The Journal of Laryngology and Otology
December 1982. Vol. **96.** pp. 1141–1147

Ossifying fibroma

A case report

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Introduction

A GREAT deal has been written about ossifying fibroma and fibrous dysplasia. Many authors divide fibrous dysplasia into two forms, polyostotic and monostotic, regarding ossifying fibroma as part of the monostotic group. Others maintain that ossifying fibroma constitutes a separate clinical and histological entity.

However, confrontation with a patient who has such a gross problem as the one described in this case report makes these distinctions irrelevant.

Clinical presentation

A fourteen-year-old Iraqi girl was referred to Professor D. F. N. Harrison at the Royal National Throat, Nose and Ear Hospital in June 1981. She gave a ten-month history of nasal obstruction from a progressively enlarging mass which completely distorted the nose and was now causing lateral displacement of the right eye with epiphora. The lesion had been biopsied in Baghdad and subsequently treated with an unknown dosage of radiotherapy.

A massive lesion occupied the right side of the nose, expanding it laterally and protruding from the nasal vestibule as a large, ulcerated, crusted mass (Figs. 1 and 2). The skin overlying it was thinned and beginning to ulcerate. There was no intra-oral extension but the mass was visible in the nasopharynx. The right eye was displaced laterally but had full range of movements. There was no evidence of diplopia or palpable cervical lymph nodes.

Radiology (Fig. 3)

Plain sinus X-rays and tomography showed a huge soft-tissue growth which was causing expansion of the nasal cavity and maxillary antra, particularly on the right side. There was almost total destruction of the right ethmoid labyrinth and the tumour had extended upwards, thinning the planum sphenoidale and eroding the side wall of the sphenoid sinus. There appeared to be contact between tumour and dura of the anterior and middle cranial fossae. The changes were consistent with pressure erosion rather than invasion.

Haematological investigation

Routine blood tests revealed a mild hypochromic anaemia with a haemoglobin of 10 g/dl. Calcium and phosphate levels were normal but alkaline phosphatase was slightly raised although within normal limits for a fourteen-year-old (128 iu/l).

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FIG. 2
Pre-operative photograph of patient—lateral view.

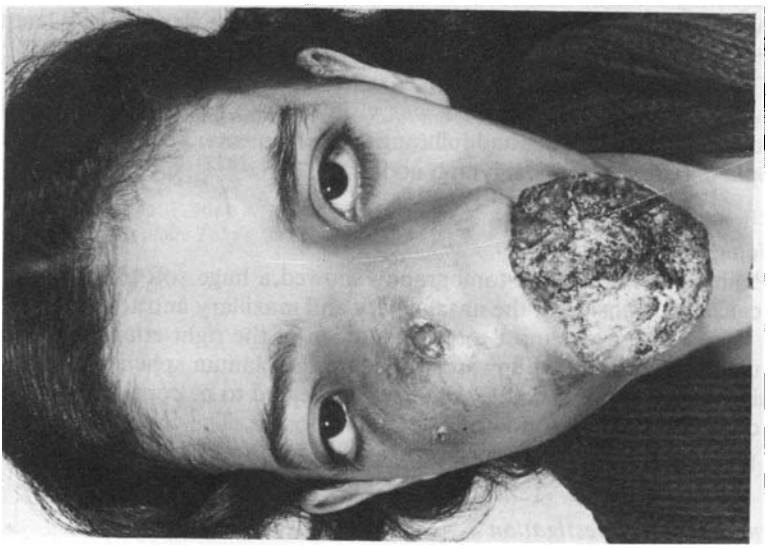


FIG. 1
Pre-operative photograph of patient—frontal view.

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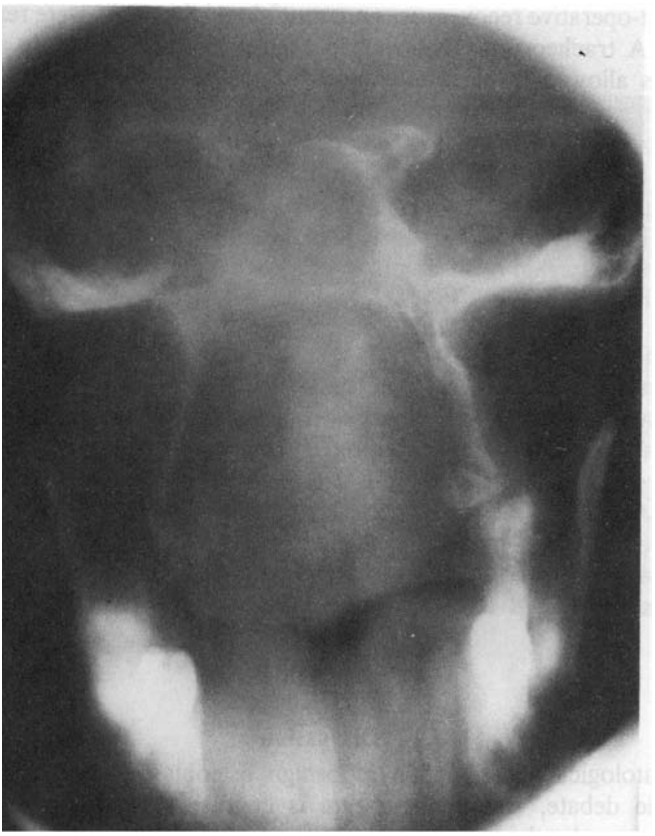


FIG. 3

Tomogram showing massive soft-tissue mass causing huge expansion of nasal cavity and maxillary antra, particularly on the right side, and considerable bony destruction.

