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Case Report
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Iodine-125 brachytherapy for the treatment of a large parotid epithelial-myoepithelial carcinoma in a child

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Abstract. Epithelial-myoepithelial carcinoma (EMC) in the salivary glands is rare. Tumours originating from salivary glands are also rare in children. Radical resection is the main treatment used for EMC in salivary glands. However, this surgery is commonly associated with functional and/or cosmetic deficits. There is also a high percentage of local recurrence after a tumourectomy. We present a typical case of recurrent EMC in the left parotid gland of an 8-year-old girl. The patient was treated with iodine-125 brachytherapy. At the 2-month follow-up, the tumour size was reduced by 80%, and at 1 year, no tumour tissue was detected on positron emission tomography/computed tomography. At the 6-year follow-up, no local recurrence or distant metastasis was found and no complications had occurred. Iodine-125 brachytherapy is a potentially appropriate alternative treatment for EMC in the salivary glands, especially for paediatric patients.

Key words: iodine-125; brachytherapy; epithelial-myoepithelial carcinoma; parotid..

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Epithelial-myoepithelial carcinoma (EMC) in the salivary gland is rare, with an incidence of less than 1% of all salivary gland tumours. EMC is now recognized as a low-grade malignant tumour.[1] Radical excision is the main treatment for EMC. However, local recurrence is common after resection. Also the extent of the surgical excision often results in significant cosmetic and/or functional deficits.

We report the case of a female paediatric patient with a large EMC, who was treated successfully using iodine-125 brachytherapy.

Case report

Clinical history

An 8-year-old girl was referred to our hospital because of a recurrent tumour on the left parotid gland. Two years previously she had noted a painless tumour in the sub-auricular region on the left side. The tumour was 20 mm × 20 mm in size. She was diagnosed with a parotid gland tumour in a local hospital. The tumour and superficial lobe of the parotid gland were removed, with the left facial nerve preserved. Pathological examination of the tumour indicated EMC. She was not given any treatment after surgery. One year after surgery, the tumour recurred in the same region and grew slowly. She had no other symptoms except for the tumour, and the results of all laboratory tests (clinical

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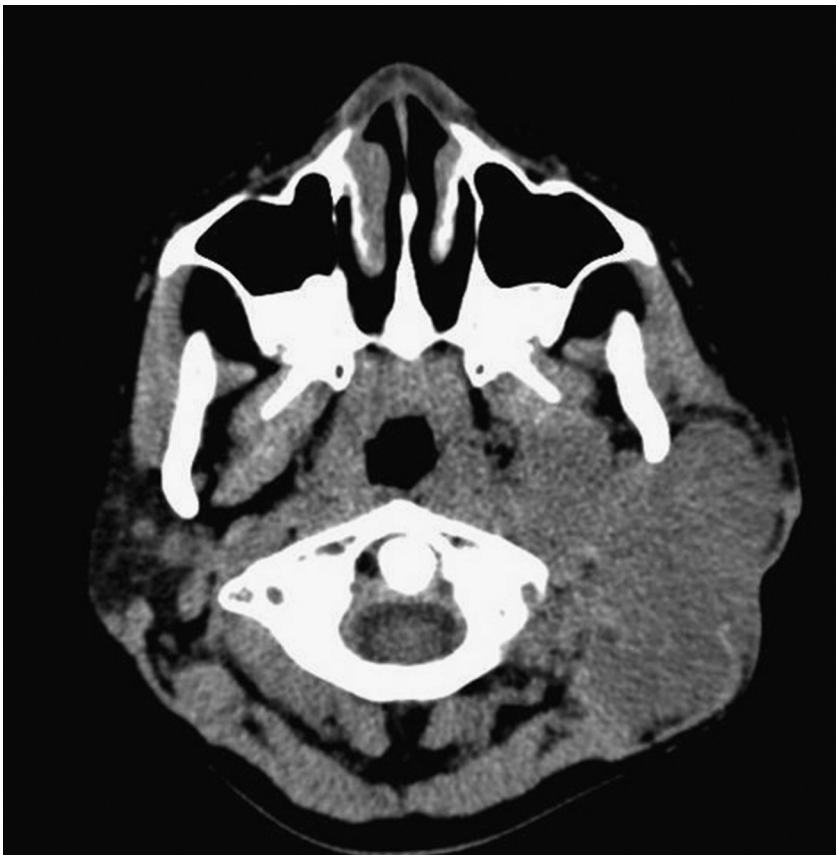

Fig. 1. CT scan showing a large tumour in the parotid region; the tumour margin is not clear.

chemistry and haematology) were within the normal ranges. She was then referred to our hospital.

