Fibrous dysplasia

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Historical review

Fibrous dysplasia is a term first coined by Lichtenstein and Jaffe (1942) to define a group of bony lesions, previously designated by such varying titles as osteogenic or ossifying fibroma, fibrous osteoma or osteofibroma. In fact, there were thirty-three synonyms listed in their paper in 1942, and the term fibrous dysplasia does not appear in the Index Medicus until 1967.

Fibrous dysplasia constitutes 2·5 per cent of all bony neoplasms, and 7 per cent of all benign bony tumours (Coley, 1960). It is uncertain whether these growths represent developmental abnormalities or are due to abnormal repair following minor trauma, (Schlumberger 1946). Changus (1957) believes that it is due to an exaggerated response of fibroblasts and osteoblasts to some unknown stimulus. Lichtenstein (1942) felt that it was caused by an alteration in activity in bone-forming mesenchyme, and that primitive fibrous tissue proliferates in the medullary cavity of bone and erodes the cortex from within.

For many years fibrous dysplasia of bone was not distinguished from primary hyperparathyroidism, and both kinds of osseous lesions were described pathologically and radiologically as osteitis fibrosa cystica. In fact, Pritchard (1952) reviewed thirty-one cases of fibrous dysplasia which were fruitlessly explored for a parathyroid tumour. However, Hunter and Turnbull (1931) in their paper on hyperparathyroidism drew attention to a localized fibrous lesion of bone in which the rest of the skeleton is normal, and which is never associated with abnormalities of calcium and phosphate metabolism. They called it local osteitis fibrosa.

McCune and Bruch (1937) published an account of a case of osteodystrophia fibrosa associated with precocious puberty and pathological skin pigmentation. The same year Albright and his colleagues (1937) described the same syndrome, which today bears his name, in spite of his objections. It constitutes a polyostotic fibrous dysplasia which is commonly unilateral, associated with brown non-elevated pigmented areas of skin on the same side as the bony lesions and in addition endocrine dysfunction, which in females is associated with precocious puberty. The following year Lichtenstein (1938) described eight cases and suggested the term fibrous dysplasia of bone to identify the disease, and in 1942,

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he and Jaffe reported fifteen further cases and reviewed the literature to include a total of ninety cases. Of these, only thirty-two showed any pigmentation, and twenty, all female, showed some endocrine dysfunction. Serum calcium, phosphate, and alkaline phosphatase was found to be normal in all cases. In fifteen cases, only a single bone was involved, but they pointed out that the monostotic form was probably more common than this. Recent literature suggests that the monostotic form outnumbers cases of Albright's disease by about forty to one. Both the polyostotic and the monostotic lesions are pathologically and radiologically identical.

Fibrous dysplasia and primary hyperparathyroidism

Until recently there had been no evidence that fibrous dysplasia is either a hereditary or a familial disease, nor was there any evidence of any congenital abnormality; however, Firat and Stutzman (1968) reported two cases of familial hyperparathyroidism associated with fibrous dysplasia in a mother and daughter. In these cases removal of the parathyroid adenoma had no effect on the course of the fibrous dysplasia. Familial hyperparathyroidism has been previously reported, but never before in association with fibrous dysplasia, and it had long been believed that there was no connection between fibrous dysplasia and primary hyperparathyroidism. However, Ehrig and Wilson (1972) describe two cases of monostotic fibrous dysplasia with primary hyperparathyroidism. In one there was a raised serum calcium, and the mandibular lesion was operated on following the removal of a chief cell adenoma. The other had a near normal serum calcium but a raised serum alkaline phosphatase. All blood chemistry returned to normal after parathyroid surgery but removal of the adenoma had no effect on the course of the fibrous dysplasia. Benedict (1962) also describes two cases of fibrous dysplasia coexisting with primary hyperparathyroidism. Ehrig feels that 5 to 10 per cent of fibrous dysplasia may be associated with primary hyperparathyroidism but this may be an artificially high figure.

The differential diagnosis of fibrous dysplasia and hyperparathyroidism in the presence of a normal serum calcium presents a problem. Hypercalcinuria may often be found in hyperparathyroidism, and disappeared following excision of the parathyroid tumours in the cases reported, so this estimation may assist in the diagnosis of hyperparathyroidism. Recently, radioimmunoassay of circulating parathormone has become available and this would make the differential diagnosis much easier.

