The Nijmegen Auricle Consultation: How We Do It?

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Abstract

Introduction: The quality of healthcare optimization for patients with an absent or a deformed auricle is of great importance. The absence of the auricle has various causes, and the management differs between patients with congenital microtia and patients with amputated ears due to a malignant tumor or trauma. Auricular reconstruction using autogenous rib cartilage grafts and auricular replacement with implant-retained prostheses are the standard techniques used at the Radboudumc, Nijmegen.

Aim: The aim of this study was to create more awareness, and to enhance the quality of care for patients with an absent or deformed auricle.

Material and methods: All caretakers at Radboudumc who are involved in the healthcare of patients with an absent or a deformed auricle either aiming for hearing revalidation, an auricular reconstruction, or the application of an implant-retained prosthesis, or a combination of these, were interviewed through a questionnaire. Patients' opinions regarding the auricle consultation were noted on a Visual Analogue Scale (VAS).

In addition, the patients who had undergone auricular reconstruction or received prosthesis over the past five years were asked to fill out the Glasgow Benefit Inventory (GBI).

Results: The level of entrance in this chain of healthcare did not influence the end point for patients visiting the Nijmegen auricle consultation. The possible three end points either with or without rehabilitation were (1) accept or wait, (2) auricle reconstruction with autologous rib cartilage grafts, and (3) auricle reconstruction by an implant-retained prosthesis. Interviews and questionnaires were evaluated to create a flowchart. The patients rated the auricle consultation with an average score of 9. All patients who underwent auricle reconstruction and filled out the GBI stated that their quality of life was improved.

Discussion: The present study describes the management of patients with an absent auricle at the Radboudumc in Nijmegen, the Netherlands. Due to the low incidence of these patients and the subsequent requirement for specialized care, patient-centered care is extremely important, and is offered at two experienced specialty centers in the Netherlands.

Keywords: Microtia; Auricular Reconstruction; Prosthesis; Hearing Loss; Quality of Life; Implants

Abbreviations

Visual Analogue Scale (VAS); Otorhinolaryngology (ORL); Glasgow Benefit Inventory (GBI); Audiological Centre (AC); Bone Conduction Device (BCD); Middle Ear Implant (MEI).

Introduction

An important goal in the management of the patients with an absent auricle seeking for auricular reconstruction or implant-retained prosthesis is to improve the quality of life. The absence of the auricle has various causes, several grades [1], and its management differs between patients with congenital conditions and patients with amputated auricle due to a malignant tumor in the region of the auricle.

Since the description of the upper and lower auricle reconstruction by Diefenbach in 1845, a number of techniques have been developed to reconstruct the absent or deformed auricle [2]. Most of the current techniques have been developed after 1950 when the number of publications and textbooks related to auricular surgery increased rapidly [3].

The introduction of osseointegrated implants in the temporal bone for bone-conduction hearing in 1977 and auricular prostheses in 1983 [4] has revolutionized prosthetic possibilities for the auricle [5]. This technique has evolved with the development of new devices and new options for the prosthetic auricle [6-8].

Currently, four different techniques are known for reconstruction of the auricle [9]. First, the surgical reconstruction of the auricle using autogenous cartilage [10]; second, using porous polyethylene (Medpor®) [11]; third, the reconstruction of the auricle by an implant-retained prosthesis of the auricle [12]; and fourth, the reconstruction of the auricle through tissue engineering, which is still in the preclinical phase. A few challenges that persist are appropriate scaffold design, cell source selection, and long-term sustainability [13,14]. Currently, studies have been conducted to overcome these challenges and to use this technique for clinical application [15].

The reconstruction of the auricle using autogenous rib cartilage or an auricular implant-retained prosthesis are the most frequently used standard techniques at the Radboudumc. Magritz and Siegert [16] described the surgical reconstruction technique with an autologous rib cartilage. This is not only the preferred technique used in the Radboudumc, they also work in close collaboration.

