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The 'hybrid' fibro-osseous lesion

by

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Introduction

In recent years the term fibro-osseous lesion has increased in popularity as a general designation for certain pathological processes of the cranio-facial skeleton (Boysen *et al.*, 1979).

These lesions characteristically demonstrate replacement of normal bone architecture by a benign fibrous tissue containing varying amounts of mineralized material (Waldron, 1970).

The term fibro-osseous lesion is not specifically diagnostic but comprises an as yet undetermined number of pathological entities (Waldron and Giansanti, 1973a). At one end of the spectrum lies fibrous dysplasia, whilst at the other end is a group of processes arising from multipotential mesenchymal blast cells situated in the periodontal membrane that have the capacity to produce cementum, bone and fibrous tissue (Hamner *et al.*, 1968). Some of these lesions are reactive in nature and others appear to be neoplastic (Waldron and Giansanti, 1973b).

The terminology and classification of fibro-osseous lesions are still confusing and controversial. There is a considerable degree of histological overlapping between fibrous dysplasia and the lesions of periodontal membrane origin (Boysen *et al.*, 1979). Adkeye *et al.* (1980) have recorded the combination of the early active phase of fibrous dysplasia coupled with a lesion similar to aneurysmal bone cyst in the production of an aggressive dysplastic rather than neoplastic 'hybrid' fibro-osseous tumour (Kramer, 1983). We now report a further but as yet to be described fibro-osseous 'hybrid' lesion, comprising the elements of ossifying fibroma, cementifying fibroma and aneurysmal bone cyst combined within the same tumour. This again emphasizes the heterogenicity of this group of pathological processes and stresses the importance of the combined clinical, radiological and histopathological approach to these lesions (Eversole *et al.*, 1972).

Case Report

Clinical presentation

A 14-year-old girl was referred to the E.N.T. Department of the Royal Gwent Hospital, Newport, with a three-month history of a painless left-sided proptosis, intermittent epiphora and ipsilateral nasal obstruction. She had normal visual acuity. No other symptoms were present.

On examination a slight proptosis of the left eye was noted. Anterior rhinoscopy revealed a grossly enlarged left middle turbinate which displaced the septum to the right and caused almost complete obstruction of the left nasal

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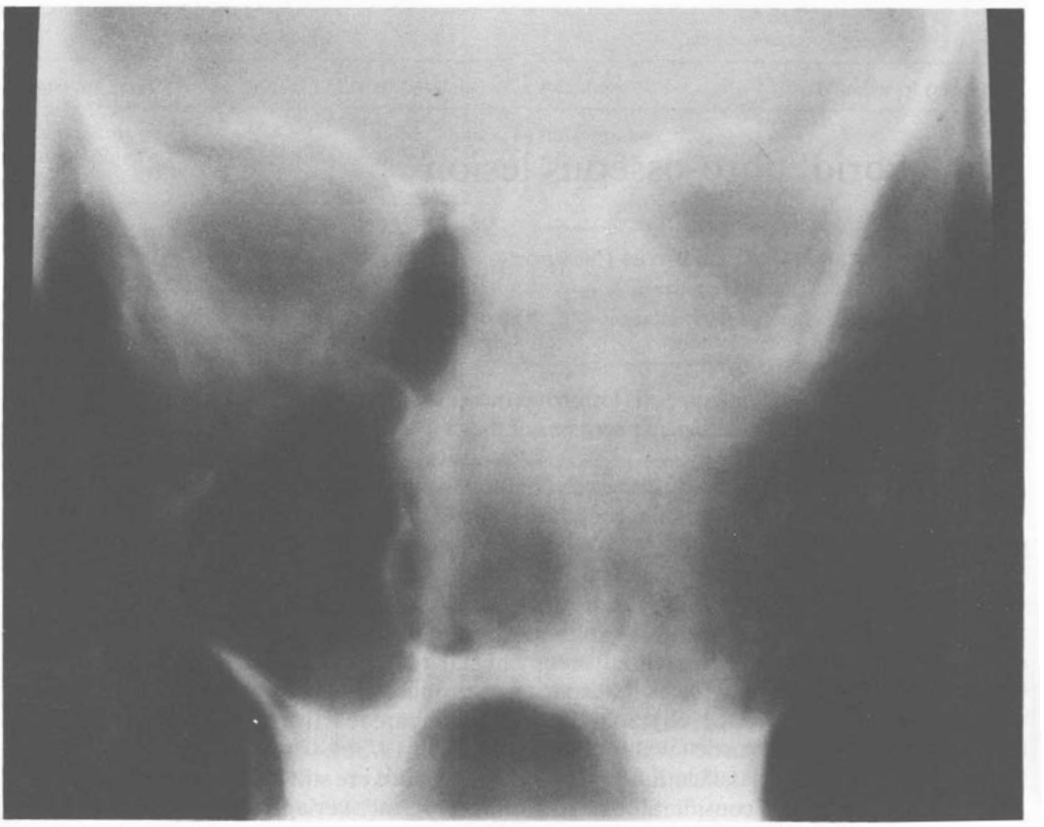