Distribution and presentation

Ramsey *et al.* (1968) describe forty-seven cases of monostotic fibrous dysplasia in cranial bones and state that it is rarely accompanied by extra skeletal manifestations. They use Belaval and Schneider's (1954) classification of fibrous dysplasia:

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Type 1—monostotic, limited to one bone;
Type 2—polyostotic (monomelic);
Type 3—disseminated, with extraskeletal manifestations.

Although their cases are equally distributed by sex, most authors find that females outnumber males by about three to one, particularly in the polyostotic variety. Eighty-three per cent of their cases had symptoms before the age of 20, and they felt that this supported the theory of a developmental defect or hamartoma. In their series the mandible was the most frequently involved bone; however, other authors have found that the maxilla is the most commonly involved facial bone (Zimmerman *et al.*, 1958).

Swelling is the most common presenting symptom associated with functional disturbance depending on the site of the bony lesion. Pain is rare in fibrous dysplasia of the head and neck, although it is common in extracranial lesions, usually because of pathological fractures. There are few reported mandibular fractures. Proptosis, visual disturbance, epistaxis and headache were also noted. No endocrine or sex abnormalities were found associated with monostotic fibrous dysplasia.

X-ray appearance

The diagnosis is made on X-ray appearance associated with the histopathological findings. The X-ray findings in fibrous dysplasis are variable and Fries (1957) has classified three types of X-ray appearance:

(1) Pagetoid with expansion of the involved area;
(2) Sclerotic;
(3) Cystic.

The appearance depends on the relationship between the amount of fibrous and osseous tissue in the lesions, and bears no relationship to the age of the lesion, which is translucent if fibrous tissue predominates, and 'ground glass' in appearance if there is much calcification. Lesions in the mandible are usually relatively radiolucent, and cysts may be seen, while in the maxilla and temporal bone, the lesions are generally more dense. Radio-diagnosis depends on the degree of suspicion. In Schlumberger's (1946) series, the diagnosis was not made in any case out of sixty-seven on the X-ray appearance, and in Ramsey's (1968) series only fifteen out of forty-seven had a correct X-ray diagnosis made. Tomography is often of value in diagnosis and in estimating the true extent of the lesion.

Associated features of Albright's disease

The number of endocrinopathies associated with Albright's syndrome and recorded in the literature in addition to precocious puberty, now include goitre, with or without hyperthyroidism, acromegaly, Cushing's disease, gynecomastia, and acceleration of skeletal growth. The pituitary gland in these cases shows evidence of prolonged stimulation and activity,

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but no evidence has yet been found of any pituitary tumours as such. Hall and Warrick (1972) feel that these disorders suggest an abnormality of the hypothalamus leading to over-production of pituitary hormones. Both Henry (1969), who reported on fifty cases of monostotic fibrous dysplasia (of which twelve were in the calavrium) and Hunter and Turnbull (1931) noted that the lesions may be reactivated by pregnancy.

Malignant change

There has been much debate on the question of malignant change in this condition. It was thought that this occurred more commonly in males and more commonly in the polyostotic form of the lesion, although Schwartz and Alpert (1964) found that equal numbers arose in the monostotic and polyostotic forms of the disease. Thirty cases have appeared in the literature in patients who had a previously histologically diagnosed fibrous dysplasia. Osteosarcoma is most common, although fibrosarcoma and chondrosarcoma have also been described. It has been estimated that 0·4 per cent of the lesions of fibrous dysplasia undergo malignant degeneration, which is about one-quarter of the incidence of malignant change in Paget's disease. Any lesion that suddenly increases in size or becomes painful must be suspected of having undergone such a change.

Of the thirty cases with proven transformation, thirteen had previous radiotherapy. It is usually stated that 3000 r is necessary to produce a sarcoma in bone, although in a bone already abnormal with fibrous dysplasia it is quite possible that a smaller dose may suffice.

Feintuch (1973) recently reported a case of chondrosarcoma of the humerus at the age of 40 in a patient who had irradiation at the age of 9 for fibrous dysplasia. Because of the rarity of this type of malignant change Feintuch postulates that the presence of cartilage suggests the possible relationship between this condition and skeletal enchondromatosis (Ollier's disease), of which 30 per cent are alleged to become sarcomatous.