Specific healthcare demands are required to treat this specific group of patients. First, attention to the hearing ability of the patient; second, availability of a passionate and specialist surgeon for the treatment of patients; and third, a multidisciplinary approach to address all issues, including psychological aspects. All members of the multidisciplinary team should be aware of the trajectory of care.

A flowchart on the patient's routing within this specific care and chain could contribute to further improve the quality of care in patients with an absent or deformed auricle.

Material and Methods

A questionnaire (Appendix I) was developed at the Radboudumc to evaluate the possible pathways for patients with a missing auricle and to aid for hearing revalidation, auricular reconstruction/prosthesis, or a combination of these. This general questionnaire included three categories. The first category comprised general questions and the second category comprised questions about the specific pathways a patient can follow. The last and third category included a few additional questions.
Using this questionnaire, caretakers from all disciplines at the Radboudumc who are involved with patients with absent auricles have been interviewed about their participation within the various pathways.

A list of caretakers was determined before the interview. It included (Appendix 2) comprising everyone who is involved in healthcare, either with high or low contribution. The answers to the questionnaire and other data acquired during these interviews were converted into a flowchart. The goal of the flowchart was to describe all possible routes within the Radboudumc as clearly as possible. The audio of the conversations was recorded with their consent.

In addition to these interviews, all patients visiting the auricle consultation over the past five years were asked to fill out a Visual Analogue Scale (VAS) to score their experiences. Moreover, the data of the quality of life were obtained from those patients who underwent auricular reconstruction or received an implant-retained prosthesis at the Radboudumc over the past five years. The instrument used was the Glasgow Benefit Inventory (GBI), which was developed particularly for otorhinolaryngological interventions [17,18]. Patient's benefit is the change in health status resulting from healthcare intervention. The GBI was developed to be patient-oriented, and to be maximally sensitive to otorhinolaryngology (ORL) interventions. The GBI is an 18-item, post-intervention questionnaire intended to be given to patients [19].

**Results**

The flowchart in figure 1 is a simplified version of the original flowchart. The Parts of the original flowchart were used to clarify the process.

**Referral to the otorhinolaryngology outpatient clinic**

Independent of the etiology, patients were directed from three different places to the ORL outpatient clinic (Figure 1). General practitioners or secondary referrers, such as internal or peripheral specialists, can choose to refer to the ORL outpatient clinic directly or indirectly via the Audiological Center (AC). The choice for either one depends on the expectations of the doctor, and the demand for care of the patient. In other words, a patient will be referred to the AC in case hearing problem is expected. If the referral is solely concerned about the auricle, the patient will be referred to the ORL outpatient clinic. Thus, the entry does not influence the outcome of the treatment, because ORL and AC work closely, both internally, referring patients to each other according to the requirement.

**Etiology**

The group of patients referred to the ORL outpatient clinic can be divided in three categories based on etiology.

1. Patients with a congenital abnormality of the auricle
2. Patients with a tumor of the auricle
3. Patients with an absent auricle by burn or trauma. This type of patient is less common, and therefore has not been extensively discussed in this paper.

**Patients with a congenital abnormality of the auricle:** The patients with a congenital abnormality were referred to the auricle consultation within the ORL outpatient clinic (Figure 2).

Microtia can arise as a sole mutation or in the context of a syndrome like Goldenhar syndrome, Treacher-Collins syndrome and hemifacial microsomia [20]. In case the patient has a concomitant cleft palate, the multidisciplinary Nijmegen cleft palate team consisting of pediatricians, cleft palate-nurses, plastic surgeons, orthodontists, phoniatrists, speech-language pathologists, oral maxillofacial surgeons, clinical geneticists, dentists, otologists and neurosurgeons, is the main caretaker. From there the patient can visit the auricle clinic.