FIG. 1

Postero-Anterior Tomogram. Anterior. A well demarcated expanding lesion involves the infero-medial orbital wall, the left ethmoid air-cells, the left side of the nasal cavity and the left maxillary antrum. The latter are opaque.

airway. The right nasal cavity was normal as were the postnasal space, mouth, hard and soft palate and pharynx. All cranial nerves were intact.

Radiology

(i) Sinus X-rays and tomograms

The left maxillary antrum, left nasal cavity, left fronto-ethmoid complex and sphenoid were all opaque. The left maxillary antrum was enlarged with bowing of its lateral wall and thinning of the bony cortex. There was bone destruction involving the medial wall of the antrum, medial wall of the orbit and orbital floor. These appearances suggested a well demarcated, expansile and destructive process (Fig. 1).

(ii) A C-T scan confirmed the tomographic findings and demonstrated superior extension of the mass through the floor of the orbit with displacement of the globe anterolaterally. No intracranial spread of the lesion was seen. The 100 window width showed apparent sparing of the anterior maxilla (Fig. 2).

Haematological investigation

Routine blood tests, including full blood count and film, urea and electrolytes and liver function tests, were normal. The E.S.R. was 35 mm. 1st Hr. The alkaline phosphatase was

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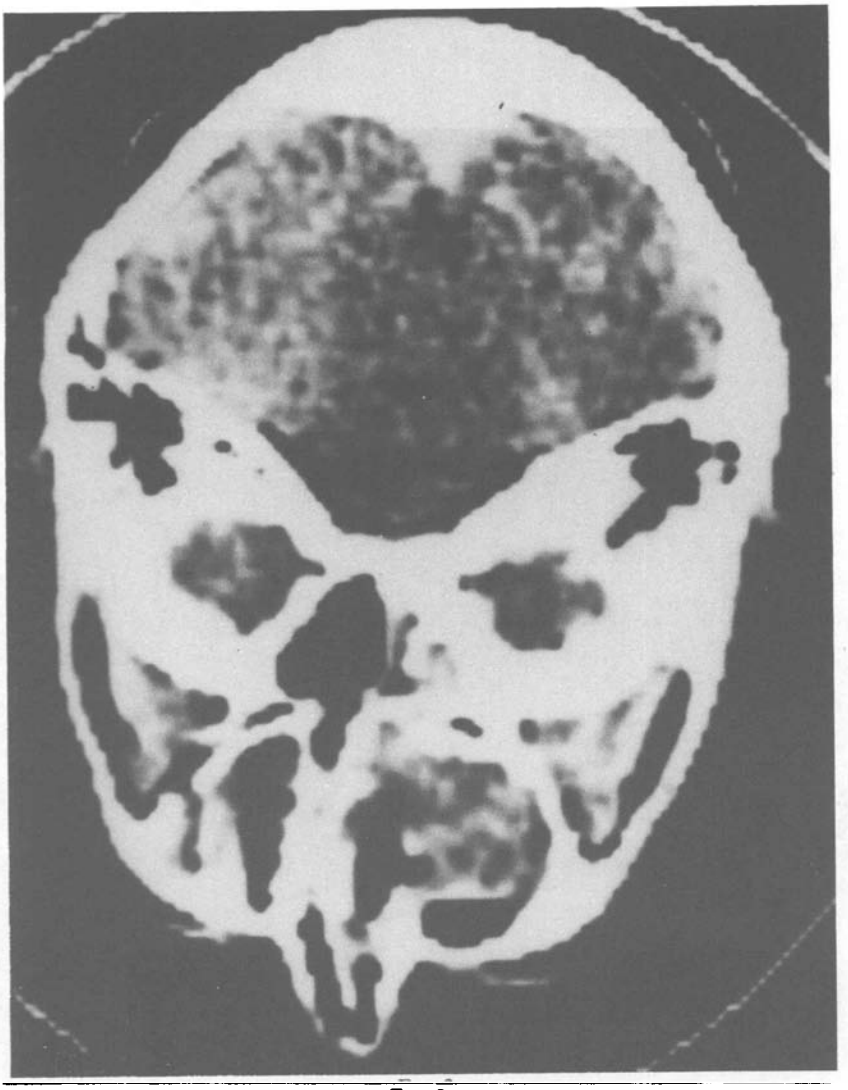


