MAXILLARY SINUS XANTHOGRANULOMA

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XANTHOGRANULOMA is rare and is essentially an inflammatory granulomatous lesion, which is tumour like and may be found in the mediastinum, retroperitoneum, lymph nodes, bones and rarely elsewhere.

The ætiology and pathogenesis of xanthogranuloma is not well understood. Three theories have been put forth explaining the genesis of the lesion.

According to the first theory fundamental derangement in the fat metabolism precipitates lipins in certain tissues. These lipins in turn initiate an inflammatory reaction and this inflammatory tissue rich in histiocytes further favours retention of lipin. Establishment of such a vicious circle leads to the formation of xanthogranuloma. The first theory as quoted by Oberling has been defended particularly by Chester (1935) who believes xanthogranuloma formation to be simply due to the deposition of lipids. Against this in xanthogranuloma, the clinical picture is distinctly inflammatory. The experimental work by Letterer (quoted by Oberling) suggests this theory is wrong.

The second theory suggests that the lesion is primarily inflammatory in nature consisting of irritative proliferation of histiocytes which later become charged with lipins.

According to the third theory, both an irritative inflammatory and a degenerative metabolism work together favouring the formation of xanthogranuloma.

The xanthogranulomatous lesion is characterized by an extensive infiltration of plasma cells, lymphocytes and macrophages with areas of sclerosis. Foam cells loaded with neutral fats and cholesterol are present in varying concentration in different areas. At places the proliferation of histiocytes is so marked as to simulate a tumour. The fibroblasts with accompanying connective tissue fibres displayed in interlaced bands are suggestive of fibrosarcoma. The terminology of xanthogranuloma is justified by polymorphism, the striking feature of the lesion which gives it its granulomatous character. Oberling (1935) suggested its relationship to the reticuloendothelioses.

Xanthogranuloma is a slow but progressively increasing lesion; it may infiltrate the surrounding tissues but none is reported to have undergone malignant transformation or to have metastasized. The presenting signs and symptoms are due entirely to the size of the lesion and vary with the site involved. It has been said that if completely removed, the lesion does not recur. Deep X-ray therapy has been used in its treatment (Oberling, 1935). The pure xanthogranuloma formation can occur at many places in the body. The purpose of this paper is to describe a case of xanthogranuloma formation in the maxillary antrum.

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Clinical Records

Case Report

K.D., 46 years old housewife, attended the out-patient department of Ophthalmology of Irwin Hospital, New Delhi, on *December 9th, 1963*, complaining of intermittent mild pain in right eye for 6 months. During the last month, the pain had increased, becoming worse on bending and associated with gradually and persistently increasing proptosis. Occasional diplopia and bleeding from the right nostril was reported. No history of fever, trauma or diminution of vision was given.



FIG. 1.
Showing slight proptosis of right eye.

Eye Examination: The right eye showed non-pulsatile, non-reduceable forward proptosis (Fig. 1). Slight restriction in movements of the right eye in all directions, particularly upwards, was noticed. Vision and fundus examination were normal. Nothing significantly abnormal seen on examination of the left eye.

E.N.T. Examination: The middle turbinate was seen touching the nasal septum causing obstruction of the right nasal airway. Slight mucopurulent discharge was seen in the right middle meatus. Slight tenderness of the right ethmoid region was noted.

X-ray of the paranasal sinuses showed hazy right antrum but no erosion of the antral walls was seen.

General Systemic Examination: Showed no abnormality.

Blood and Urine: Routine examination within normal limits.

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Diagnosis

Provisional diagnosis of right maxilloethmoidal sinusitis was made, but the presentation of quite marked proptosis with restricted movements of about a month's duration, associated with occasional epistaxis and the age of the patient aroused suspicion of malignancy of right postero-superior medial group of nasal sinuses (Ohngren classification).

Treatment

Right antrum puncture was performed on *December 24th, 1963*, but pumping in the fluid was very painful and there was no returning fluid. On the suspicion of blockage of the natural ostium, another trocar and cannula was passed in right antrum. Wash out was again attempted but again there was no returning fluid.

On *December 28th, 1963*, the right antrum was opened under G.A. through the antero-lateral wall by a sub-labial approach. The antrum was completely filled up with pale rather lobulated soft mass, firm at places. Manipulation caused very slight bleeding. It gave the impression of being a benign growth; the antrum was cleared of the whole mass without much bleeding. A small deficiency was seen in the medial aspect of the right antral roof and rest of the walls were intact and looked healthy. The tissue was sent for biopsy.

The patient had an uneventful recovery marked by regression of proptosis, disappearance of pain and opening up of the right nasal airway. The patient was discharged from the hospital on *January 6th, 1964*, hoping for still further improvement. Tissue biopsy report was xanthogranuloma (Figs. 2 and 3).