**Patients with a tumor of the auricle:** The patients with a tumor of the auricle were referred to the head and neck oncological outpatient clinic within the ORL outpatient clinic (Figure 3). Working of the oncology clinic includes an intake by a nurse and a combined intake with an otorhinolaryngologist, oral maxillofacial surgeon, and a radiotherapist. A different, subsequent treatment may include dietitian, X-ray of the lungs, Computerized Tomography (CT) scan, Magnetic Resonance Imaging (MRI) scan, ultrasound, or screening consultations by geriatriest, anesthetist, or dentist. Additionally, in our 3D laboratory it can be determined if sufficient temporal bone volume is present to allow implant placement. Obviously, patients at risk for an auricle amputation will opt for this consultation.

![Figure 1: Simplified flowchart.](image)
Because of priority reasons [21], patients will be referred only to the auricle consultation after the tumor removal instead of within the oncological treatment. Although radiotherapy increases the risk of implant loss, an implant-retained prosthesis is still a treatment option [22].

**The auricle clinic**

The auricle consultation (Figure 4) comprises an otologist, an anaplastologist, and a psychologist. A psychologist assists in the communication between a doctor, a patient, and parents, and gives psychological support, if necessary.

At this consultation, which is held approximately once a month, the patient will get all information they need, including the possibilities for hearing rehabilitation, and all the options regarding reconstruction of the auricle. Between June 2011 and May 2016, the auricle consultation held 31 meetings, at which 153 patients were seen in a total of 197 visits. In most cases, the patient was brought only for a few weeks after their birth. An implant-retained prosthesis can only be provided when the patient is six to seven years old; although it is preferable to wait until at least eight years of age to have psychologically more matured child prior to making these decisions. An auricular reconstruction can be provided when the patient is ten years old [23]. During the first visit at the auricle consultation, most patients are too young for an auricular reconstruction or a prosthesis. Therefore, it is extremely important to inform the patient or his/her caretakers about the trajectory of care, the time schedule, and the
possible obstacles a family of a child with microtia could encounter. After the hearing is optimized, and a strong relationship is built between the patient and the doctor with its team, they can together decide upon a possible next step regarding the auricle, always in combination with the hearing revalidation.

Results section-1: Outcome and Endpoints

For the outcome and end points, see figure 5.

A. Regarding the strategy in case of absence of an auricle, three end points can be defined:

I. Accept and wait: After the patient visited the auricle consultation, he/she can choose to leave the absent or deformed auricle untreated. We empower people to believe in themselves. When the patient should change his/her mind, they can always visit the auricle consultation again. The same strategy can be followed for hearing in case of congenital aural atresia. Between 2011 and 2016, 45% of the patients choose to accept and wait. This group also includes patients who already underwent an auricular reconstruction elsewhere or have already received an implant-retained prosthesis. They visited the Radboudumc for a second opinion, and ultimately, decided to do nothing about their hearing.

II. Auricular reconstruction: This is offered patients who have attained the age of ten years, or at least, if the patient’s chest circumference is at least 63 cm; only then, sufficient rib cartilage is available for reconstruction of the auricle [23].

The use of porous polyethylene (Medpor®) for auricular reconstruction has a few advantages compared with autologous rib cartilage. Only one operation is needed to reconstruct the auricle. In addition, no rib cartilage needs to be harvested with donor-
site morbidity; and therefore, a reconstruction with Medpor® can be performed at an earlier age. However, only few surgeons are trained at this, leading to a relative lack of understanding of the nuances of this specific type of surgery. Therefore, reconstruction with Medpor® is known to have a higher chance of repulsion, infection, and deformation of the auricle [24,25]. At our center, in combination with our philosophy, we have chosen to in principal use only Medpor®, in revision cases where there is no useful rib cartilage left.

When a patient opts for an auricle reconstruction with autologous rib cartilage, a schedule for the sequence of operations is made. The second operation is planned six months after the first operation. Sometimes, a third operation is necessary to perform some refinements. At the Radboudumc, in the studied period about 8% of the patients eventually chose to get their auricle reconstructed with rib cartilage.