FIG. 2

CT Scan (100 window width). The left maxillary antrum is expanded by a soft tissue mass that has destroyed its medial wall and is invading the ethmoid air cells and the left side of the sphenoid sinus. The nal septum is deviated.

slightly raised at 145 iu/L but within a range considered normal by the laboratory for a girl of her age.

Biopsy

Following admission to hospital the patient was examined under anaesthesia and biopsies were taken from a partly cystic left middle turbinate which on probing discharged brown thin fluid. Deeper biopsies revealed a soft grey-brown tumour mass occupying the maxillary antrum and ethmoid sinuses.

Histopathology

Macroscopically the biopsy material consisted of tissue containing fragments of bone.

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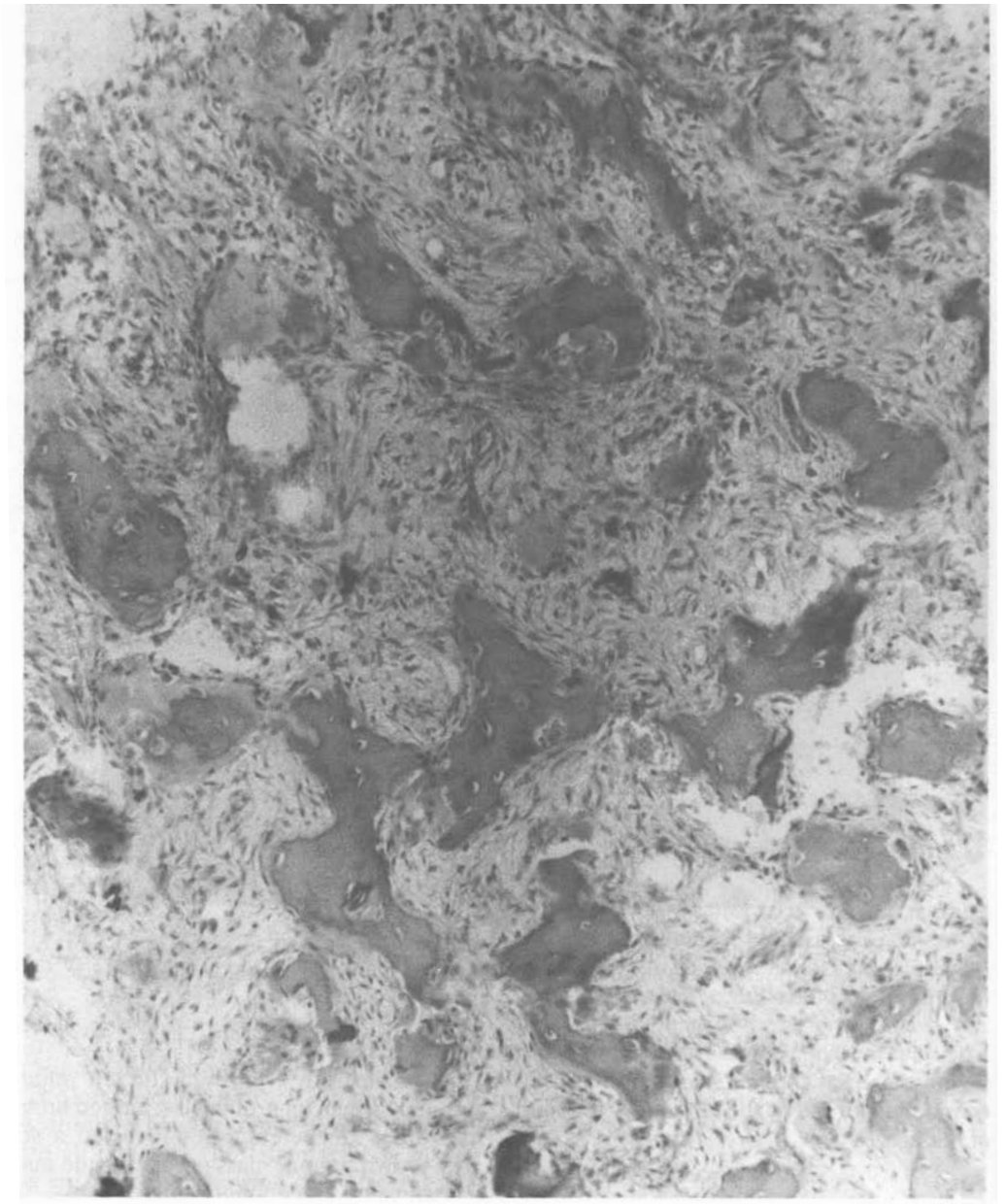