Metastases may occur in any of these sarcomata, and the occasiona, solitary metastasis in lung may be treated by local excision. Howeverl lymph nodes dissection is never done for this condition. It carries a poor prognosis, but in fact, no poorer than for *de-novo* osteogenic sarcoma. The two-year survival of osteogenic and fibrosarcoma in fibrous dysplasia is 50 per cent, the five-year survival is 20 per cent. The average onset of spontaneous malignant change in fibrous dysplasia is fourteen years from the first symptom. Following radiotherapy the average onset is eleven years.

Treatment

Radiotherapy has not been shown to have any noticeable effects on the growth and progress of any lesion of fibrous dysplasia, and its use in this condition is never indicated. Similarly, chemotherapy has not been shown to have any effect and should not be used.

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The choice of treatment therefore, is limited to either radical or conservative surgery, or even no surgery at all. The range of surgical approaches which has been used is very extensive. In some cases major surgical procedures have been performed, such as the case of Young and Putney (1968) who reported an orbital exenteration and maxillectomy because of exophthalmos. There have also been several cases described of a transcranial approach for fronto-ethmoidal lesions, indicated for encroachment of the tumour on vital structures such as the eye or the base of the skull.

Case histories

Miss P.Y.

An 8-year-old white female presented to an ophthalmologist in March 1969 with a nine month history of a bulging left eye, and a four month history of left nasal obstruction and fullness of the lateral aspect of the nasal bridge. There was no diplopia or loss of visual acuity.

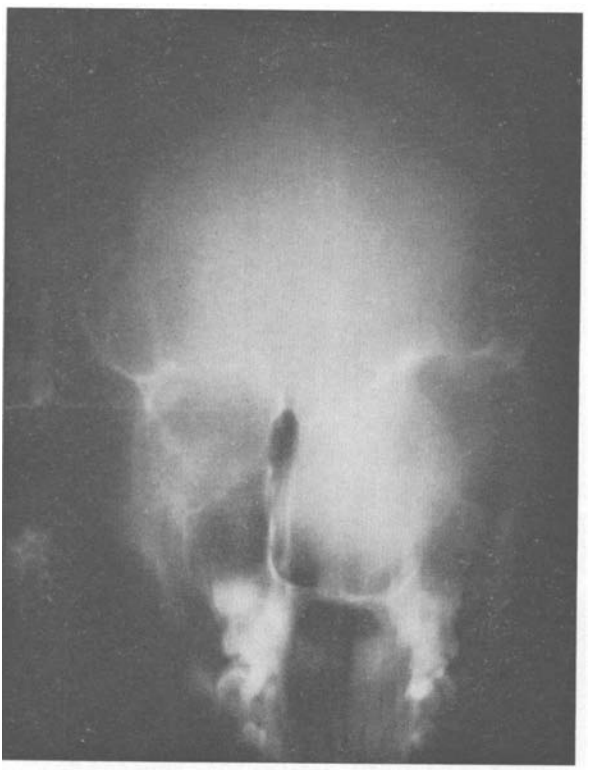


FIG. 1.
Miss P. Y.
Posterior-anterior tomogram of frontal bone and orbits. Large soft tissue mass encroaches on left orbit and left nasal cavity. Left maxillary antrum filled with fibrous mass.

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X-rays showed a large mass in the left nasal cavity with extension into the left orbit, ethmoid and maxilla (Fig. 1). A left carotid angiogram showed no vascularity of the tumour, but splaying out of the maxillary vessels was seen, and a left orbital venogram showed separation of the superior ophthalmic veins.

The case was then referred to one of the authors who examined the nose and found a large fleshy, apparently vascular tumour replacing the middle turbinates, touching the septum and extending downwards to fill the middle meatus of the nose. A punch biopsy showed a cellular fibro-vascular stroma with numerous deposits of bone but no rim of osteoclasts. The specimen had a gritty consistency and a diagnosis of fibrous dysplasia was made (Fig. 2). Serum calcium and phosphate were normal.

