Case Report / Olgu Sunumu

Retroauricular inflammatory myofibroblastic tumor: a case report

Retroauriküler enflamatuvar miyofibroblastik tümör: Olgu sunumu

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Inflammatory myofibroblastic tumors are rare lesions which show benign histological features with highly local aggressive growth. These tumors usually tend to involve lungs. Head and neck location is very rare. In this article, a four-year-old patient who underwent a procedure for a retroauricular mass which evolved into inflammatory myofibroblastic tumor was presented in the light of data from the literature review.

Key Words: Inflammatory myofibroblastic tumor; pediatric age group; retroauricular area.

Enflamatuvar miyofibroblastik tümörler, benign histolojik özellikler ve genellikle lokal agresif gelişim gösteren lezyonlardır. Bu tümörler, çoğunlukla akciğerleri tutar. Baş ve boyunda yerleşimine çok ender rastlanır. Bu makalede, literatür incelemesinden elde edilen veriler ışığında, enflamatuvar miyofibroblastik tümöre dönüşen retroauriküler bir kitlenin cerrahi ile alındığı dört yaşındaki bir olgu sunulmuştur.

Anahtar Sözcükler: Enflamatuvar miyofibroblastik tümör; çocukluk yaş grubu; retroauriküler bölge.

Inflammatory myofibroblastic tumors are also known as inflammatory pseudotumors, plasma cell granulomas, pseudo pseudosarcomatous myofibroblastic proliferation, myofibroblastomas, xanthomatous pseudotumors and atypical fibromyxoid nodules.[1] These tumors, although showing benign histologic characteristics, are clinically locally aggressive.[2,3] They are most commonly encountered in the lungs. The upper respiratory tract is less commonly involved, with the larynx, trachea, oropharynx and nasopharynx accounting for 11% of extrapulmonary cases.[4] In the head and neck area, they can be diagnosed mostly in the orbita, rarely in the maxillary sinus, nasopharynx, infratemporal fossa, pterygopalatine fossa, larynx, pharynx and seldomly at the skull base.[5,6]

CASE REPORT

A four-year-old male patient was referred to our clinic for a painless progressively enlarging mass behind his left ear for four months. Physical examination revealed a 3x3 cm, mobile mass in the left retroauricular area (Figure 1). On microscopic examination, the external ear canal and tympanic membrane were normal. Fine needle aspiration biopsy was repeated twice, but did not yield sufficient diagnostic cellular material. On magnetic resonance imaging (MRI), a 3x4 cm solid mass with slightly irregular borders was demonstrated in the

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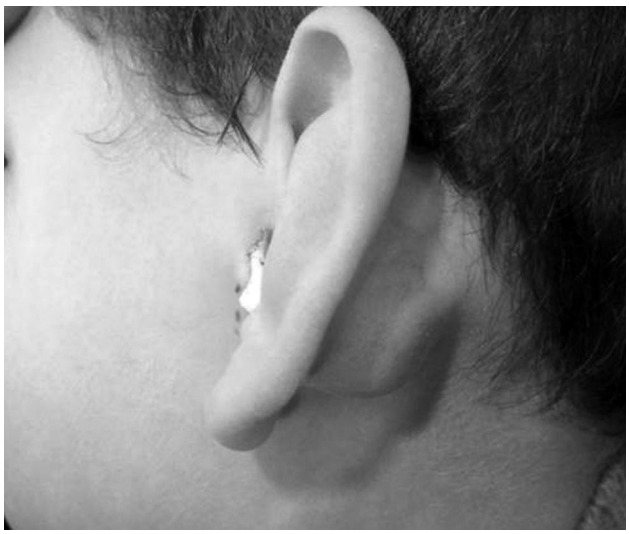

Figure 1. *The retroauricular mass.*

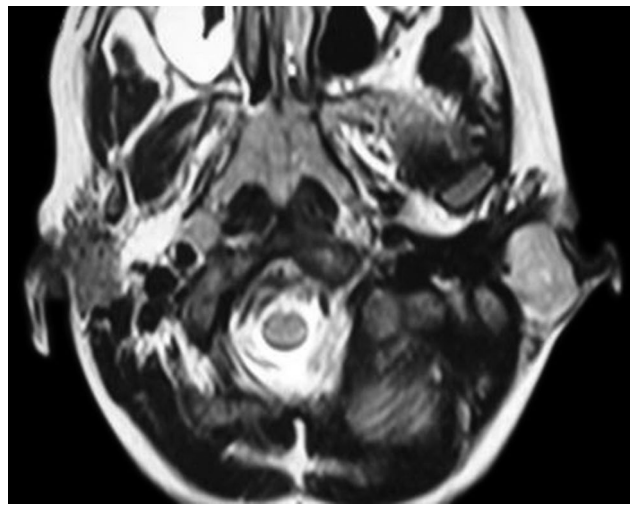

Figure 2. *A 3x4 cm solid mass lesion with slightly irregular borders located at the left retroauricular area.*

left retroauricular area (Figure 2). Within one month during which the patient was followed, the mass invaded the conchal cartilage and its overlying skin, hemorrhage from the external ear occurred, and invasion of the retroauricular area skin was observed. Due to the aggressive clinical behavior of the mass, suspicions of a possible malignant tumor were raised and surgery to remove the tumor was planned. During surgery, it was observed that the mass destroyed conchal cartilage and exited from the external ear by keeping the auricular skin. The mass was 4x4 cm and adherent to surrounding tissues (Figure 3). The tumor was completely excised along with part of conchal cartilage and external ear canal skin. No destruction in the temporal bone was observed. The surgical defect was closed primarily. The surgical margins were all negative on frozen section. According to the final pathology result, the mass was diagnosed as inflammatory myofibroblastic tumor. There were no problems in the postoperative period and the patient was evaluated by pediatric oncology, and it was decided to follow the patient for possible multi-centric tumors.