FIG. 3

Fragments of irregular bone in a fibrous stroma. There is very little osteoblastic or osteoclastic activity. HSE ×32.

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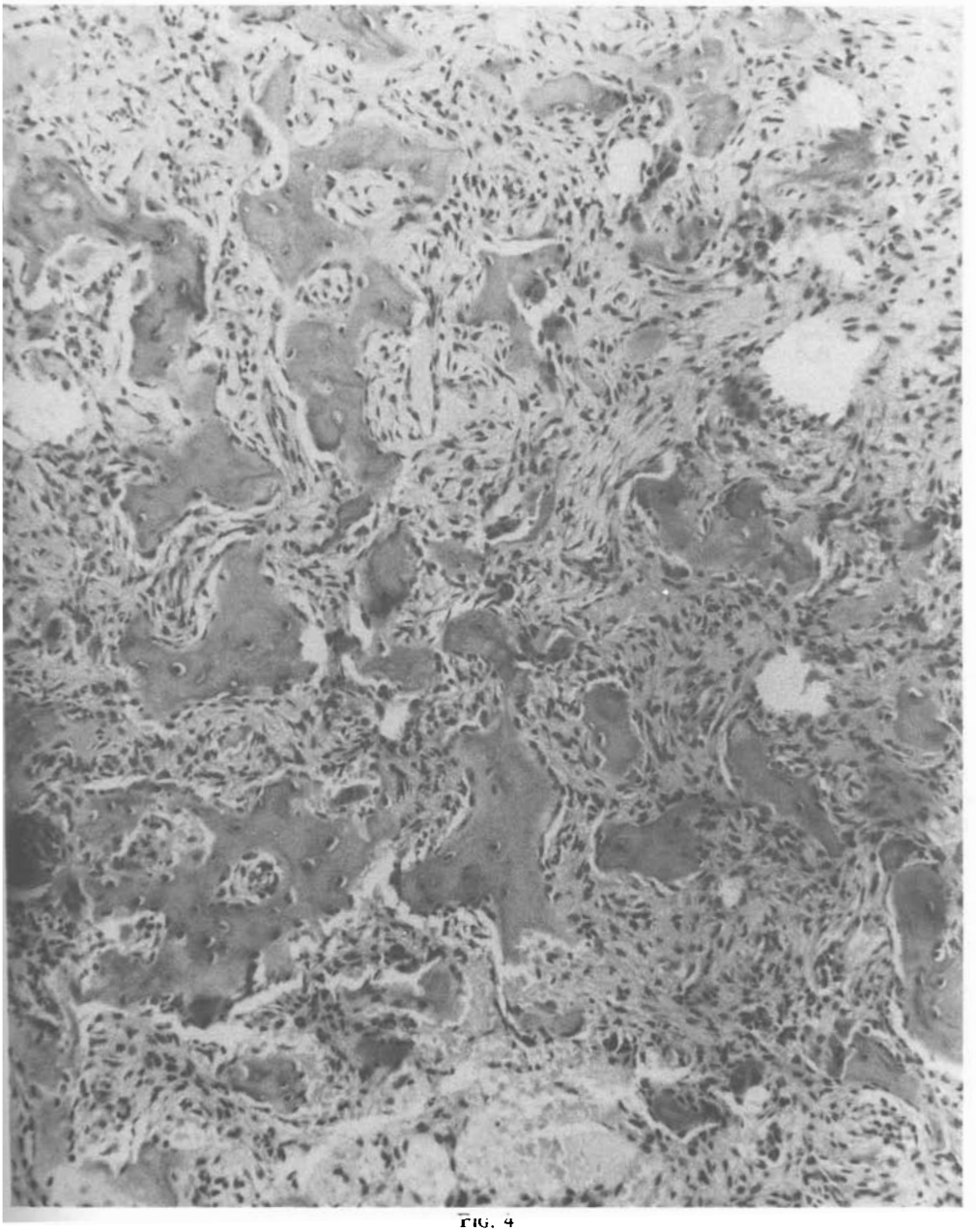


