Myospherulosis of the Mandible Presenting as a Multilocular Lesion: A Case Report and Review of the Literature

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Abstract Myospherulosis is an uncommon foreign body reaction that occurs in tissues exposed to antibiotic and oil-based ointments. Radiographic imaging of this lesion may suggest other diagnoses. Treatment consists of conservative surgical removal, and is usually curative. We report herein a case of myospherulosis presenting as an extremely unusual multilocular radiolucent lesion.

History

A 74 year-old Caucasian female was referred to the oral pathology clinic of our institution by her dentist for the evaluation of an asymptomatic lesion in the right mandibular ramus. The lesion was detected as an incidental finding on a routine panoramic radiograph. The patient denied having any previous surgery in the involved area, aside from an uneventful third molar extraction in her youth. Her relevant medical history includes high blood pressure, eczema and alleged contact allergies to certain metals. She does not smoke or drink alcohol. Clinically, there was no expansion of the cortices or any significant mucosal anomaly.

Radiographic Features

A panorex and a cone-beam computed tomography (CBCT) were ordered and showed a well-defined, non-expansile, multilocular radiolucency of the right posterior mandible, above the lower alveolar nerve (Fig. 1a, c). The anterior portion of the lesion was in continuity with an area of diffuse medullary sclerosis, consistent with localized condensing osteitis. Surgical exploration and a bone biopsy were performed under local anesthesia after informed consent. During the procedure, a lobulated cavity was identified and contained a black, viscous, tar-like substance (Fig. 2). The mandibular lingual cortex was eroded in several areas (Fig. 1b, d). There was no cystic lining or any other sign of pathology.

Diagnosis

Histologically, the entire specimen consisted of black spherules of varying size, admixed with amorphous eosinophilic debris, consistent with a diagnosis of myospherulosis (Fig. 3). The whole cavity was conservatively debrided and the patient showed no sign of recurrence 7 months later.

Discussion

Myospherulosis, also known as spherulosis or spherulocytosis, consists of a rare foreign-body reaction in tissues more often resulting from a contact with petrolatum-based products. The diagnosis of myospherulosis is confirmed by histology and can be detected by imagery. Myospherulosis has been reported in various anatomic sites, including the

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Fig. 1 **a** Panoramic view of the right mandibular ramus presenting as a multilocular radiolucency, located above the lower alveolar nerve. **b** Coronal, **c** vertical oblique and **d** axial views of CBCT displaying a multilocular lesion in the right mandibular ramus

Fig. 2 Oil-based substance removed from bone and floating in buffered formalin

nose, paranasal sinuses, eye, upper eyelid, oral cavity, lower lip, brain, liver, skin, breast and kidney [1–10].

Myospherulosis was initially reported by McClatchie et al. in 1969 [4]. The condition was termed “myospherulosis” because of its location in skeletal muscle and

Fig. 3 Characteristic histologic appearance of black spherules of varying size and amorphous eosinophilic debris (hematoxylin-eosin, magnification $\times 20$)

histologic features. The latter includes the formation of “spherules”, which appear in cystic-like spaces, known as “parent bodies”. Foreign body-type giant cells and eosinophilic infiltrates may be present [7].

The first case of myospherulosis in the United States was identified by Kyriakos in 1977 [1]. He described sixteen cases of “spherulosis” lesions in areas of previous surgery in the nose, paranasal sinuses and middle ear. The causative agent was thought to be an unidentified endospore-forming fungus. His hypothesis emerged from the common surgical use of gauzes filled with unsterilized petrolatum-based antibiotic ointment.

A year later, Rosai [11] stipulated that the spherules were not actually of fungal origin but rather derived from the contact of erythrocytes with lipid-containing material, the brownish color being due to the degradation of hemoglobin.

The first cases involving the jaws were reported in 1980 by Dunlap and Barker [12]. Later, Lynch et al. [13] reported six additional cases in the oral cavity.

