp tenderness or fluctuance.

Routine laboratory work including a white cell count, hemoglobin, and urinalysis was normal. Skull films demonstrated gross and relatively uniform thickening of the left lateral vault (parietal, adjacent frontal, occipital) to 30–35 mm, with obliteration of the diploic space and inner table irregularity but no focal osteolysis (Fig. 1). The lesion
crossed the midline to the midright parietal vault, and tomography demonstrated extension into the base to include the entire floor of the left middle fossa to the body of the sphenoid bone, posterior and lateral wall of the left orbit, and the left temporal bone lateral to the otic capsule and base of left pterygoid process (Fig. 2). The sinuses were normal. Minimal lateral cortical thickening of the vertical ramus of the left mandible was present. Films of the pelvis and thorax were normal. Repeat physical examination of the skull and scalp after review of the skull x-rays revealed no bruits to auscultation, but did show a slight increase in scalp temperature over the left parietal area (99.6°F) as compared to the right (98.6°F) as measured by a thermistor. An electroencephalogram was reported as abnormal because of excessive fast activity without focal abnormalities.

The absence of an osteolytic component to the widened vault and the eccentric location of the lesion, in addition to a normal central skeleton, were considered atypical for Paget’s disease. Fibrous dysplasia, however, was considered as an acceptable radiographic diagnosis. A left common carotid angiogram with multiframe subtraction demonstrated 1 cm inferior and 1.4 cm medial displacement of the left cerebrum (as a transfalx herniation from left to right) due to invagination of the vault. No focal left cerebral hypervolumia was present (Fig. 3). Marked enlargement of the left middle meningeal, ophthalmic, and anterior meningeal arteries (and grooves) was noted. The intracranial internal carotid artery and branches were slightly attenuated but uniform in caliber. The cerebral venous drainage was not recognizably deformed or altered, and the circulation time was normal.

Operation. The initial perforator opening of a left parietal craniectomy was placed just posterior to the coronal suture, approxi-
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Fig. 2. Left: Frontal skull x-ray film (Caldwell) demonstrating bilateral thickening of the vault, temporal, orbital, and middle fossa bones. Right: Frontal hypocycloidal tomogram showing mid aspect of the middle fossa.

mately 4 cm from the midline. The calvarium was approximately 1 cm thick and exceptionally dense. After the inner table had been removed, tenacious, fibrous material was encountered which could not be teased away from the dura. Another perforator opening placed just inferior to the first one uncovered yellowish-green pus in the epidural space. A gram stain of this material demonstrated a dense network of violet-staining fibrils with club-like extensions, compatible with either actinomycosis or nocardiosis. The parietal bone was removed in small pieces to accomplish a rectangular craniectomy approximately $6 \times 10$ cm. Bone was thickest in the midparietal area, where it

Fig. 3. Left common carotid angiogram. Left: Arterial phase, frontal projection, showing displacement of the left cerebrum due to invagination of the vault. Right: Arterial phase, lateral projection, showing very large middle meningeal channels (solid arrows), moderately large ophthalmic artery (and anterior meningeal branch), and average size superficial temporal artery (open arrow).
measured 40 mm. The epidural space contained alternating patches of claylike, greenish-yellow fragments with focal deposits of purulent material. At the conclusion of the procedure, the wound was thoroughly irrigated with bacitracin and penicillin, and drains were inserted into the epidural space. The dura was not opened.

Postoperative Course. Anaerobic cultures of both resected bone plate and the epidural deposit grew actinomycosis. The patient's wound healed primarily. Until the diagnosis of actinomycosis was established by culture, treatment consisted of massive intravenous penicillin as well as sulfadiazine therapy. The latter medication was included to cover the eventuality of a nocardial infection. The dosage of penicillin was 16 million units (intravenous continuous drip) daily for 6 weeks. The patient had no seizures or evidence of neurological deficit while in the hospital. He was discharged on oral penicillin G therapy (4 million units per day) as well as anticonvulsants. Since discharge the wound has remained well healed, but he had required frequent rehospitalizations for recurrent seizures and alcoholism. Despite admonishments, he has continued with "spree drinking" and has taken anticonvulsant medication and antibiotics only erratically. Repeat skull films and electroencephalograms demonstrated no significant change over the past 24 months.