FIG. 4

Irregular bone in a fibrous stroma. Osteoblasts and osteoclasts are present in this area. HSE ×32.

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FIG. 5

Dilated endothelial lined vascular spaces present within the tumour. HSE ×20.

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Microscopically the sections demonstrated a fibro-osseous lesion. It was formed mainly of irregular bony trabeculae in a fibrous stroma (Fig. 3). Some of these trabeculae were rimmed with osteoblasts and occasional osteoclasts were also seen (Fig. 4). The fibrous stroma varied in cellularity from one area to another but without any mitotic activity. Many large dilated endothelial lined vascular spaces were also noted in the stroma (Fig. 5) whilst in addition, lying free within it, were small dense solid 'psammoma-like' bodies or cementicles (Fig. 6).

As features of both aneurysmal bone cyst and others suggestive of ossifying fibroma and cementifying fibroma were present this was considered to be a fibro-osseous lesion of 'hybrid type'. Sections were submitted to Professor I. R. H. Kramer at the Institute of Dental Surgery, The Eastman Dental Hospital, London, who confirmed the histological diagnosis.

Prognostically, it was felt that the lesion was benign but could recur unless completely excised.

Treatment

The tumour was approached *via* a left extended lateral rhinotomy and noted to occupy the maxillary antrum, infero-medial part of the orbit, ethmoids, fronto-ethmoid region and sphenoid. It appeared to have a fairly well-defined edge which glistened grey in places. It was possible to remove the tumour in its entirety. The bony margins were thinned out and the inferior orbital margin was loose but preserved. In places the tumour was attached by fibrous strands to the surrounding structures but its relatively defined margin aided in its identification and excision. The cavity was packed with Whitehead's varnish (which was removed one week later under anaesthesia), and the wound closed in layers. Eighteen months later the incision is almost unnoticeable; there has been no sign of recurrence and the only feature of note is a very slight asymptomatic orbital asymmetry, which has persisted since surgery.

Discussion

The majority of fibro-osseous lesions encountered clinically tend to fall within the broad categories of fibrous dysplasia or fibro-osseous lesions of periodontal membrane origin.

In the differential diagnosis of these lesions one must include: Fibrous dysplasia (monostotic and polyostotic), ossifying fibroma, cementifying fibroma, the cementomas, fibrous osteoma, osteoblastoma and osteoma. Difficulty in diagnosis may be compounded by the presence of infection in the form of periostitis or chronic osteomyelitis (Adekeye *et al.*, 1980).

Histopathology

Fibrous dysplasia can be divided into its two variants, monostotic and polyostotic. Monostotic dysplasia is characterized by involvement of a single bone (Lichtenstein and Jaffe, 1942). Polyostotic dysplasia involves two or more bones and, should the skull and facial bones be the site of involvement, the term cranio-facial fibrous dysplasia is used (Daves and Yardley, 1957). Both variants seem to have a predilection for the maxilla or mandible.