Treatment

It was possible to remove the entire tumour *via* a right lateral rhinotomy approach. An enormous mass, 13×7×6 cm., was found attached by fibrous strands to several areas in the nasal cavity, and it protruded into the nasopharynx. It seemed to be attached primarily to the cribriform plate area, being adherent to dura, which was seen to be intact after removal of the mass. The tumour was covered by a large necrotic crust where it protruded from the vestibule, and it was cystic and soft. Posteriorly the mass was firm but encapsulated, and this allowed complete removal.

There had been considerable bony loss due to pressure. The palate was substantially eroded, more on the right, leaving a free-floating alveolus; and there was considerable loss of the maxilla. The sphenoid and ethmoid complex were grossly enlarged and partially eroded. A large part of the septum had disappeared, leaving a single layer of mucosa on the left side.

The incision was closed with removal of redundant skin and refashioning of soft tissues internally to partially reconstruct the septum. The cavity was then packed with Whitehead's varnish packs.

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The post-operative recovery was uneventful and the packs were removed three days later. A tracheostomy, which had been performed to ensure an adequate airway, was allowed to close after twenty-four hours. A prosthesis was introduced at the end of the operation, allowing oral feeding to begin three days after the operation. It is interesting that an alkaline phosphatase done at this time had fallen to within the normal range (107 iu/l).

Although there was lateral displacement of the eye, the overall cosmetic result was good (Figs. 4 and 5). However, it is the intention of Professor Harrison to review the patient again in six months when a second procedure may be necessary to improve her appearance.

Histology (Fig. 6)

The tumour was composed of spindle-shaped cells, some of which were fibroblasts and fibrocytes, and others osteoblasts forming dense groups. Throughout the tissue were calcified particles and fragments of newly woven bone.

In the centre of the lesion there was a considerable amount of necrotic tissue, and the periphery of the lesion was surrounded by a fibrous capsule in which newly formed bone was found.

There were no mitotic figures and no evidence of malignant change. Our pathologists considered this to be an ossifying fibroma, a benign osteoblastic lesion.

Discussion

The histological classification of benign osteoblastic lesions is an area of considerable debate, and the literature is confusing regarding the distinction between fibrous dysplasia and ossifying fibroma. Indeed, many authors contend that there is no distinction.

Ossifying fibroma was first described specifically by Montgomery in 1927, fifteen years before Lichenstein and Jaffe (1942) described fibrous dysplasia. Fibrous dysplasia is generally divided into a polyostotic and a monostotic form, and the histological controversy principally surrounds whether ossifying fibroma constitutes part of the monostotic form or is a separate condition.

Schlumberger (1946) regards both lesions as variants of the same dysplastic process, whereas other authors such as Thoma (1956) prefer to separate the two conditions. Sherman and Sternburgh (1948) and Sherman and Glauser (1958) believe that monostotic fibrous dysplasia and ossifying fibroma may be distinguished radiologically on the basis that the margins of the lesion are diffuse in fibrous dysplasia whereas ossifying fibroma has a clearly demarcated edge.

Differentiating between them on purely histological grounds is more difficult. Pindborg and Kramer (1971) distinguish them primarily on the presence of a capsule but the suggestion that an osteoblastic rim or the presence of lamellar bone precludes a diagnosis of fibrous dysplasia is not supported by authors, such as Waldron and Giansanti (1973). Eversole *et al.* (1972) believe that fibrous dysplasia and ossifying fibroma are separate conditions distinguishable on a clinical and radiological basis.

Another distinction that is made is that ossifying fibroma is said to enlarge after cessation of skeletal growth, whereas fibrous dysplasia usually ceases to progress (Williams and Faccini, 1973).

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FIG. 5
Immediate post-operative photograph of patient—lateral view.