In April 1969, November 1969, December 1969, and October 1970, repeated partial excision through external ethmoid or lateral rhinotomy incisions was

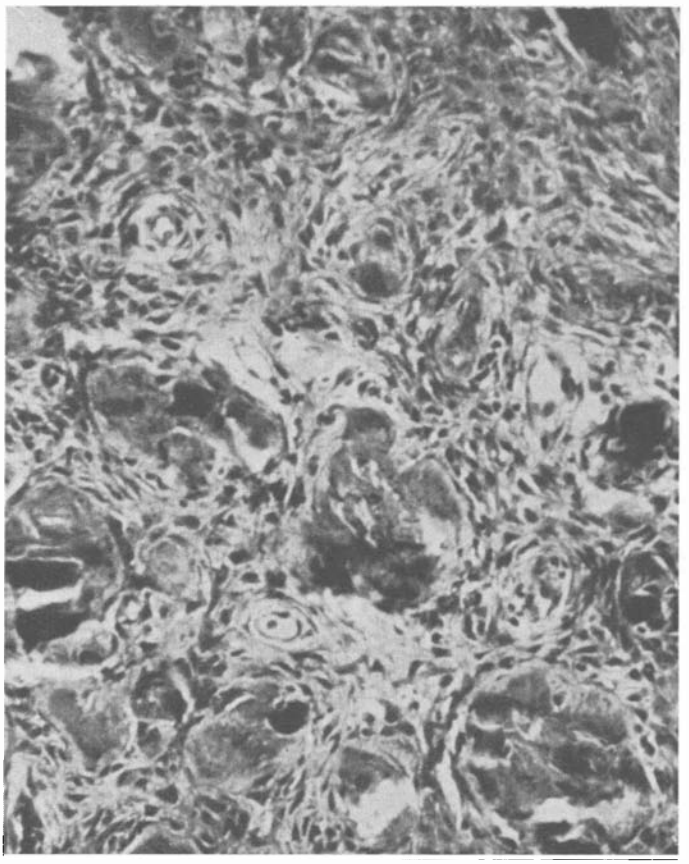


FIG. 2.

Miss P. Y.

Fibrous dysplasia, stained with Trichrome, showing cellular fibrovascular stroma with deposits of bone.

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attempted (Fig. 3). Large amounts of tumour were curetted out at each occasion and a great deal of bleeding always ensued, needing up to three units of blood to be transfused at any one operation.

The lesion continued to progress in size (Fig. 4), with increasing exophthalmos until October 1970, when a decrease in the visual acuity was found. It was associated with fullness of the left optic disc but no atrophy. This time a raised serum alkaline phosphatase of 310 M.I.U./ml. was found. X-rays

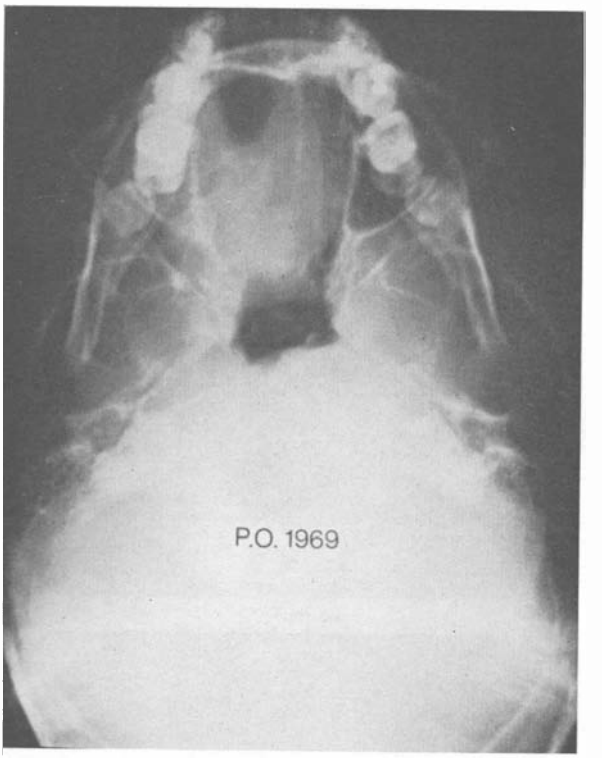


FIG. 3.
Miss P. Y.
Submentovertical view demonstrating operative defect anteriorly in the soft tissue mass.

showed encroachment on the left optic foramen and extension into the posterior ethmoid and sphenoid. A further left lateral rhinotomy was performed and 10 grams of tumour was removed but this made no difference to the symptoms and it was therefore decided to explore the patient via a craniotomy and left frontal bone flap. The periosteum was elevated, orbital decompression was performed and a large volume of tumour weighing 50 grams was removed from the orbit and the nose. The patient withstood the procedure well.