Fibrous dysplasia is characterized by an osseous maturation arrest at a metaplastic woven bone stage (Lichtenstein, 1938; Reed, 1959). It is considered to be a benign self-limiting lesion of adolescents which in growing endosteally tends to follow the general structure of the bone which is then thickened and deformed (Boysen *et al.*, 1979). Cavities such as sinuses will be similarly deformed.

Ossifying fibroma was first reported by Menzel in 1872. Montgomery (1927) is credited with describing it specifically as a clinical entity. There has been much confusion in the past over this lesion and many synonyms were introduced into the literature to describe it, such as: Osteofibroma, fibro-osteoma, monostotic fibrous dysplasia and localized Paget's disease.

Ossifying fibroma belongs to a group of lesions with marginal definition, ease of

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enucleation and which may give the impression of being neoplastic (Adekeye *et al.*, 1980). Kramer (1973) believes that ossifying fibroma behaves like a benign tumour. However, in view of the anatomical complexity of the sinuses, it may recur unless adequately exposed and exenterated.

The periodontal membrane is believed to give rise to the formation of: Ossifying fibroma, cementifying fibroma and cementoossifying fibroma (Tomeo, 1978). Hamner *et al.* (1968) suggested that these lesions could be differentiated from one another based upon the presence or absence of calcification, the staining properties of haemotoxylin and eosin and the degree of fineness of parallel birefringent lines seen with polarized light microscopy.

Waldron and Giansanti (1973b) feel that cemento- and ossifying fibroma are different stages of the same disease process and that attempts at specific diagnosis can be arbitrary and subjective, depending on the interpreter's feelings as to whether or not the amount of cementum or bone present in a lesion is significant. They state that there are few pure product lesions and even those in which cementum or bone predominate usually show some of the other hard tissue.

The differentiation on pure histological grounds between fibrous dysplasia and the ossifying/cementifying fibroma group centres on a number of characteristic features (Waldron and Giansanti, 1973b). These are: (a) The character of the stroma; (b) the nature, arrangement and shape of the bony trabeculae; (c) the presence or absence of osteoblastic rimming, lamellar bone, spheroids or psammoma bodies (calcium hydroxyapatite deposits) (Damjanov *et al.*, 1978).

This classical histological differentiation, however, has become less clearly defined as common histomorphological features are noted to occur, though to a varying degree, in both groups (Boysen *et al.*, 1979; Eversole *et al.*, 1972).

The occurrence of 'hybrid' fibro-osseous lesions, exemplifying the histological heterogenicity of the group, has become increasingly recognized though to date infrequently documented (Kramer, 1983). Adekeye *et al.* (1980), have cited the combination of aneurysmal bone cyst with the early active phase of fibrous dysplasia in producing an aggressive dysplastic rather than frankly neoplastic tumour. Our case represents yet another form of hybrid lesion within the fibro-osseous spectrum, in this instance, demonstrating the different histological features of aneurysmal bone cyst, ossifying fibroma and cementifying fibroma.

Radiology

Many authors distinguish between fibrous dysplasia and ossifying fibroma on radiological grounds, contending that the lesions with delineated boundaries represent ossifying fibromas whereas the diffuse blending lesions are monostotic fibrous dysplasia (Hamner *et al.*, 1968; Sherman and Glauser, 1958; Waldron, 1970).

The radiographic appearance of ossifying fibroma is that of an oval, spherical or multilocular lesion with a well-circumscribed osteolytic boundary. In large maxillary tumours an 'egg-shell capsule' of thin bone is apparent along the outer border of the tumour (Sherman and Sternberg, 1948). The lesion appears to grow expansively filling the sinuses and other cavities with destruction of neighbouring bone. Dental root resorption may occur. Calcification within the tumour mass tends to occur in a haphazard fashion with areas of radiolucency and density intermingled. The anterior maxilla seems to be free of involvement (Waldron and Giansanti, 1973a).

