Persistent orbital inflammation following complete excision of deep dermoid cysts.

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Abstract

Piecemeal excision of dermoid cysts carries the risk of implanting epithelial fragments into orbital fat, which is well-recognized to continue secreting oily debris, inciting chronic, often granulomatous inflammation. The authors present the clinical and histological details for two patients with persistent lipogranulomatous inflammation for years after piecemeal excision of deep orbital dermoid cysts, in the absence of any residual epithelium. The importance of copious saline lavage -- to 'float-out" and reduce microscopic lipid droplets is also emphasised.

In the absence of infection, inflammation after excision of a dermoid cyst will arise from the response to necrotic tissues or to extracellular lipid outside the cyst lumen. Whilst the clinical and histopathological features for spontaneous lipid leakage from the cyst lumen -- causing chronic, often granulomatous, inflammation within the cyst wall and neighboring tissues -- is very well recognized and widely reported,[1-5] – there does not appear to be any description of the pathological changes of persistent inflammation after excision of deep orbital dermoids ***and in the absence of any residual epithelium***. Deep orbital dermoid cysts often occupy multiple cavities within the orbit and surrounding bones, making intact excision very unusual.[4]

We present the clinical and histological details for two patients who required repeated anterior orbitotomy to settle persistent orbital inflammation after complete epithelial excision of deep orbital dermoid cysts. The study adhered to the tenets of the Declaration of Helsinki.

Clinical Cases

Patient 1 A 21-year-old female presented with a 4-year history of a palpable, non-tender mass in the right lacrimal gland area. She had 3mm right relative proptosis, but normal visual functions and ocular motility. CT confirmed a large intraosseous dermoid in the greater wing of the sphenoid (Figure 1A). The cyst was removed through an upper eyelid skin-crease incision, it being necessary to evacuate the contents during surgery to allow access to the deep aspects of the mass; all of the epithelium was removed, any bone surfaces thoroughly cleaned with dry swab and any of the evident free lipid/keratin content was aspirated from the surgical field.

Although visual functions, exophthalmometry ocular motility remained normal, the patient had persistent and variable upper eyelid swelling and redness after surgery, this not settling with 3 short courses of oral corticosteroids. Imaging showed some patchy opacities near the right orbital roof, compatible with mild chronic inflammation. The possibility of an area of sequestered residual epithelium prompted a re-exploration of the area at 18 months after first surgery: at surgery a “woody” periosteum, with abnormal white nodules and abnormal neighboring orbital fat, was located and some of these nodules exuded a yellow, paste-like, “oily” fluid during excision. Histopathological examination revealed mild chronic inflammation of the orbital fat, with some multinucleate foreign-body giant cells,

and large intercellular lacuni from which free lipid had been washed out during processing (Figure 1B); the lacrimal gland also displayed granulomatous inflammation, probably secondary to the inflammation in the neighboring orbital fat and periosteum (Figure 1C). There was no evidence of any residual epithelium from the original dermoid cyst, and the orbital inflammation settled rapidly after excision of these abnormal tissues, with no recurrence over 19 years after surgery.

Patient 2 Having had 4 years of left upper lid swelling and proptosis, a 31-year-old male was found, on CT scan, to have a well-defined extraconal cyst with scalloping of the adjacent orbital roof (Figure 1D). A large, highly-inflamed dermoid cyst was adherent to the surrounding tissues and was drained and excised through an upper lid incision; histopathology showed a chronically-inflamed dermoid cyst.

Five years later, the patient reattended with 6 months of left upper eyelid swelling and slight redness; the lid was tender and swollen, with 2mm left relative proptosis and slight conjunctival chemosis. CT scan showed a small low-density mass superolaterally within the left orbit and patchy localized opacities, compatible with inflammation (Figure 1E); the symptoms failed to settle with a course of flurbiprofen. Inflamed, hyperemic tissues containing firm yellow cysts were found alongside the lacrimal gland at repeat orbitotomy, histopathological examination extensive fibrosis with empty spaces associated with foamy macrophages -- an appearance suggestive of spilled lipid ingested by these macrophages, without any epithelium being present (Figure 1F). The inflammation improved rapidly and there has been no further inflammation over 16 years after surgery.

