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CLINICAL RECORD

Transcatheter arterial embolisation for paediatric inflammatory pseudotumour of the maxillary sinus

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Abstract

Background: Inflammatory pseudotumours are mostly seen in the lung, and occasionally in the head and neck region including the sinonasal area. Reported treatment modalities comprise corticosteroid treatment, surgical excision and radiotherapy. The latter option is required because wide surgical resection may be difficult for head and neck lesions, especially in children. However, clinicians should be aware of the risk of late-onset side effects of radiotherapy in children.

Case report: We present a two-year-old girl with a massive inflammatory pseudotumour of the maxillary sinus. Transcatheter arterial embolisation was performed, and the lesion was successfully managed without additional therapy. There was no evidence of recurrence over the next five years.

Conclusion: This is the first report presenting the utility of arterial embolisation for inflammatory pseudotumour.

Key words: Maxillary Sinus; Inflammatory Pseudotumour; Plasma Cell Granuloma; Inflammatory Myofibroblastic Tumour; Embolization, Therapeutic

Introduction

Inflammatory pseudotumour is an uncommon, benign, non-specific granulation consisting of inflammatory cells, histiocytes and fibroblasts.[1] A very early report described two cases of inflammatory pseudotumour of the lung, and numerous cases in various organs have subsequently been reported.[2,3] However, the pathogenesis and biological characteristics of inflammatory pseudotumour are still under debate. Inflammatory pseudotumour is frequently observed in the lungs and orbits, but also occasionally in the head and neck region, including the sinonasal tract.[1,4–6]

Although a generally accepted therapeutic strategy for inflammatory pseudotumour does exist, it is not well established. Standard options include corticosteroids, surgical excision and radiotherapy.[7–10] Because of anatomical limitations, wide surgical resection of inflammatory pseudotumour arising in the head and neck region is often difficult, especially in children. To the best of our knowledge, no previous study has reported the effect of arterial embolisation of inflammatory pseudotumour in the head and neck region.

In this case report, we present a two-year-old girl with a massive inflammatory pseudotumour of the maxillary sinus, who was successfully managed with arterial embolisation.

Case report

A two-year-old girl had initially presented to the dental clinic with a short history of discomfort over the right maxillary bone. The patient's medical history had been unremarkable.

She had undergone extraction of a right upper fifth deciduous tooth. After the extraction, a gingival mass in the right mandible had appeared. The mass had been biopsied, and histological examination had revealed respiratory epithelium with chronic inflammation and fibrous tissue.

The patient was admitted to our hospital.

Physical examination identified a reddish, granular, mass lesion with aggressive enlargement and frequent haemorrhaging.

Computed tomography scanning of the paranasal region showed a diffuse soft tissue density in the right maxillary sinus, with severe destruction of the maxillary bone. Magnetic resonance imaging (MRI) of the paranasal sinuses demonstrated an abnormal, enhancing soft tissue mass (Figure 1). A plain radiograph of the chest was normal.

Laboratory blood analysis showed an increased white blood cell count (11 300/μl), platelet count (58.7×10^4/μl), serum alkaline phosphatase activity (541 IU/l), serum calcium level (10.5 mg/dl) and C-reactive protein level (0.8 mg/dl). A decreased red

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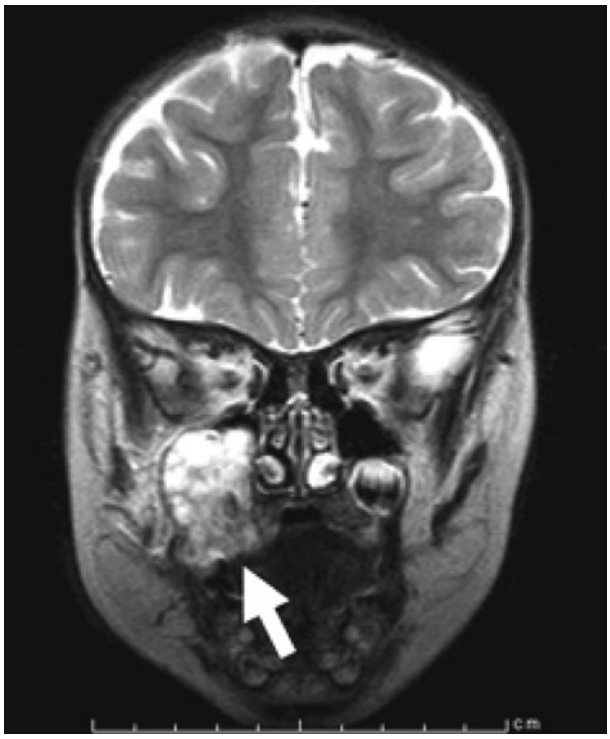

FIG. 1
Coronal magnetic resonance imaging scan showing a massive, non-homogeneous lesion in the right maxillary sinus (arrow).

blood cell count ($3.61 \times 10^6/\mu l$) and haemoglobin concentration (9.9 g/dl) were also observed.

