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CASE REPORT

Question Mark Ear Deformity: A Combined Method for Correction

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Abstract Question mark ear deformity or Cosman ear is a very uncommon congenital alteration. The deformity includes a cleft between the posterior helix and the lobule, an increase in anterior projection, an abnormal superior third that modifies the superior crura and the scaphoid fossa, partial or complete absence of the antihelix, transposition of the lobule and antihelix (severe cases), and postauricular tags. The authors present a case of moderate question mark ear deformity treated using Mustarde sutures and two cartilage grafts to correct the support and the contour defect. Adequate correction of the deformity and symmetry was achieved for both ears. The technique described in this report is suitable for minimal to moderate defects.

Keywords Congenital web neck · Secondary surgical correction · Severe case

Question mark ear deformity is a congenital alteration of the pinna rarely reported in the literature. It was first described by Vincent et al. [1] in association with urogenital anomalies. After that report, Cosman et al. [2] presented two similar cases and described the similarity to the question mark.

Congenital auricular deformities affect 0.8–2.4 of 10,000 live births, with a lower incidence among Caucasians and Afro-Americans than among Hispanics and Asians. In the latter group, the incidence can be as high as 55% of the general population [3].

The Cosman ear includes six different characteristics in most cases, with a wide range of severity: a cleft between the posterior helix and the lobule, increased anterior projection, an abnormal superior third that modifies the superior crura and the scaphoid fossa, partial or complete absence of the antihelix, transposition of the lobule and antihelix (severe cases), and postauricular tags.

The deformity can occur uni- or bilaterally. The incidence is not clear, nor is the etiology. However, in some cases, this deformity appears related to and behaves as a minimal form of the auriculocondilar syndrome [4], in which micrognathia (with glossoptosis and upper respiratory problems), prominent cheeks, stenotic ear canals, and hearing loss can be found.

Most reports describe isolated cases [5–10]. The largest reported series included 32 patients from China [11] and proposed a classification to aid in recommending specific treatment. This deformity is not incorporated in any other classification, but it may represent a considerable defect.

We present our experience correcting a moderate case of question mark ear deformity using different techniques, with a satisfactory result and no complications.

Case Report

An 8-year-old girl presented with a right ear malformation. The left ear lacked definition of the superior crura. The right ear showed a middle-third defect that included a cleft over the posterior helix as well as absence of the distal scapha and the superior crura. The height of the right ear was 61 mm and that of the left ear was 55 mm (Fig. 1).

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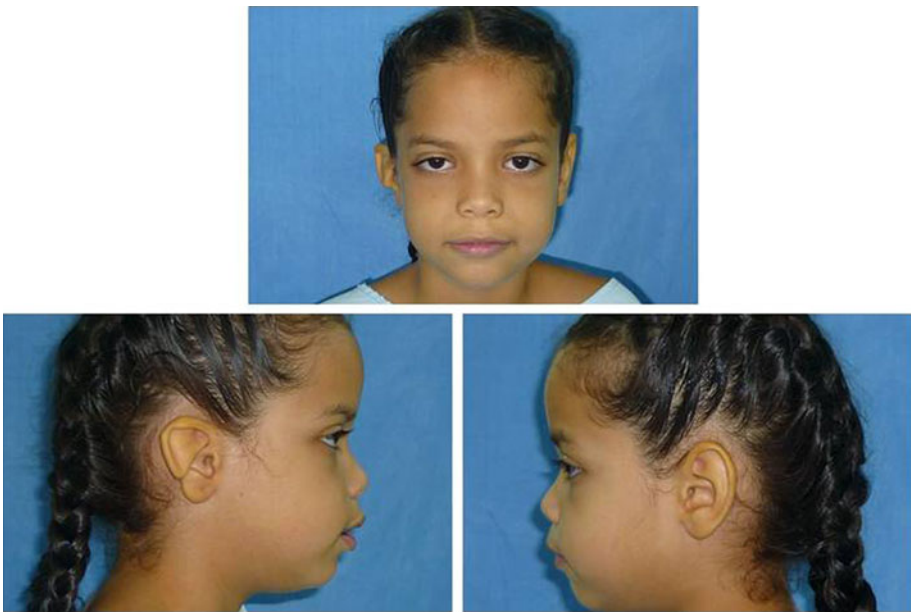