DISCUSSION

Inflammatory myofibroblastic tumors are rare and originate from nonspecific granulation tissue.

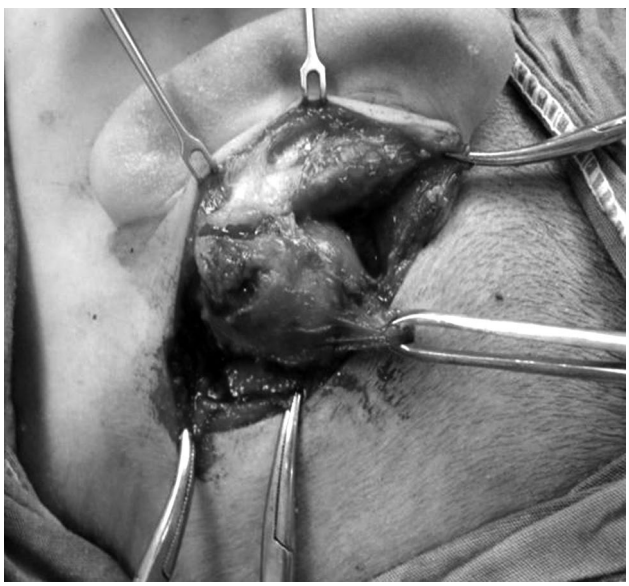

Figure 3. *Conchal cartilage invasion by the tumor.*

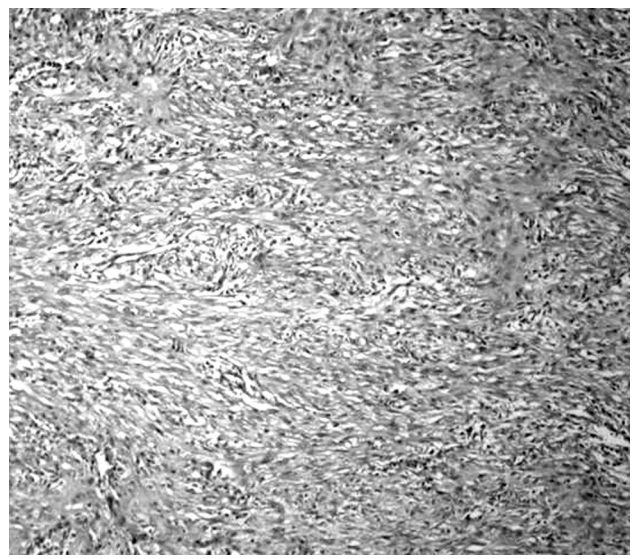

Figure 4. *Dense chronic inflammatory cells are followed up between the myofibroblastic cells (H-E x 100).*

They are located mostly in the orbita at the head and neck area. Our literature review revealed only three cases located within the middle ear and mastoid area. External ear and retroauricular locations have not been reported. Their etiology and pathophysiology are not clear but infection and chronic inflammation are posited. There was no history of chronic trauma or infection in our case.

Inflammatory myofibroblastic tumors located in the lungs mostly occur in the middle-age group while extrapulmonary tumors are more common in the first and second decades.[1] Our patient is in the infant age group as compatible with the literature.

Since it is a soft tissue tumor, the preferred imaging method is MRI. In our case, preoperative MRI examination helped significantly in planning the surgery. The major differential diagnosis should be made with head-neck malignant tumors; especially sarcomas which are common in the pediatric age group. Definitive diagnosis is made with histopathological examination. In the presented case, fine needle aspiration biopsy was repeated twice; but it did not yield sufficient diagnostic cellular material.

On postoperative histological examination, myofibroblasts and plasma cells were seen among chronic inflammatory cells (Figure 4). It has been observed that myofibroblastic cell density correlated with recurrence and local aggressiveness.[1] Clinical behavior of the tumor is variable. In some cases, spontaneous regression may occur, but sometimes the tumor may persist or even enlarge and invade surrounding areas.[7] In our case, we observed that the tumor enlarged rapidly and destroyed the conchal cartilage within one month.

The choice of treatment is guided by the location of the lesion. The treatment options for orbital inflammatory pseudotumors include systemic steroid therapy, radiotherapy, complete surgical resection, alone or in combination.[1] Systemic steroid therapy has been initially successful in controlling up to 78% of orbital lesions.[4] Radiotherapy is useful for recurrent cases and in cases when steroid therapy has proved to be ineffective.[1,4,7] Due to the rarity of temporal bone-located inflammatory myofibroblastic tumors, there is no established treatment modality. However, complete surgical resection should be seen as the main treatment option. In our case, complete surgical resection was performed. Recurrence rates after surgical resection have been reported between 10% and 25% for extrapulmonary inflammatory myofibroblastic tumors.[1] During the two year follow-up period of our case, there was no recurrence.

Consequently, inflammatory myofibroblastic tumor of head and neck region is a rare clinical entity that should be kept in mind in the differential diagnosis of pediatric head and neck tumors.

Declaration of conflicting interests

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