In case of a malignant tumor of the auricle, the success rate of an autologous rib cartilage reconstruction is too low to be a serious alternative for an implant-retained prosthesis (see end point III).

III. Prosthesis: After the auricle consultation, the patient can opt to have his/her auricle reconstructed by an implant-retained prosthesis. Parallel to the referral, at the Centre of Special Dental Care, an otologist will arrange a CT scan, and will involve the 3D-lab within the process.

The 3D-lab uses the CT-scan to:

1. Mirror the opposite auricle in case of unilateral microtia,
2. To observe if sufficient temporal bone is present to allow implant installation at the desired symmetric position,
3. To manufacture a surgical template for placing the implants as accurate as possible.

When the exact positions of the implants are determined by the 3D lab along with confirmation by an otologist, the implants will be placed in the operating room using the prefabricated surgical template. In case of implant placement in the same session of the ablative surgery, 3D preoperative implant planning is not always beneficial, as sometimes the temporal bone is removed perioperatively.

When Branemark implants are used, the implants are uncovered after a three-month osseo-integration period, the abutments are placed, and the wound is closed. In case Vistafix® implants are used, abutments are placed in the same session as implant installation (single stage surgery), and loading can be started after 4 weeks. These implants use the same Bi300 implant with some differences in the concomitant abutment, which have shown optimal results over the years [26].

An anaplastologist uses the 3D-printed mirrored auricle to manufacture the prosthesis. The advantages of prosthesis are that the surgery for the implants is short and the implants can be placed in patients from four years of age, when the skull has outgrown the anaplastologist also conducts the follow-up. However, when implant-related complications appear, an otologist or a maxillofacial surgeon can be consulted.

At the Radboudumc, in the studied period about 10% of the patients eventually chose to acquire an auricular prosthesis at the Radboudumc, including two patients who were dissatisfied with their autogenous auricular reconstructions in the past.

B. Regarding the strategy in hearing, four end points can be defined:

I. Accept and wait: In comparison to the strategy in case of absence of an auricle, when a patient changes his/her mind, one can always visit the auricle consultation again and decide to go for hearing rehabilitation by means of a bone conduction device (BCD) or middle ear implant, among others. At first, an audiogram is conducted before proceeding to one of the three end points.

II. Atresia repair: The surgical opening of the external meatus improves hearing in patients with an atresia type 1 and atresia type 2a [29]. In case of patients with atresia type 2b or 3, there are expected to be too unfavorable to reconsider surgery, especially in comparison with the results that are currently achievable with BCD, or middle ear implant (MEI) [30,31]. To decide which hearing implant suits best for a patient, the expert opinion of an otologist is obligatory. This depends mostly on their bone conduction thresholds, in combination with risks of the middle ear surgery and inability to effectively use MRI scanning in certain cases [32-34]. In addition, patients with milder atresia might opt for accept and wait or a (semi) implantable hearing aid.

III. BCD: A BCD, on a softband, is offered to patients from 6 months of age. Approximately 6% of the patients with microtia were bilateral, and their sound deprivation necessitates even earlier intervention [35]. All these patients need a BCD, preferably two, on a softband from three months of age [36]. When the skull is thick enough mostly around the fourth year, a bone implant can be placed. During the placement of these implants, a possible reconstruction of the auricle eventually has to be taken into account regarding the positioning of the implant.

At the auricle consultation in the studied period 45% of the patients opted for a BCD.

Results section 2: Patient outcome measurements

Regarding the flowchart of this study, only physicians and other employees at the Radboudumc were interviewed. Every interviewed person gave consent to record the conversation.

Patients were not interviewed to inventory their point of view at these different pathways. However, patient’s satisfaction was reviewed by means of another measures, namely a generic questionnaire (VAS) after they visited the auricle consultation. Patient’s satisfaction showed an average score of 8.7 (N = 25, range 7–10), standard deviation = 0.78).