Fig. 1 Preoperative view

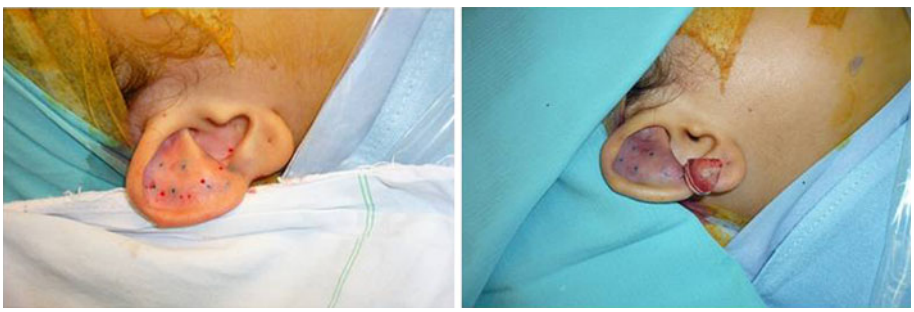

Fig. 2 Intraoperative view. *Right* Mustarde marks. *Left* Conchal cartilage graft

Surgery was planned to correct the middle-third deficiency and the upper prominence. The procedure was performed with the patient under general anesthesia.

From the left ear (normal ear) using a posterior incision, we harvested a conchal cartilage graft to be used in the contralateral ear. After sharp scoring of the cartilage, three Mustarde sutures were performed to recreate the superior crura using Prolene 5-0.

On the right ear, we also made a posterior incision and performed a subpericondrical dissection. The cartilage graft then was tailored to correct the cleft where no cartilage support was found and skin deficiency was moderate. A second graft was tailored to complete the antihelix contour that was absent on the middle third (Fig. 2).

After placement of both cartilages, Mustarde sutures (Prolene 5-0) were placed to form the superior crura and the antihelix, including plication of the graft in the cleft. External sutures then were placed to guarantee the desired contour and immobilize the cartilage grafts. Both ears were sutured with Vicryl 5-0 and Monocryl 5-0. After 7 days, moderate edema was found, but the desired contour remained.

The wound healed uneventfully, and the corrected auricle looked similar in shape and size to the left auricle after 1 month of follow-up evaluation. Minimal epidermal loss was observed in relation to the external fixation, but no additional scars or marks were observed later. The final result after 6 months is shown in Fig. 3. It can be noted that the contour of the antihelix is irregular, possibly secondary to an abnormal rotation of the second graft. A minor correction of this remaining abnormality might be performed after the scarring process is completed. The patient is satisfied and can now do her hair in any style and go to school without any concerns.

Discussion

Question mark ear deformity is a rare but morbid congenital defect. There is no consensus about the etiology or the appropriate treatment.

The development of the external ear has recently been a subject of debate. As understood traditionally, both the first and second branchial arches contribute to auricular