Two months later in April 1971 there was still evidence of disease around the optic canal on X-ray (Fig. 5), the the alkaline phosphatase was still elevated at 340 M.I.U./ml. It was therefore decided to irradiate the lesion because of

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its clinically malignant progress, and 3000 R of Cobalt 60 was given through a 5 by 5 cm. field. Following this the child has been followed up and remains well at the present time. The optic foramen is now clearly visualized for no further progress of the lesion has been seen, although the serum alkaline



FIG. 4.
Miss P. Y.
October 1970.

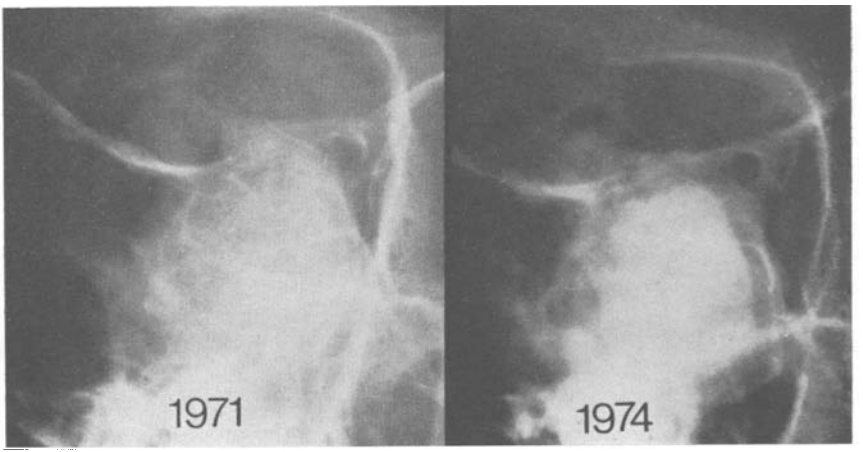


FIG. 5.
Miss P. Y.
Optic canal views. Marked improvement in bony margin of the left optic canal following treatment over a period of three years.

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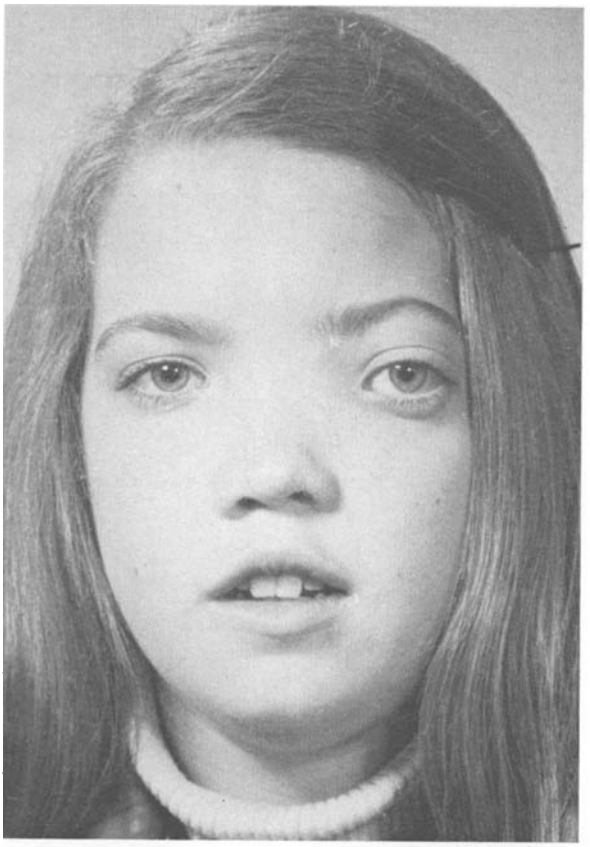


FIG. 6.
Miss P. Y.
Now.

phosphatase is still elevated at 302 M.I.U./ml. There is now no evidence of tumour in the nose (Fig. 6).

Miss C.M.