A malignant tumour was highly suspected.

As the patient suffered frequent, substantial bleeding, she underwent embolisation of the major feeding vessels prior to re-biopsy. Angiography under general anaesthesia revealed that a branch of the maxillary artery was the main feeding vessel of the mass, with a branch of the facial artery also supplying a small amount of the circulation (Figure 2a). Embolisation was achieved by injecting polyvinyl alcohol via a catheter into the branch of the maxillary artery; this resulted in almost complete disappearance of the previously visualised angiography enhancement (Figure 2b). After embolisation, another biopsy was taken from the lesion.

Histological examination revealed reactive granulation. The regeneration of stratified squamous epithelium was observed. There was fibrosis and a mild myxoid change in the interstitium. Infiltration of inflammatory cells including neutrophils was observed. The number of vessels was increased, and vascular endothelial cells were enlarged. There was no evidence of malignancy or angiosarcoma. The pathological diagnosis was inflammatory pseudotumour (Figure 3).

After arterial embolisation, bleeding stopped and the mass gradually reduced. The patient was followed up using MRI scans. The mass completely disappeared without any additional treatment, and no recurrence was observed in the five years after embolisation (Figure 4). The patient's right upper permanent teeth

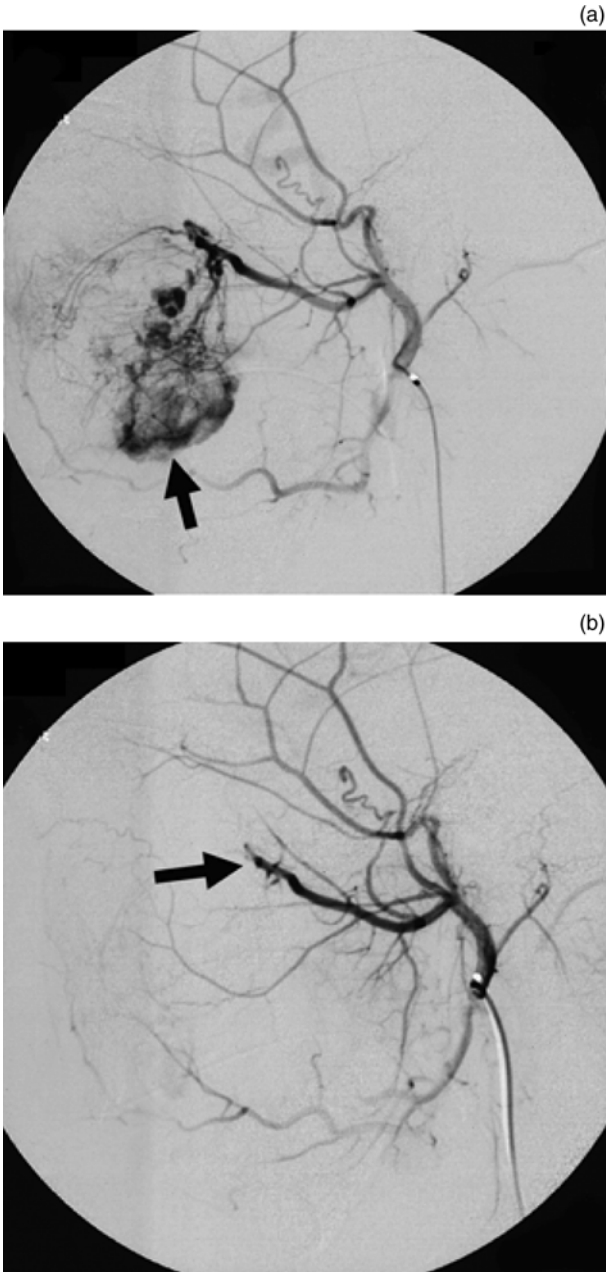

FIG. 2
Lateral view of transcatheter angiography, (a) before and (b) after arterial embolisation. (a) A branch of the maxillary artery is the main feeding vessel of the lesion (arrow). (b) Contrast enhancement disappears after arterial embolisation (arrow).

begin to appear. Facial development was not affected, and the patient had no subsequent clinical symptoms.

Discussion

Inflammatory pseudotumours are composed of a proliferation of myofibroblasts against a background of chronic inflammatory cells, such as plasma cells, lymphocytes and eosinophils. Because of this condition's pathologically heterogeneous composition, a variety of names have been applied, including plasma cell granuloma, pseudosarcomatous myofibroblastic proliferation, xanthomatous pseudotumour, inflammatory myofibroblastic tumour and atypical fibromyxoid nodule.[11]