Discussion

It is widely documented that epithelial desquamation and accumulation of glandular secretions causes slow enlargement of dermoid cysts,[2,3] and that lipid seepage from unoperated cysts causes chronic inflammation and foreign-body giant-cell granulomas. [2,3,6,7] Intramural and pericystic inflammation is common, even in childhood,[2,5] and pericystic lesions within the surrounding fat have been termed ‘satellite inflammatory pseudocysts’.[3]

Inadvertent intra-operative rupture of dermoids occurs in 10-28% of cases[1,5,8] and deep orbital or intraosseous dermoids, being larger or multiloculated, generally need piecemeal removal.[4,6,9] Such

piecemeal removal carries risk of sequestering epithelial fragments into orbital fat, such implanted epithelium being well-recognized to continue secreting oily debris that incites a chronic, often granulomatous, inflammation.[3,5,6,7,10] Inflammation persisting for years after complete excision of the epithelial lining of a dermoid cyst does ***not***, however, appear to have been reported, and we describe the clinical course, imaging and histopathology for two such patients.

Although impossible to exclude clearance of minimal residual epithelium through inflammatory cytolysis, it would seem likely that the diffuse chronic inflammation (shown at re-exploration) was due to a persistence of small amounts of lipid spilled at initial surgery. Extracystic lipid (as a result of iatrogenic or spontaneous rupture of intracranial or spinal dermoids) has been reported to induce recurrent or persistent inflammation in cerebrospinal fluid channels, this being asymptomatic, or causing headache, seizures, cerebral ischemia, and aseptic meningitis.[11,12]

Prior to these cases, it had been our practice to clear visible debris from the operative field by aspiration, and by "swab-abrasion" of the bone. Since these two cases, we now also copiously irrigate the operative field with (~60ml) saline after piecemeal excision of any cyst -- when it will be seen to "float out" a very large number of fine lipid droplets that are otherwise missed with simple aspiration. Since adopting this saline-lavage, we have not observed another case of lipid-induced chronic orbital inflammation in about 150 procedures. Although irrigation after cyst rupture has been recommended,[1,3,5,6,10] the vast majority of specialist surgical textbooks do <u>not</u> suggest this practice. In a series of 27 dermoid cysts ruptured during surgery, 20 were managed with suction and irrigation and one (3.7%) developed persistent inflammation.[6]

Our cases provide the first histological report of inflammation persisting for years after piecemeal excision of dermoid cysts, apparently in the absence of any retained epithelium -- in other words, inflammation due to the persistence of free lipid, rather than lipid release from persistent epithelium. It also appears likely that our original technique for removing spilled lipids failed to clear micro-droplets, and the importance of copious saline lavage -- to 'float-out" and reduce microscopic lipid droplets -- is highlighted by our subsequent experience.

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Legends

Figure 1

(Patient 1) CT scans showing **(A)** a deep right orbital dermoid cyst before excision, involving the greater wing of the sphenoid of the orbital fat. **(B)** After 18 months of intermittent orbital inflammation, excision of abnormal periosteum from the superotemporal quadrant showed obliterated fibrotic fat, mild chronic inflammation (arrow), clusters of histiocytes, and a multinucleate giant cell (encircled), without any epithelial remnants being present. **(C)** The lacrimal gland showed well-formed granulomata (encircled) and an intervening lymphoplasmacytic infiltrate (arrow).

(Patient 2) CT scans of **(D)** a large dermoid cyst expanding the left orbital roof, and **(E)** persistent patchy opacity of orbital fat 5 years after its excision (encircled). **(F)** Removal of these areas of abnormal orbital fat showed a dense fibrotic stroma with chronic inflammation (white arrow), including lipid-laden histiocytes (black arrow). Removal of these abnormal tissues led to a rapid and complete resolution of symptoms.

All photomicrographs stained with Hematoxylin & Eosin (x10 objective power).