On physical examination, there was a hard, immobile, painless, and irregular mass in the left sub-auricular region; the mass margin was not clear. Her facial movement was intact and mouth-opening movements were not limited. The superficial skin over the mass was normal. The parapharyngeal wall had not expanded. No enlarged lymph nodes were found in the neck.

A computed tomography (CT) scan revealed an enhanced tumour with cystic lesion; its margin was not clear and the tumour was approximately 70 mm × 70 mm × 60 mm in size, located in the left parotid gland region. The upper bound of the tumour was located inferior to the zygomatic bone and the lower bound was at the lingual bone level. The jugular artery and vein were not clear (Fig. 1). Chest radiography showed no metastasis.

Treatment

On 20 July 2007, a radical resection was planned and performed through a standard 'lazy S' cervicomastoid–pre-auricular incision. After the parotid gland and tumour were exposed, the facial nerve was found to be surrounded by the tumour and thus could not be preserved. The child's parents refused to allow sacrifice of the facial nerve. Instead, a conserved tumour resection was performed. Pathological examination confirmed the tumour to be an EMC (Fig. 2).

The patient's parents agreed to the use of iodine-125 seed implantation brachytherapy. Based on the CT scan, the tumour outline was defined as the clinical target volume (CTV) and 1 cm outside of the CTV was defined as the planned target volume (PTV). The prescribed dose was 120 Gy. The radioactivity of each seed was 0.8 mCi and it was planned to implant a total of 97 seeds. On 2 August 2007, 97 iodine-125 seeds were implanted in the target positions under CT guidance.

Follow-up

Two months after the iodine-125 seed implantation, the mass had decreased significantly in size. The functions of the facial nerve were normal and the girl had experienced no discomfort. A CT scan showed the tumour size to be reduced by about 80% (Fig. 3).

One year later, no cancerous tissue could be detected on positron emission tomography/computed tomography (PET/CT). The skin had become rough and black. Six years later, a local physical examination was completely normal and the superficial skin had become normal. No local recurrence or distant metastasis was found (Figs 4 and 5).

Discussion

Salivary gland tumours are rare in a child, and only 2.5% of all head and neck malignancy in childhood is of salivary gland origin.[2] According to the World Health Organization pathological classification, EMC is a rare neoplasm of the salivary gland with an incidence of less than 1% of all salivary gland tumours. EMC is a low-grade malignant neoplasm with a high propensity for local recurrence after resection. Many authors have reported radical surgical resection to be the only and best treatment for EMC,[3] but incomplete surgical excision is associated with a high percentage of local recurrence and distant metastasis. Previous studies have reported about 40% of local recurrence and 14% of distant metastasis after a tumourectomy.[4] Postoperative adjuvant radiotherapy and/or chemotherapy have been used by some medical doctors, but their benefits remain unclear. Thus, long-term follow-up is necessary postsurgery for EMC patients.

Surgical excision is the mainstay of treatment for salivary gland tumours. However, quite a number of patients are not candidates for surgery due to the location or size of the tumour making the tumour unresectable, or because the patient is in a poor condition and would not be able to tolerate surgery; the patient may also refuse surgery. For these patients, radiotherapy is an alternative to surgery. Most recently, Mendenhall et al. reported the treatment of patients with stage I–III salivary gland carcinoma using radiotherapy alone. They achieved 10-year overall survival and local control rates of 65% and 75%, respectively. For patients with stage IV salivary gland carcinoma, the 10-year overall survival and local control rates were 21% and 21%, respectively.[5]

Due to the possibility of permanent functional and/or cosmetic deficits from the side effects of routine external radiotherapy, the use of radiotherapy for