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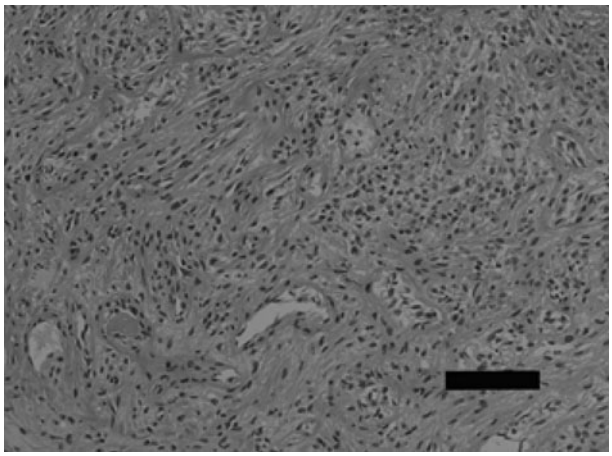

FIG. 3
Photomicrograph showing inflammatory pseudotumour (H&E; bar = 100 μm)

Several classification systems have been reported.[12] For example, inflammatory pseudotumour has been sub-categorised histologically into the following: (1) lymphoid subset (mostly lymphocytes with minimal fibrosis); (2) granulomatous subset (diffuse infiltration of lymphocytes, plasma cells, eosinophils and histiocytes, with a variable degree of fibrosis); and (3) sclerosing subtype (increased fibrosis with limited inflammatory infiltrate).[13] Newlin *et al.* reported that early lesions tended to be predominantly lymphoid, and that advanced lesions contained a greater component of fibrosis.[14]

There have recently been several reported cases of inflammatory pseudotumour of the lung or liver with extensive immunoglobulin (Ig) G4 positive plasma cell infiltration.[15,16] Microscopic examination of the latter case revealed a slight plasma cell infiltration, together with fibrosis and the infiltration of other inflammatory cells, including neutrophils; IgG4-positive cells represented less than 1 per cent of the total cell population.

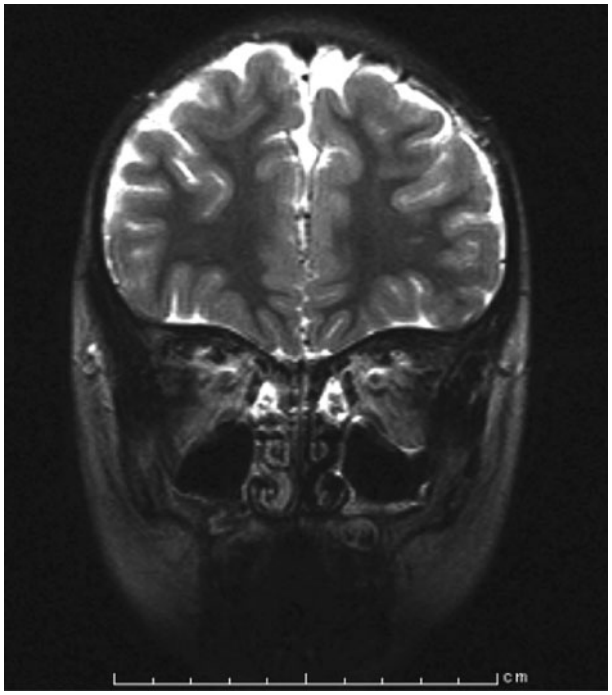

FIG. 4
Follow-up coronal magnetic resonance imaging scan taken after arterial embolisation, showing complete remission of the inflammatory pseudotumour.

Previous studies have reported effective treatment of inflammatory pseudotumour using high-dose corticosteroids, surgical excision and radiotherapy. Surgical resection has generally been proposed, with radiation therapy for unresectable or recurrent cases.[7,9] Maruya *et al.* reported that high-dose corticosteroid treatment should be carefully applied prior to consideration of resection or radiotherapy.[4] A literature review indicates that radiotherapy has been proposed for lymphoid-predominant lesions, while granulomatous lesions may respond better to high-dose corticosteroids. Sclerosing lesions with a large fibrotic component exhibit more aggressive features, and show little response to high-dose corticosteroids or radiotherapy.[9,14,17–19]

- **The pathogenesis and characteristics of inflammatory pseudotumour are controversial**
- **Surgical excision, high-dose corticosteroids and radiation therapy have been used to treat this condition**
- **A case of inflammatory pseudotumour in the maxillary antrum is presented, in which arterial embolisation was successful, suggesting an additional therapeutic option for this condition**

Ahuja *et al.* reported successful combination therapy of a proximal haemophilic pseudotumour arising in a child with mild haemophilia, using arterial embolisation prior to surgery. However, no previous report has presented the utility of arterial embolisation as the sole treatment for inflammatory pseudotumour.[20]

The precise effect of embolisation on tumour cells remains largely unknown.[21] The ischaemia and hypoxia generated by arterial embolisation are possible mechanisms for the observed reduction in inflammatory pseudotumour mass.

Conclusion

Wide surgical resection of inflammatory pseudotumour is difficult for head and neck lesions, especially in children. Furthermore, radiotherapy may have late-onset side effects in children. Hence, we propose that arterial embolisation may be a therapeutic option for inflammatory pseudotumour presenting in children.

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