The radiographic appearance of fibrous dysplasia, on the other hand, is extremely variable and depends on the amount of fibrous tissue deposition present, its distribution and the degree of calcification in relation to the duration of the lesion. When areas of condensation are interspersed with areas of radiolucency the classical 'ground-glass' or 'orange-peel' appearance is seen (Daramola *et al.*, 1976). The borders of the lesion are difficult to define precisely as the dysplastic process thickens and distorts the involved bone. The transitional zone between normal and dysplastic bone may be 1 cm. thick in

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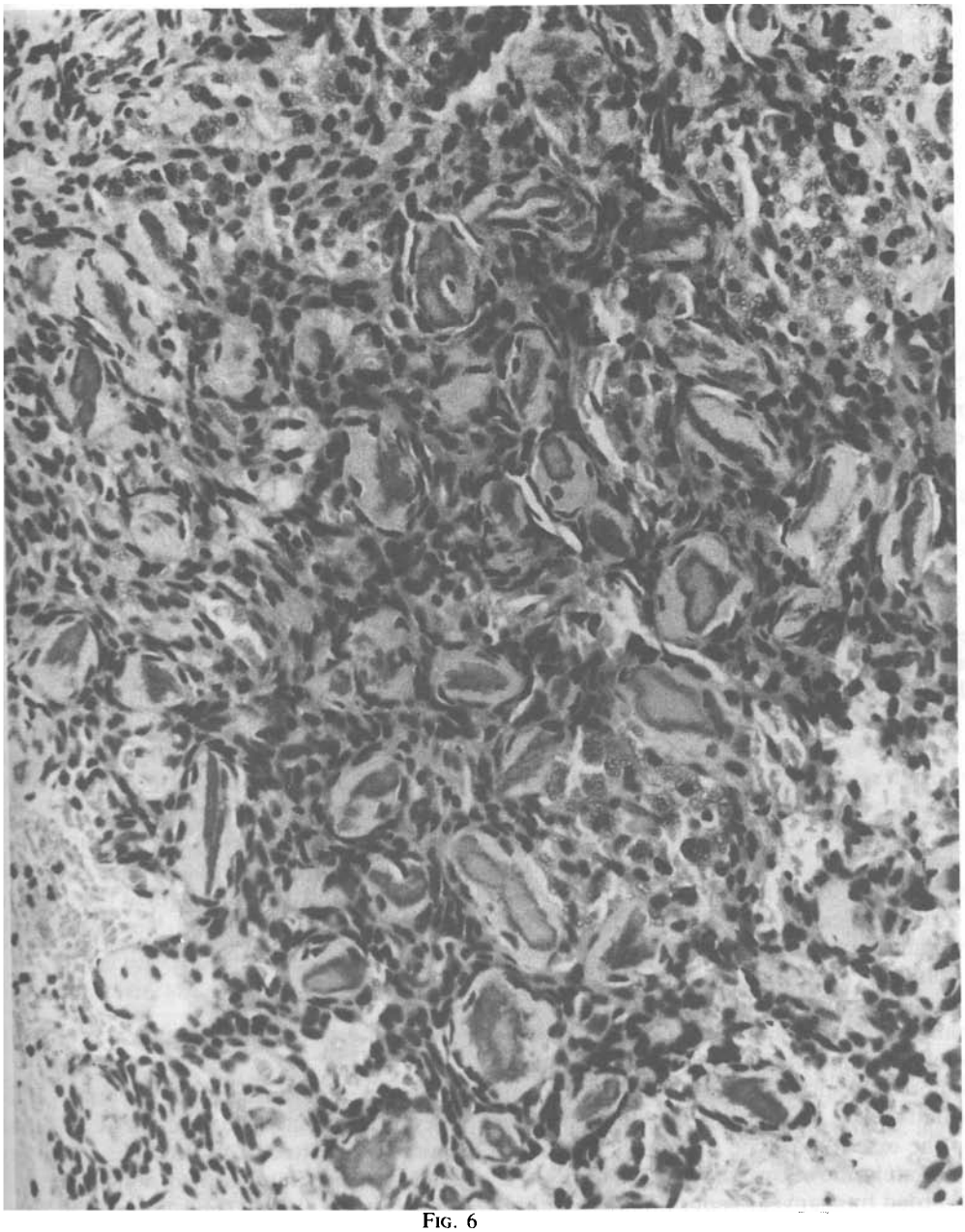


FIG. 6

Fibrous stroma containing small calcified cementicles. HSE ×80.