Discussion

Human actinomycosis has a well-recognized anatomical distribution, with sites of predilection for the cervicofacial region, abdomen, and thorax. In Cope's series of 1330 cases of actinomycosis, the incidence of cervicofacial lesions was 63%, abdominal 22%, and thoracic 15%, with evidence of central nervous system involvement in only 10 cases, or less than 1%. In contrast, Harvey, et al., reviewed the records of 27 cases of proven actinomycosis at the Johns Hopkins Hospital and found a different distribution; abdominal lesions accounted for 63% of the cases, cervicofacial lesions 24%, and thoracic lesions 13%, while the central nervous system or cranial vault were involved in 15%, but these latter were all secondary to disease in other anatomical sites.

Isolated brain abscess is the commonest neural complication of the disease, and is usually secondary to involvement of the temporal bone or paranasal sinuses. Actinomycotic brain abscess can simulate tumors of the third ventricle or posterior fossa. Extension to the subarachnoid space can induce an arteritis similar to that seen with tuberculous or syphilitic meningitis. Bolton and Ashenhurst have reviewed the pertinent clinical features of 18 cases of central nervous system actinomycosis, the majority of the cases being single brain abscesses with only one case of epidural infection. Clinical experience has demonstrated that surgical excision of an isolated abscess coupled with penicillin therapy offers a reasonably good prognosis.

Actinomycosis of osseous structures in the head and neck, other than the mandible, is rare, but infections in the maxilla, temporal bone, and cervical spine have been reported. In general, the osteoblastic reaction to fungal bone infections is usually less than that seen with bacterial invasion, namely, staphylococcus or streptococcus, but does vary with individual microorganisms and the mechanism of dissemination. Hematogenous spread of fungi to bone generally produces a lytic lesion, whereas direct extension may result in a marked osteoblastic reaction. It is of interest that actinomycosis, a disease resulting from an organism whose classification lies between bacteria and fungi, may produce a marked osteoblastic response in infected bone though a broad spectrum exists. Cryptococcosis results in less bony proliferation than in either blastomycosis or sporotrichidomycosis.

This case presents several interesting diagnostic and therapeutic features. Once an epidural abscess had been identified secondary to either nocardia or actinomycosis, the past history of mandibular osteomyelitis assumed heightened significance and strongly suggested the latter disease. Growth on culture with varying incubation conditions is the only method, however, for resolving this differential diagnosis, and will show actinomycosis to be microaerophilic, and nocardia, aerobic. Differentiation of these two organisms is critical since nocardiosis is best treated with sulfonamides and actinomycosis by penicillin.

The natural history of the disease process
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in this patient may have extended over a 22-year period. The pathogenesis of human actinomycosis is uncertain since the organism pursues an ambient existence in gingival and tonsillar crypts. One theory holds that either a chronically infected tooth or a mandibular injury, such as might occur with a tooth extraction, provides a portal for entry into the mandible. This patient received inadequate antibiotic therapy at the onset of his disease, and since anaerobic cultures of pus were not obtained the organism was not identified. The calvarial and epidural lesions were indolent until intractable seizures finally brought him to neurosurgical attention. It was deemed impracticable to eradicate and resect the entire epidural infection and involved bone, and the process has remained controlled on long-term, but intermittent, antibiotic therapy.

Summary

A case of actinomycosis of the skull and epidural space has been reported, presenting a 6-year history of seizures that started 22 years after a presumed actinomycotic infection of the mandible. Radiographic studies and operative findings demonstrated remarkable thickening of the left hemicalvarium and base associated with deformity and displacement of intracranial contents. This and other types of actinomycotic infections of the nervous system have been reviewed and the therapy discussed.

References


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