A 9-year-old white girl presented in October 1960 with a three month history of swelling of the left side of the face and a mass in the roof of the mouth, which on examination was fluctuant. An X-ray showed an opaque expanded left maxillary antrum. A left intranasal antrostomy was performed and pieces of greyish fibrous tumour were removed. A diagnosis of 'fibrous tumour' was made.

In September 1961 she presented at this hospital with the same symptoms, but in addition, had nasal obstruction and discharge. X-rays showed a dense mass in the antrum and nose, extending through the hard palate, floor, lateral wall and roof of the antrum (Figs. 7 and 8). The nasopharynx, medial orbital wall and sphenoid were also encroached upon. Biopsy through the nose showed fibrous dysplasia. A Caldwell-Luc incision was made and tumour was removed

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through the lateral wall of the nose and from the antrum. The bony palate was found to be largely destroyed.

Over the next ten years, the child underwent eight separate surgical procedures for relief of nasal obstruction and a great amount of bleeding always ensued, up to 3 units of blood being transfused on each occasion. During the



FIG. 7.

Miss C. M.
Occipitomental X-ray.
Posterior-anterior tomogram of facial bones.

Marked enlargement of left maxillary antrum with displacement of alveolar margin of maxilla and mal-occlusion of teeth in left side. Mass occupies left nasal passage and displaces nasal septum to right side.

same period she also had numerous epistaxes and on several occasions had to be admitted for this. Serum calcium and phosphate estimations were always normal but a slightly raised alkaline phosphatase of 115 M.I.U./ml. and 125 M.I.U./ml. were found in 1970 and 1971. In May 1972 a Wilson transpalatal incision was performed and a partial removal of the floor of the maxilla and the hard palate was performed. Again a great deal of bleeding was encountered. This was repeated in December 1972, and a much larger opening was made in the nasopharynx to give the patient a reasonable nasopharyngeal airway. Since then the patient has had occasional epistaxes which have required only

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local treatment and there is no evidence that the disease is active at present. The patient is now aged 23.



FIG. 8.
Miss C. M.
Posterior-anterior tomogram of facial bones.
Marked enlargement of left maxillary antrum with displacement of alveolar margin of maxilla and mal-occlusion of teeth in left side. Mass occupies left nasal passage and displaces nasal septum to right side.

Miss D.C.

This 10-year-old girl presented in 1965 with a bony mass occluding the left external auditory meatus. She had no visible tympanic membrane, and a left conductive hearing loss. X-rays and tomography suggested a diagnosis of fibrous dysplasia (Fig. 9). There was a generalized reaction in the bone surrounding the external auditory meatus. Serum calcium and phosphate were normal. Through a post auricular incision, the external auditory meatus was curetted and drilled out down to the annulus and grafted. The tympanic membrane remained intact. No blood was transfused. The biopsy showed a typical appearance of fibrous dysplasia. The child has remained well and asymptomatic since that date.

Each of these cases presents its particular problem and they will be discussed individually.

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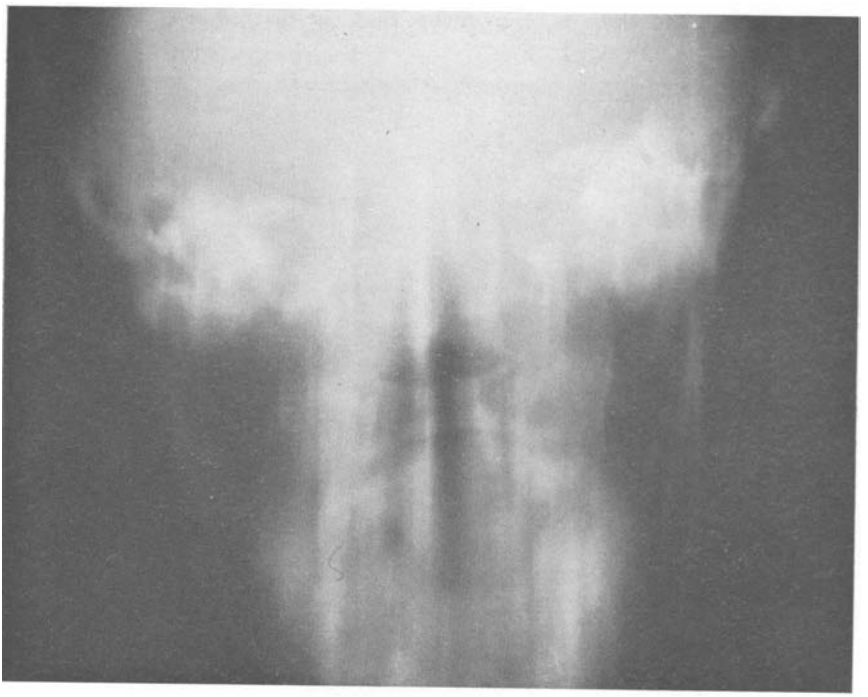


