A Rare Case of Infantile Hemangioma Requiring Ear Reconstruction

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1. Introduction

Infantile hemangiomas (IHs) are the most common soft-tissue tumors in infancy. Ear deformity due to IH is well-known, and surgical correction is sometimes necessary. However, to our knowledge, there are no cases of cartilage transplantation. We present a rare case of ear reconstruction of the lower two-thirds, lost due to IH, along with a literature review.

2. Case report

A 6-year-old girl underwent partial ear reconstruction of her left ear. She was born with a weight of 3470 g as a full-term newborn. A small red macule on her left auricle gradually became swollen and ulcerated. She presented to a local hospital at 1 month of age and was diagnosed with hemangioma but received no treatment. Access to the expertise was limited in her hometown.

Her tumor continued growing and began bleeding; thus, she was referred to our hospital at 3 months of age.

Her left ear was disfigured with a partial loss of cartilage and a red mass with multiple ulcers (Figure 1). The patient underwent a pulsed dye laser (SPTL-1b Syneron Candela) treatment as a proliferating IH. The ulcers healed after four procedures. Propranolol therapy was not applied because it was off-label use in Japan at that time. At the age of 5 months, the IH started to involute, and laser therapy was discontinued.

Her left ear lacked a part of the helix and lobule. The tragus, antitragus, and concha were preserved. Mature scars were left around her preexisting lobule area, and the postauricular skin was thinner than normal (Figure 2). At the age of 4 years, ear reconstruction, including cartilage transplantation, was started. Because any flap necrosis was worrisome due to fragile soft tissue condition, a 3-step surgery was conducted.

Rib cartilage transplant was performed as the first step. The sixth and seventh cartilages were harvested, and a base frame with the lobule and helix unit was constructed. The shape of the ear was designed on the postauricular skin according to the contralateral side. The skin was incised at the posterior edge of the design. A thin skin flap was carefully raised, including the scars. The cartilage frame was inserted under the skin flap. We postponed an additional skin incision for the ear and rib cartilage connection, considering the risk of flap necrosis.

Figure 1. First visit view of the patient’s left ear at three months old. Helix is already lost, and multiple ulcers are located on the IH. The borderline of lobule is vague. IH, infantile hemangiomas.
of potential skin necrosis. Some of the harvested cartilages were stored for ear elevation under her chest skin, and the rest was diced and replaced inside the perichondrium. No skin necrosis was observed after the first surgery.

The second step of surgery was conducted 4 months later. The rib cartilage frame and rest of the ear cartilage were connected firmly with 38-gauge wire (Figure 3). No complications were observed after the second step of surgery.

Seven months later, the third step surgery for ear elevation was conducted. The skin was incised along the posterior edge of the reconstructed ear cartilage. A triangle skin flap was raised to wrap the posterior surface of the ear lobe.

Figure 2. Preoperative view of left ear at four years old. A part of helix and lobule are lost. Tragus, antitragus and concha were preserved. Scars were located around her preexisting lobule area.

Figure 3. First step surgery (A) Transplanted ear frame was constructed from sixth and seventh rib cartridge. Ear lobe frame is pointed by arrow. B, Ear frame was inserted under the skin through postauricular incision. A unit of transplanted cartridges was not connected to the original cartridge in order to prevent skin flap ischemia. Note the ischemic color of the flap over the cartridge.
Previously banked cartilage was taken, shaved, and fixed at the mastoid surface of the reconstructed ear cartilage as a strut. A square-shaped superficial mastoid flap was raised and covered the cartilages (Figure 4). The rest of the raw area was covered by a full-thickness skin graft from the groin. Good shape and projection of the ear were achieved without any complications at 5 months after the third surgery (Figure 5).

Discussion

Infantile hemangiomas are the most common soft tissue tumors in infancy. They have a period of rapid growth (proliferating phase) followed by gradual involution (involuting phase).

Because most IHs tend to regress spontaneously, treatment is only required for complicated cases. However, without any treatment, residual consequences occur in 70% of cases, namely telangiectasia, excessive fibrofatty tissue, and skin laxity, and previously ulcerated lesions usually leave scars.1 Indications for treatment include life-threatening IHs, IHs with functional impairment, and IHs accompanied by disfigurement. In cases of IHs with manifest obstruction or ulceration, immediate therapy is required. In the proliferative phase, therapy is directed at the induction of growth arrest and remission. After incomplete regression, excessive fibrofatty tissue and scars can raise cosmetic concerns that need to be addressed.1 The emergence of propranolol has reduced tragic destruction by IHs, but almost half of patients treated with propranolol still require surgical intervention.2 This patient’s ear required its reconstruction, considering the scars and fibrofatty tissue left around the preexisting lobule area, which could have made the skin flapless durable to ischemia. Fu et al reported that skin necrosis occurred in 5.39% of microtia patients treated using the Brent and Nagata technique cartilage transplant.3 Skin necrosis can lead to cartilage infection that should be avoided.3 We suggest that a safer 3-step reconstruction was preferred in this case.
In addition, we transplanted the cartilages frame to reconstruct both the middle and lower thirds of the ear, including the lobule and achieved a smooth continuous outline of the ear. A small notch at the cartilage joint point and slight posteriority rotation was noted, but they were acceptable. The rotation is estimated to be the result of relatively heavy reconstructed lower ear and posterior scar contracture.

In the majority of past reports, the middle third of the ear has been reconstructed by cartilage transplants, while the lower third has been rebuilt by soft tissue.4,5 Reconstruction method of middle and lower thirds of the ear has not been established. Our method is worth considering as an option.

Conclusion
We introduced a rare case of ear IH that required cartilage transplantation. We chose a 3-step surgery, considering the risk of necrosis derived from fragile soft tissue condition. A cartilage frame was inserted to reconstruct the lower two-thirds of the ear, including the lobule. Good appearance and projection of the ear were obtained without complications. To our knowledge, no study has reported on cartilage transplants for IH-affected ear, and lower two-thirds of ear reconstruction is rare. In the case of IH-affected ear reconstruction, we propose to choose a safe and personalized method according to each defect and condition of soft tissue.

References