Most cases of oral myospherulosis are thought to originate from the use of tetracycline-containing petrolatum-based ointments at the site of dental extractions. Antibiotic ointments are usually placed in the alveolar bone in order to prevent alveolar osteitis. This practice is controversial and the efficiency of petrolatum-based ointments remains to be statistically demonstrated [14]. Most cases of myospherulosis appear to be iatrogenic in origin, although other factors may be linked, such as the release of endogenous lipids via trauma and, putatively, parasitic infection [15]. Sindwani et al. [16] suggest that patients who develop myospherulosis could have lipid sensitivity and display a greater tendency towards immune-mediated reactions.

Table 1 Differential diagnosis for multilocular radiolucencies of the jaws

	Site of predilection	Typical radiographic features
Myospherulosis	Mandible	Non-expansile
Ameloblastoma	Mandibular molar-ramus area	Expansile with thick, angulated septae
Odontogenic keratocystic tumour	Mandibular molar-ramus area	May be multifocal in Gorlin syndrome
Cherubism	Posterior mandible and maxilla	Symmetrical and bilateral
Central giant cell granuloma	Mandibular region, anterior to first permanent molar	May be multifocal in hyperparathyroidism Fine septae at a 90° angle and expansile
Odontogenic myxoma	Mandible	Fine septae at a 90° angle and expansile “Honeycombed” pattern
Hemangioma	Mandible, zygoma and orbital region	Expansile
Central mucoepidermoid carcinoma	Mandible	Expansile
Botryoid odontogenic cyst	Anterior region of mandible	Well-delineated “grape-like” pattern

Myospherulosis of the jaws typically occurs within bone, at the site of extraction of mandibular third molars [17]. It is usually discovered as an incidental finding on dental radiographs, and may present clinically as an asymptomatic soft tissue swelling. In rare cases, patients may present pain and purulent discharge. Surgical exploration reveals black, greasy, tar-like foreign material in association with a bony defect [18]. Radiographically, myospherulosis presents as a well-circumscribed unilocular radiolucency, although our case presented as an extremely unusual multilocular radiolucency. The differential diagnosis for multilocular radiolucencies includes the following: ameloblastoma, odontogenic myxoma, odontogenic keratocystic tumour (odontogenic keratocyst), cherubism, central giant cell granuloma, central mucoepidermoid carcinoma, hemangioma and other rare lesions (Table 1). In our opinion, this list should include myospherulosis, whenever there is a history of previous extraction, especially the site of mandibular third molars. Surgical exploration and biopsy is required for final diagnosis. Myospherulosis might be overlooked in dentistry on account of its rare occurrence and lack of associated symptoms.

Treatment of myospherulosis consists of complete conservative removal of the foreign material with curettage of the affected area. Recurrence is rare. All excised tissue should be submitted for histopathologic examination in order to rule-out superimposed pathologies, such as aspergillosis [19]. Immunostaining for hemoglobin or antibody-based staining methods may be used to distinguish myospherulosis from endospore-forming organisms, such as *Coccidioides* and *Rhinosporidium seeberi* [20].

The common denominator of myospherulosis in the literature is the use of a topical antibiotic ointment [12, 17, 21–23]. This raises questions on the safety of antibiotic

ointments. Water-soluble antibiotic pastes, as opposed to petrolatum-based ointments, may reduce the risk of granulomatous foreign-body reactions [16, 24], but there is no clear evidence that myospherulosis may be avoided.

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Compliance with Ethical Standards

Conflict of interest All authors declare that there are no financial conflicts associated with this report and that there is no funding source.