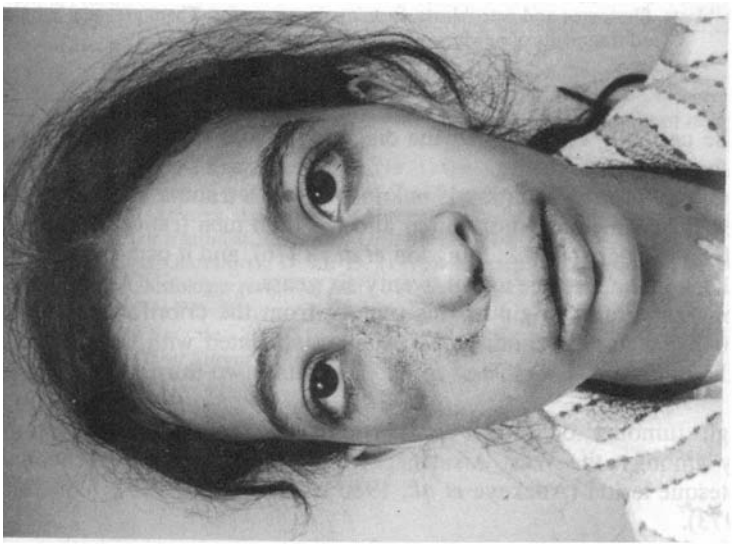


FIG. 4
Immediate post-operative photograph of patient—frontal view.

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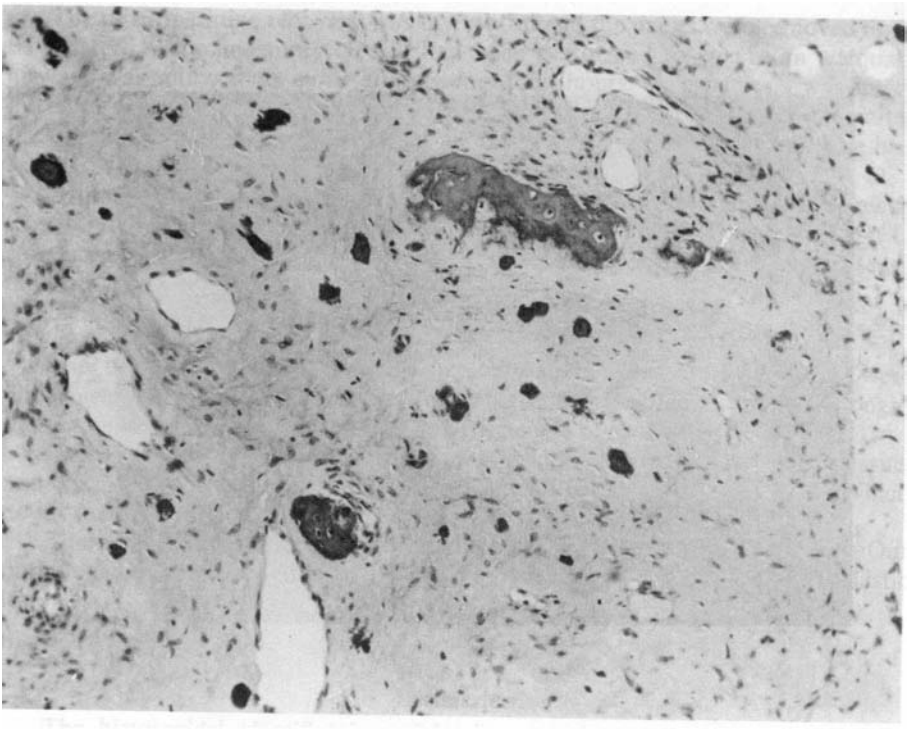


FIG. 6

Photograph of histological appearance showing a tumour composed of spindle-shaped cells, some of which are fibroblasts, fibrocytes and osteoblasts, forming dense groups. There are calcified particles and fragments of newly woven bone throughout the tissue.

Our case certainly conforms to the diagnosis of ossifying fibroma clinically, histologically and radiologically.

The aetiology of this condition is unknown though trauma and infection have been suggested. Women are more often affected than men (ratio 2:1) (Anand *et al.*, 1967; Eversole *et al.*, 1972; Langdon *et al.*, 1976), and it occurs most often in young adults, with an average age of twenty-six years.

The apparent site of origin of this tumour from the cribriform plate area is extremely unusual as this condition is usually associated with the jaw. In their review of the literature, Eversole *et al.* (1972) found two-thirds occurring in the mandible.

Although tumours of similar proportions are reported in the literature, particularly amongst the West African population, we have rarely encountered such a grotesque lesion (Adekeye *et al.*, 1980; Anand *et al.*, 1967; Williams and Faccini, 1973).

All authors are agreed on surgery as the primary treatment of both ossifying fibroma and fibrous dysplasia and that there is no place for radiotherapy. In fact it is positively contra-indicated as it may precipitate malignant change (Slow *et al.*, 1971).

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Conclusion

The classification of benign osteoblastic conditions is confused. Whether ossifying fibroma and fibrous dysplasia are separate conditions or represent part of a spectrum of fibro-osseous disease may be of less importance than the grossly disfiguring effect which these lesions can have.

Fortunately in this case an enormous encapsulated lesion was amenable to complete removal with an acceptable cosmetic result.

Acknowledgements

I would like to thank Professor D. F. N. Harrison for allowing me to report this case and for his help and advice. I would also like to thank Dr. L. Crome for his assistance with the histopathology and Mr. Connelly for the illustrations.

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