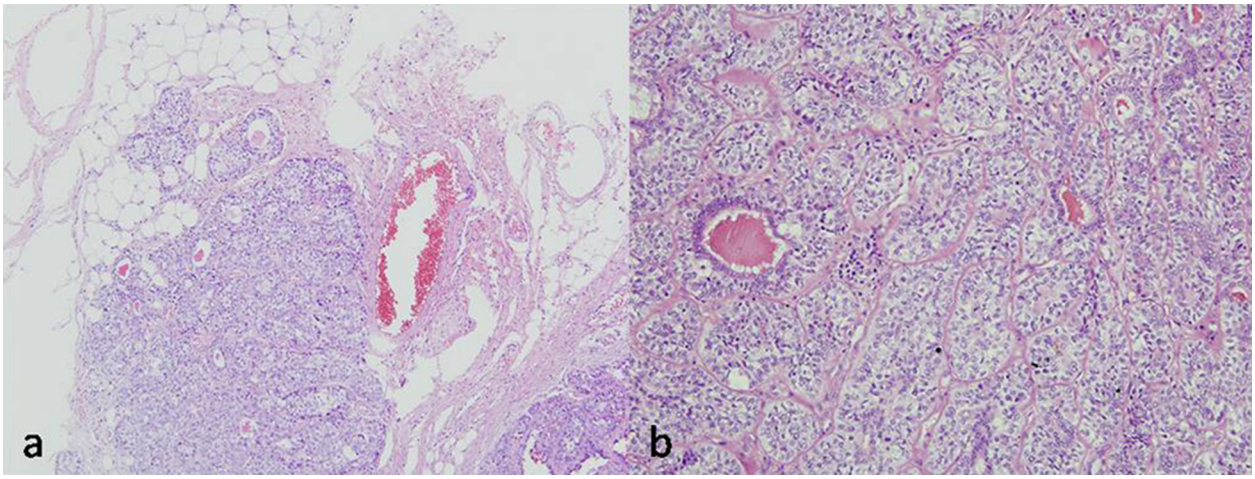

Fig. 2. Histopathology showing epithelial-myoepithelial carcinoma (haematoxylin and eosin stain; original magnification (a) 100×, (b) 200×).

salivary gland tumours in children remains controversial.[6] Here, we report the successful use of iodine-125 brachytherapy for the treatment of a young EMC patient. At the 6-year follow-up, no local recurrence or distant metastasis was detected and there were also no functional or cosmetic deficits.

The photon energy of an iodine-125 seed is low (27–35 keV) and its ray irradiation distance is short (17–20 mm). The energy is focused on a target area and reduces sharply with distance, which reduces the damage to the surrounding tissues and minimizes the risk to the adjacent vital structures. The dose of the seed in a target area is over twice that obtained with routine external radiotherapy. There is much clinical data indicating iodine-125 brachytherapy to be a promising alternative method to external radiotherapy for some malignant tumours, such as those of the head neck region and prostate cancers.[7–10]