FIG. 9.
Miss D. C.
Posterior-anterior tomogram petrous apices.
Increased bony density of left temporal bone, especially middle and external ear structures. External auditory canal narrowed. Left mastoid process enlarged and sclerotic.

Discussion

Fibrous dysplasia of the temporal bone is thought to be uncommon because only fifteen cases have been described in the literature. In the records of this hospital we have detected an additional case presenting as an acute on chronic mastoiditis in a patient who had been operated on two or three times previously for a lesion of the external auditory canal (Fig. 10). The most common presenting symptoms in these cases are local swelling and deafness due to obliteration of the external auditory meatus. In one case reported in a 14-year-old there was thought to have been previous minor trauma. Cohen and Rosenwasser (1969) present a case which caused a facial palsy and erosion of the whole of the temporal bone extending into the fronto-parietal region and onto the malar bone. Both the sinus and the dura were exposed and the internal carotid artery had to be tied during the course of surgery. It is fortunate that not all cases are like this. The temporo-mandibular joint has also been affected, as well as middle ear erosions of the incus and stapes superstructure. Conservative treatment is usually adequate to relieve symptoms, and in many cases

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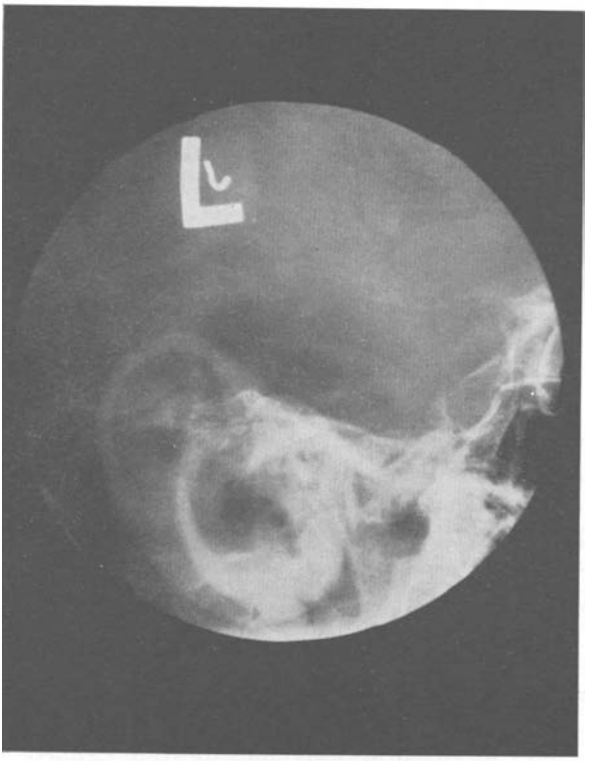


FIG. 10.

Laws view left mastoid.

Mastoid process grossly enlarged and very dense. Large antral radiolucent shadow indicating presence of abscess in the bone.

no treatment has been required once the diagnosis has been made, if the patient was otherwise asymptomatic.

Fibrous dysplasia of the maxilla usually presents as a facial deformity, but may also present to the dentist with loosening of teeth and deformity of the alveolar ridge, or to the Ear, Nose and Throat surgeon with nasal obstruction and recurrent epistaxes with or without nasal infection. The diagnosis in our case, as in most, was made on a combination of X-ray appearance and histopathologic findings following biopsy. That the condition was an aggressive one is evidenced by the recurrent nature of the surgery that had to be performed and also by the raised alkaline phosphatase, which is said by some authors to indicate an actively growing lesion with much osteoclastic resorption. In addition, each surgical intervention caused a great deal of bleeding, adding to the difficulty of adequate surgery. Apart from juvenile angiofibroma it is unlikely that any other benign lesion in the nose could cause such continued and severe bleeding from the operation site.