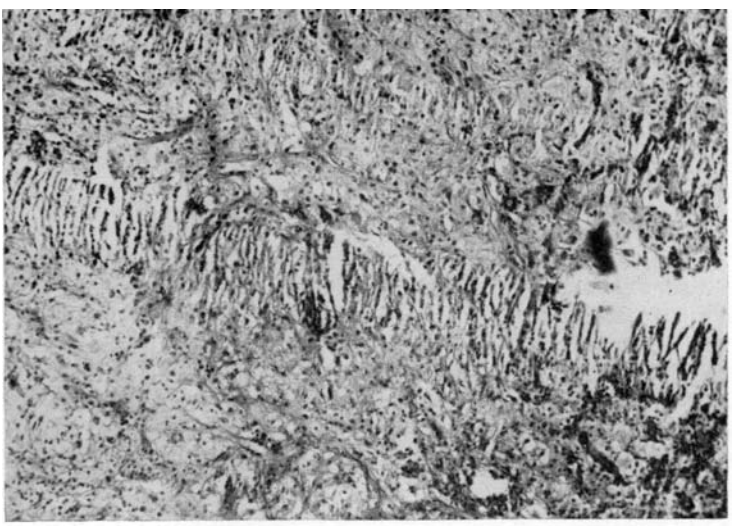


FIG. 2.

Section of piece of tissue removed from maxillary sinus showing foam cells and chronic inflammatory cells. ×100.

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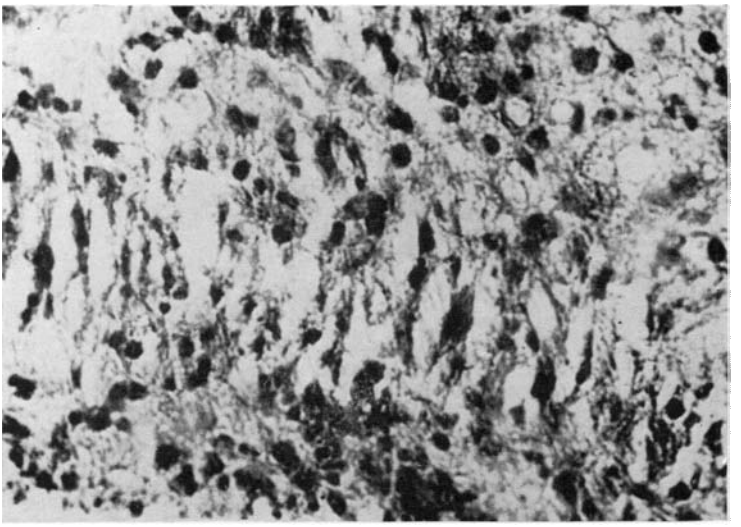


FIG. 3.

Section of piece of tissue removed from right maxillary sinus showing foam cells and chronic inflammatory cells. ×450.

On *March 2nd, 1964*, about two months after discharge the patient reported back with increasing proptosis, pain and partial blockage of right nose. A swelling 2x 1 cm. was palpable in the right orbit at the upper and lateral region. Exploration of right orbit was contemplated but before doing so, acquiring knowledge about the clinical behaviour of the disease after surgery in right antrum was thought essential. The right antrum was again opened through the previous route on *March 7th, 1964*. A similar tumour was found filling the antrum. Biopsy was taken and again xanthogranuloma was diagnosed.

Quick reappearance of the almost completely removed lesion discouraged radical exploration of the orbit. To confirm the extension of the antral lesion in the right orbit, lateral orbitotomy was done on *March 26th, 1964*. A small piece of tissue removed was sent for biopsy and xanthogranuloma was reported (Fig. 4).

The patient refused exenteration of the right antrum and orbit. The lesions were exposed to deep X-ray irradiation but with poor results.

Discussion

Unless favoured by the local influence, how can such a lesion originate at a particular site such as retroperitoneal tissue or the antrum as in the case reported here. We believe in the interaction of a local inflammatory factor and deranged cholesterol metabolism together in the formation of xanthogranuloma. This concept can explain its formation in the antrum. Acute or chronic infection frequently occurs in the maxillary antrum but such rarity of xanthogranuloma in the antrum cannot be well explained. The lesion is basically expansive but destructive invasion does also occur. It explains its extension into the orbit. In

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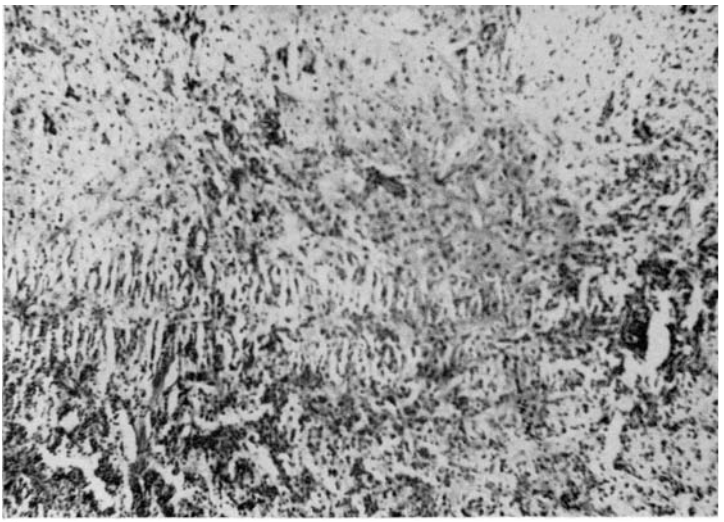


FIG. 4.
Section of piece of tissue removed from right orbit. ×100.

the case reported above, xanthogranuloma was a slow growing lesion and the complete removal was followed by quick recurrence which is in favour of it being of an inflammatory nature.

Summary

A case of xanthogranuloma in the right maxillary antrum invading the right orbit is reported. The ætiology and pathogenesis is reviewed. The lesion appears frankly inflammatory. Partial removal is followed by quick recurrence.

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