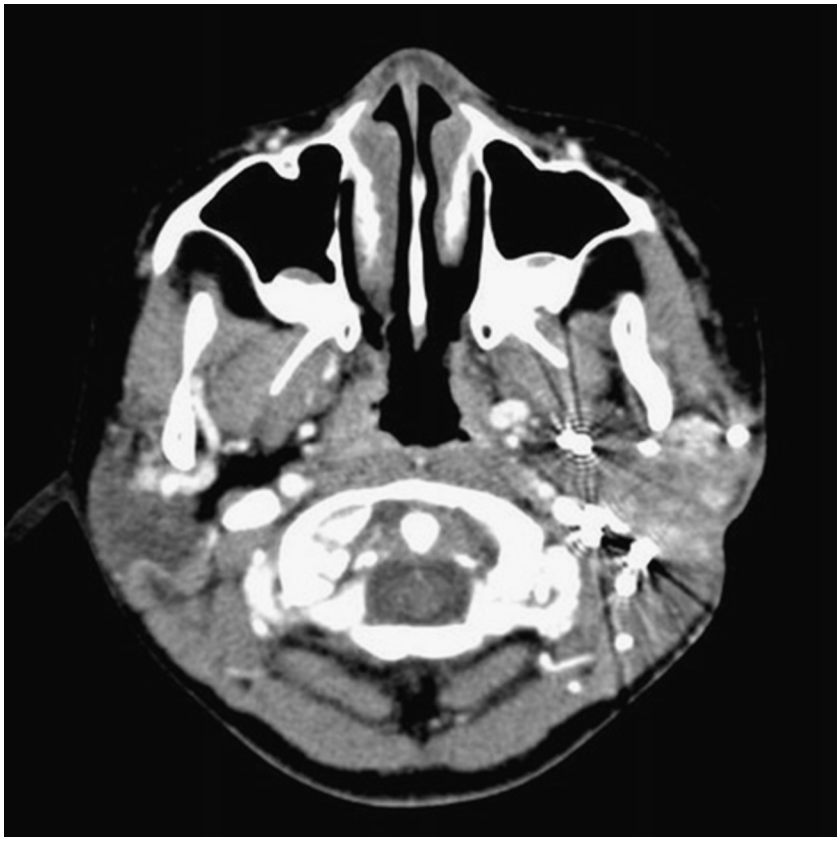

Fig. 3. CT scan showing the tumour size reduced by about 80% at 2 months after the implantation.