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places (Waldron and Giansanti, 1973a). As with ossifying fibroma the anterior maxilla seems immune and dental root resorption may occur but less frequently (Walter *et al.*, 1979).

Our case, despite being a 'hybrid' lesion, still fell into the radiographic pattern of ossifying fibroma demonstrating a well-demarcated, expanding and destructive lesion with bowing of the lateral wall of the maxillary sinus, an egg-shell capsule in places and sparing of the anterior maxilla.

Clinical and surgical aspects

It is impossible to differentiate between fibrous dysplasia and a fibro-osseous lesion of periodontal ligament origin by clinical means alone. Both groups can cause facial deformity, proptosis and nasal obstruction and present in the same manner as did our 'hybrid' lesion.

Elevation of serum alkaline phosphatase may occur but, as in our young patient, may not be considered excessive for a child or adolescent. As this enzyme may be raised in polyostotic fibrous dysplasia (Firat and Stutzman, 1968), osteogenic sarcoma, osteitis fibroma cyctica, Paget's disease and a myriad of metastatic neoplasms (Wallack, 1974), its specific diagnostic significance is of little value.

Although we continue to subdivide fibro-osseous lesions of periodontal ligament origin into several categories (e.g. ossifying fibroma, cementifying fibroma, cemento-ossifying fibroma and 'hybrid' lesions) Waldron and Giansanti (1973b) believe that these histopathological terms have little clinical or prognostic significance. A distinction should only be made between the treatment and prognosis of clinically aggressive lesions and those which are asymptomatic (El Deeb *et al.*, 1979). The main task is to try to decide pre-operatively whether or not one is dealing with a monostotic fibrous dysplasia or one of the lesions mentioned above. This will have a direct bearing on the extent and type of surgery indicated. Ultimately, however, the ease of excision is regarded by many surgeons and oral pathologists as the most reliable feature in differentiating between fibro-osseous lesions of periodontal ligament origin and fibrous dysplasia (Tomeo, 1978).

Most fibro-osseous lesions of periodontal membrane origin are small, benign, painless and frequently situated at or near the apex of a tooth. Surgical intervention is not indicated unless the lesion becomes painful or starts to increase in size.

Rarely, however, these lesions can behave in a frankly neoplastic and aggressive fashion (Tomeo, 1978). Unchecked they can cause marked destruction and intracranial spread (Boysen *et al.*, 1979). Our 'hybrid' lesion fell into this category. The treatment is surgical. Adequate surgical access to the tumour is mandatory if total exenteration is to be achieved. Although benign histologically, tumour recurrence may occur if incompletely excised (Kramer, 1983).

In our case, a grey and in places, glistening capsule (Walter *et al.*, 1979) was noted which aided tumour enucleation but excision tended to be rather bloody owing to the aneurysmal cystic vascular component of the lesion.

Radiotherapy is contraindicated in these conditions lest malignant transformation should occur as in the case of fibrous dysplastic lesions where Slow *et al.* (1971) estimated that the incidence of malignancy rose from 0.4 per cent to 44 per cent following therapy.

In conclusion, fibro-osseous lesions may behave in a reactive, dysplastic or neoplastic manner (Eversole *et al.*, 1972). The importance of these lesions to the E.N.T. surgeons lies in the fact that, apart from involving the nasal cavity and sinuses as demonstrated, they may also involve the temporal bone (Levine *et al.*, 1981) and its neighbours, the parietal (Seitz *et al.*, 1980) and occipital bones (Yamashita *et al.*, 1977).

Summary

A hitherto unreported 'hybrid' fibro-osseous lesion involving the nasal cavity and sinuses is reported. This lesion histologically demonstrated features of ossifying fibroma, cementifying fibroma and aneurysmal cyst. Whilst appearing benign in nature it behaved in a neoplastic manner which necessitated

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complete surgical excision. A combined clinical, radiological and histopathological approach is advocated and discussed in relation to the treatment of these lesions.

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