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Fibrous dysplasia of the ethmoid presents even more of a problem. It has been reported to occur more commonly in younger patients than the average lesion of fibrous dysplasia, in fact, in Ramsey's *et al.* (1970) case the patient was only 3 when first seen. It also tends to take on a more 'malignant' course, growing faster and threatening vital structures such as the optic nerve and the base of the skull.

Patients usually present with facial asymmetry and proptosis. Diplopia is uncommon and loss of visual acuity may occur later. The X-ray appearance is usually that of a dense mass. Because of the rapid progress of the lesion a preliminary diagnosis of a malignant condition is often made. The lesion is usually deeply seated and it may be difficult to get an adequate biopsy.

Our case tends to support the view of Fraser *et al.* (1969) that external ethmoid and lateral rhinotomy incisions afford a very limited access to the lesion, and the considerable bleeding at the time of surgery makes adequate excision difficult. In addition, when loss of visual acuity supervenes, with the lesion threatening the optic canal, a more radical approach to surgery is justified. Fraser describes a bi-frontal flap above the hairline and stripping off the dura from the roof of the orbit. This approach gives good access to the lesion and also a chance to control the haemorrhage by tying off both the anterior and posterior ethmoidal arteries. Through a similar approach in our case, a large volume of tumour was removed weighing about 50 grams, and progress of the lesion was controlled. It is unfortunate that the patient was given in addition a course of radiotherapy since it makes it difficult to assess whether the surgery or the radiotherapy was more significant in controlling the disease. In view of the fact that radiotherapy has never been shown in the literature to have any effect on fibrous dysplasia, it is reasonable to assume that it was the surgery in this case that played the more significant part. If this girl was to present again today, we are quite sure that no radiotherapy would be given. She has been asymptomatic for the last two years, but it is interesting to note that the serum alkaline phosphatase is still markedly elevated in spite of the finding on X-ray that the lesion is more sclerotic.

A strikingly similar case to ours has been described by Chhangani *et al.* (1968) fibrous dysplasia of the ethmoid presenting with proptosis who had had a Moure's lateral rhinotomy followed by progression of the tumour causing diminished visual acuity, necessitating an exposure by reflection of the left frontal lobe of the brain giving relief of symptoms.

The measurement of serum calcium, phosphate and alkaline phosphatase should always be performed in cases of fibrous dysplasia. It has previously been stated on numerous occasions that these estimations are always normal in fibrous dysplasia, while they are usually abnormal in primary hyperparathyroidism. Recent evidence suggests that primary hyperparathyroidism and fibrous dysplasia may occur together and that

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the serum calcium may occasionally be elevated in both the polyostotic form and in the disseminated form of fibrous dysplasia but never in the monostotic, unless there is concomitant primary hyperparathyroidism. The serum phosphate level is always normal. The alkaline phosphatase level is said to be raised in all types of fibrous dysplasia with the presence of an active lesion and it is always raised in the disseminated type. However, in our cases the alkaline phosphatase in two of three patients was elevated in spite of a clinically inactive lesion. It would not seem, therefore, that the level of alkaline phosphatase is any indication of the activity of the lesion. A measurement of urinary calcium and parathormone by radioimmunoassay would seem to be the most satisfactory way of differentiating fibrous dysplasia from primary hyperparathyroidism.

Each of the above cases has something of interest, as mentioned in the discussion. In addition, they demonstrate that recurrences, as quoted in the literature, do not take place as often as persistence of the tumour from inadequate surgical exposure.

Abstracts

Three cases of monostotic fibrous dysplasia of the cranial bones are presented.

Several theories of the etiology of this uncommon condition are put forward and the relationship between monostotic and polyostotic fibrous dysplasia, Albright's disease, and hyperparathyroidism is examined.

The difficulty in diagnosis and the ways they may present to the Ear, Nose and Throat surgeon are discussed.

It is suggested that conservative surgery is the best mode of treatment for maxillary or temporal lesions. However, ethmoid lesions, particularly when the optic foramen or base of skull are eroded, may need a neurosurgical approach. The use of radiotherapy in any case is deprecated.

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