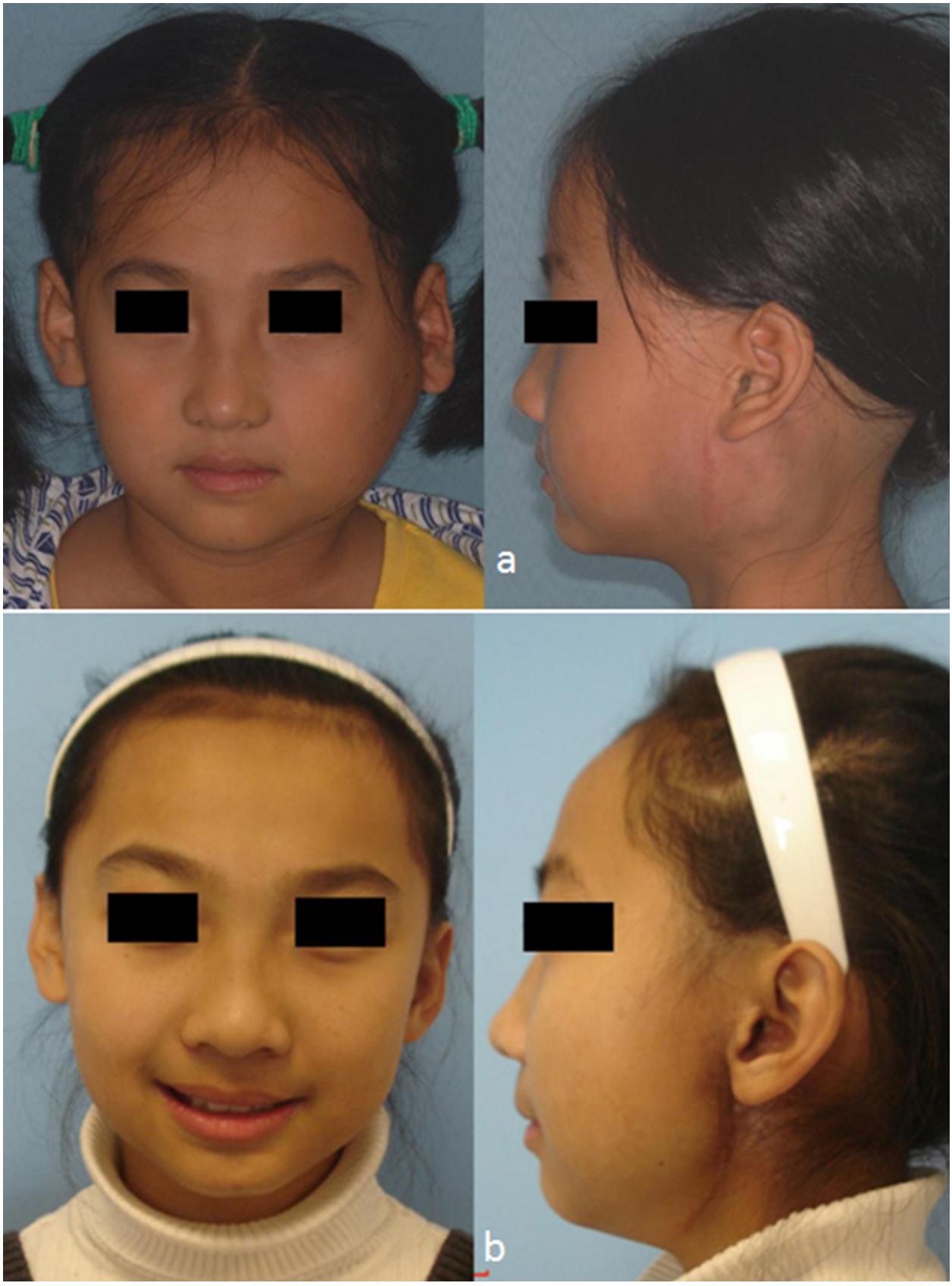

Fig. 4. (a) Anteroposterior and lateral views of the left large parotid gland tumour before seed implantation. (b) Anteroposterior and lateral views of the left parotid gland region at the 6-year follow-up: no mass and normal skin.

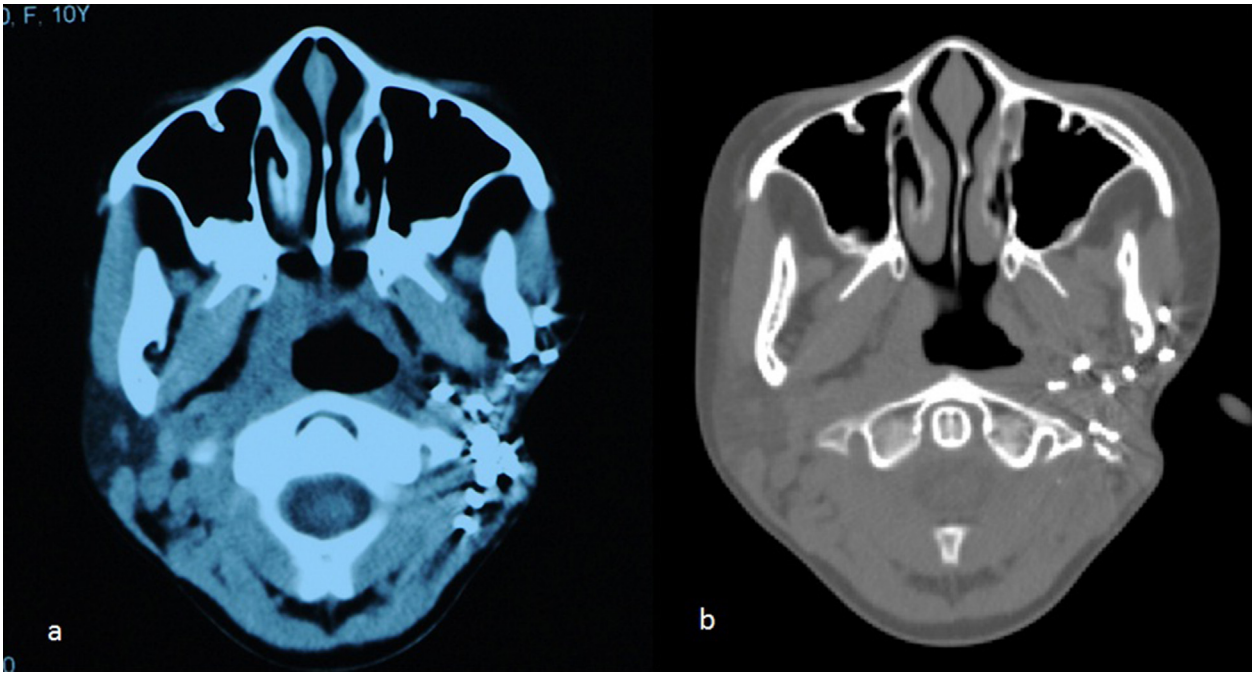

Fig. 5. CT scans showing no tumour recurrence at 3 years (a) and 6 years (b) after seed implantation.

In conclusion, iodine-125 brachytherapy is a potentially appropriate alternative treatment for unresectable EMC in children.

Funding

None.

Competing interests

None declared.

Ethical approval

The study was approved by the Ethics Committee of Peking University School of Stomatology.

Patient consent

Patient consent was obtained.

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