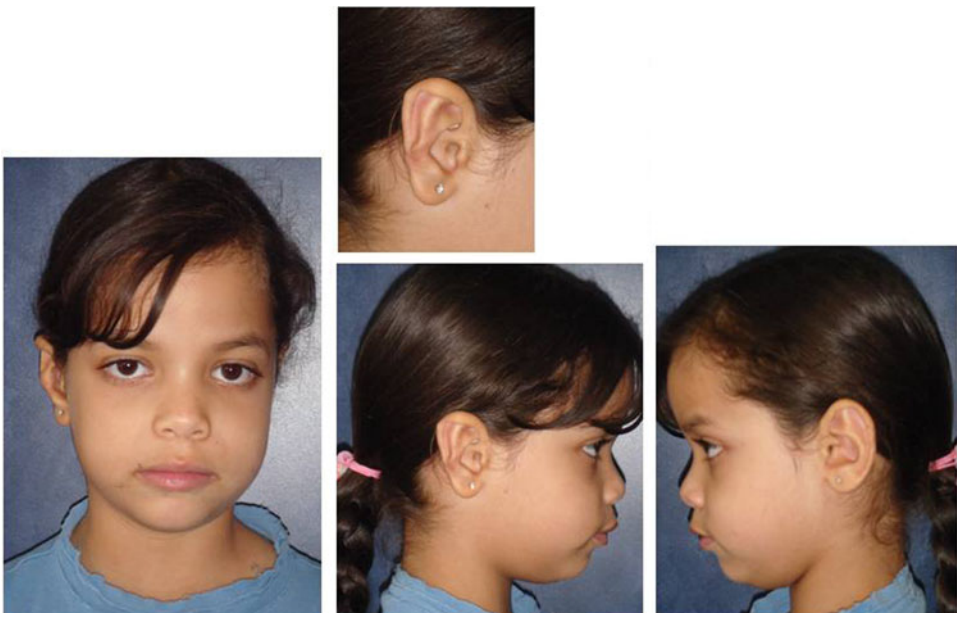

Fig. 3 Clinical results for both ears after 6 months

development. The pinna begins to form during the 5th week, when six hillocks appear. The three anterior hillocks (first arch) will form the tragus, the helical root, and superior helix, and the three posterior hillocks (second arch) will form the antihelix, antitragus, and lobule [12].

More recently, Porter and Tan [3] have suggested that these hillocks are incidental and not fundamental to the development of specific auricular components. They are transitory and represent temporary foci of intense mesenchymal proliferation. The division of the dorsal parts of the arch into a ventral and dorsal component is more important. The ventral division will form the tragus, antitragus, lobule, helix root, and anterior helix. The dorsal division will form the antihelix, inferior crus, triangular fossa, and concha. The remainder of the external ear (superior and posterior helix, superior crus, and scaphoid fossa) will derive from the free ear fold, which develops separately.

Numerous theories have been proposed to explain the etiology of this congenital anomaly, but none seems to be satisfactory. It is clear that some type of disturbance must occur to produce the congenital auricular cleft. Different reports have mentioned the traditional theory, proposing a lack of fusion between the fifth and sixth hillocks or on the other side. A real defect of the mesenchyme during the proliferation phase can be present.

Patients presenting with question mark ear deformity must be carefully studied to discard associated craniofacial or urogenital malformations. When the plastic surgeon truly knows that it is an isolated deformity, surgery is planned to correct the defect. The appropriate age for the surgery still is a topic of debate, but indications for otoplasty to correct similar defects such as prominent ears might be acceptable for these cases. The technique chosen should be adjusted to the severity of each case and the amount of height that needs to be corrected.

Different techniques have been reported for the correction of this malformation including cartilage resection [2], double z-plasty [2], postauricular flaps, chondrocutanoeus flaps [8, 9, 13], chondrocutaneous graft, cartilage sharp scoring, Mustarde-type sutures, conchomastiodal fixation, and tissue expansion [11].

In mild and moderate cases, the middle-third cleft is evident, but the skin has enough laxity to be used as external coverage. Therefore, the final correction is performed by modifying only the structural support, so contour and height are restored only with cartilage grafts, as in the reported caser.

For more severe cases, the structural support must be corrected. However, in most cases, local soft tissue coverage is deficient, so local flaps are the reasonable option. It is important to clarify that those reconstructions requiring only one surgery are the best choice if they are available. For correcting severe cases, chondrocutaneous grafts are an unsafe option because of the large size needed and the significant donor-site morbidity of the normal ear. But even for less complex deformities, these grafts are not necessary due to the skin laxity at the same level of the cleft that allows molding and complete correction of the contour deficiency.

Tissue expansion might be considered an option for severe cases, bilateral compromise, or previously failed reconstructions, in which available local tissues are deficient. It is important to remember that this procedure requires two surgeries and that the incidence of local complications in this particular area is high.

We report one case of moderate question mark ear deformity, which was treated using Mustarde sutures and

two cartilages grafts to correct the support and the contour defect present in this deformity because of the middle-third hypoplasia. The height deficiency was corrected using the antihelix plication (Mustarde sutures), a safe technique used also in otoplasty with good long-term results. In addition, we applied two cartilage grafts from the contralateral concha using a minimal posterior incision that allowed modifications in the normal ear to achieve symmetry. There were no evident scars for either ear because all the incisions were posterior. The postoperative result was satisfactory, with adequate correction of the deformity and symmetry for both ears. Only a minor contour deficit was present after 6 months of follow-up evaluation.

The advantages of this surgical technique include only one surgical time, minor donor-site morbidity, inconspicuous scars, and minor dissection. The possible disadvantages are the unpredictable behavior of cartilage grafts because of long-term rotation and reabsorption. The technique described in this report is suitable for minimal to moderate defects using simple techniques with low morbidity.

Conflict of interest None.

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