References

- Kyriakos M. Myospherulosis of the paranasal sinuses, nose and middle ear: a possible iatrogenic disease. *Am J Clin Pathol.* 1977;67:118–30.
- Kini U, Babu MK. Ocular spherulocytosis. *J Clin Pathol.* 1996;49:857–8.
- Biedlingmaier JF, Aronsky MA, Whelan PJ. Myospherulosis of the upper eyelid as a complication of endoscopic sinus surgery. *Am J Rhinol.* 1997;11:345–7.
- McClatchie S, Warambo MW, Bremner AD. Myospherulosis: A previously unreported disease? *Am J Clin Pathol.* 1969;51:699–704.
- Hornie N, Shimoyama T, Ide F. Myospherulosis of the lower lip. Report of a case. *Int J Oral Maxillofac Surg.* 1993;22:234–5.
- Mills SE, Lininger JR. Intracranial myospherulosis. *Hum Pathol.* 1982;13:596–7.
- Lui PC, Luk IS, Lee CK, Lui YH, Leung CY, Choi CH. Hepatic myospherulosis complicating portal vein embolization. *J Clin Pathol.* 2004;57:155–8.
- Waldman JS, Barr RJ, Espinoza FP, Simmons GE. Subcutaneous myospherulosis. *J Am Acad Dermatol.* 1989;21:400–3.
- Ferrell LD. Myospherulosis of the breast. Diagnosis by fine needle aspiration. *Acta Cytol.* 1984;28:726–8.
- Le Gall F, Huerre M, Cipolla B, Shalev M, Ramee MP. A case of myospherulosis occurring in the perirenal adipose tissue. *Pathol Res Pract.* 1996;192:172–81.

11. Rosai J. The nature of myospherulosis of the upper respiratory tract. *Am J Clin Pathol*. 1978;69:475–81.
12. Dunlap CL, Barker BF. Mysospherulosis of the jaws. *Oral Surg Oral Med Oral Pathol*. 1980;50:238–43.
13. Lynch DP, Newland JR, McClendon JL. Myospherulosis of the oral hard and soft tissues. *J Oral Maxillofac Surg*. 1984;42:349–55.
14. Kolokythas A, Olech E, Miloro M. Alveolar osteitis: a comprehensive review of concepts and controversies. *Int J Dent*. 2010;. doi:[10.1155/2010/249073](https://doi.org/10.1155/2010/249073).
15. Kühnel TS, Kazikdas KC. Spherulocytosis of the maxillary sinus: a case report. *Auris Nasus Larynx*. 2006;33:461–3.
16. Sindwani R, Cohen JT, Pilch BZ, Metson RB. Myospherulosis following sinus surgery: Pathological curiosity or important clinical entity? *Laryngoscope*. 2003;113:1123–7.
17. Wallace ML, Neville BW. Myospherulosis. A case report. *J Periodontol*. 1990;61:55–7.
18. Fisher SC, Horning GM, Hellstein JW. Myospherulosis complicating cortical block grafting: a case report. *J Periodontol*. 2001;72:1755–9.
19. Panayiotopoulou M, Freedman PD, Weber F, Lumerman H. The synchronous occurrence of aspergillosis and myospherulosis of the maxillary sinus. Report of a case with review of literature. *Oral Surg Oral Med Oral Pathol*. 1987;63:582–5.
20. Travis WD, Li CY, Weiland LH. Immunostaining for hemoglobin in two cases of myospherulosis. *Arch Pathol Lab Med*. 1986;110:763–5.
21. Belfiglio EJ, Wonderlich ST, Fox LJ. Myospherulosis of the alveolus secondary to the use of Terra-Cortril and Gelfoam. Report of a case. *Oral Surg Oral Med Oral Pathol*. 1986;61:12–4.
22. Ide F, Saito I, Otsuka K, Hori M, Kudo I. Myospherulosis of the mandible. Report of a case and review of the literature. *Int J Oral Surg*. 1984;13:65–8.
23. Moore JW, Brekke JH. Foreign body giant cell reaction related to placement of tetracycline-treated polyactic acid: report of 18 cases. *J Oral Maxillofac Surg*. 1990;48:808–12.
24. Culver WT, Leonard DW, Wilhelmsen CL, Bolger WE. Experimental myospherulosis of the paranasal sinuses: a histologic rabbit study. *Am J Rhinol*. 2000;14:131–7.