Followed by this generic questionnaire, all patients who underwent any type of intervention were asked to fill out an instrument, or so called quality of life questionnaire, namely the GBI [17,18]. Patients who opted for an auricular reconstruction by rib cartilage or implant-retained prosthesis were asked to fill out the benefit instrument after surgery. In all, 66% of the patients filled in the questionnaire. The mean follow-up time was 2.35 years (maximum = 10.75 years, minimum = 5.03 months). All the patients stated that their quality of life was improved and that they felt better post the auricular reconstruction or implant-retained prosthesis. Except for one, who stated that nothing changed with respect to quality of life. Overall, patients stated that they were more optimistic, less ashamed, showed more self-esteem, participated more in social activities, and felt less uncomfortable.

The satisfaction about the first visit of the patients at the auricle consultation was reviewed later. 100% of the patients that received this questionnaire completed and returned this questionnaire and they rated the overall auricle consultation with an average score of 9 (range 7–10). They all stated that they received enough information at the auricle consultation.
Discussion

This study describes the treatment management for patients with an absent auricle at the Radboudumc in Nijmegen. The prevalence of patients with microtia (1:10,000), and the incidence of tumors of the external ear canal is low (1:6,000,000) [37-39]. Therefore, the awareness of this healthcare chain is relatively low, and multidisciplinary care must be offered at centralized specialty centers. Patient-centered care is extremely important, and is offered at specialized expertise centers, of which there are two in the Netherlands (Radboudumc, Nijmegen and WKZ, Utrecht). The importance of this study is to evaluate the current patient flow and patient satisfaction of the treatment itself. The evaluation entailed interviews of professionals involved in this chain of healthcare, and assembled information in a flowchart.

One of the most important findings of this study was that the level of entry did not determine the end point of the patient with an absent auricle. Likewise, the choice of the peripheral doctor did not influence the end point of the patient. Patients were offered the same options as any other patient. As such, the patient is not dependent on the way healthcare is organized. Despite that, the choice of entrance does not influence the end point of the patient; this pathway is not always the most efficient or desired pathway for each patient with an absent auricle. The flowchart will ensure that professionals are acquainted with the patients care, and therefore can inform patients better.

This study attempts to describe a flowchart of the tertiary healthcare for patients with an absent auricle in its total aspect. To the best of our knowledge, these flowcharts have not been described before in the Netherlands. Other well-known centers from all over the world, with great expertise in treating patients with absent auricles have describing their workflow. Three of these centers (Magritz, Recklinghausen, Germany; Wilkes, Alberta, Canada; and Staffors, Gothenburg, Sweden) have been contacted. They stated by personal communication that they organized the healthcare for patients with an absent auricle in a similar way, just like the flowchart we describe.

Minor discrepancy was seen in the results of the interviews. The different end points and numbers described in this retrospective analysis showed that indeed every patient is different, and indicated patient-centered care. Some caretakers stated that every oncology patient wanted their amputated auricle back in form of an implant-retained prosthesis, while others stated that this was not the case for all patients. In literature, there is still some debate about the use of a BCD in case of an unilateral hearing deficit [30,40-42]. BCD implantation in patients with a unilateral conductive hearing loss is mostly conducted to reestablish binaural hearing [43]. Lieu et al. [44] stated that unilateral hearing loss in children has a significant negative effect on their speech language development, and suggest that the delay in speech language development affects their performance in school. Lieu et al. [45] in another research concluded that school-aged children with unilateral hearing loss show increased rates of grade failures, needed additional educational assistance, and perceived behavioral issues in classroom. This suggests that it is beneficial for a children with unilateral hearing loss to wear their BCD in classroom. In a study by de Wolf et al. [46], it was stated that a BCD could be particularly beneficial for children with unilateral hearing loss in the school environment; the exact mechanism especially about binaural hearing and cross hearing were not completely objectified yet.

In case of bilateral microtia, a different surgical scheme for autologous auricular reconstruction is preferable. In our best knowledge, only one article describes the management of surgery in these patients [47]. We suggest to harvest bilateral rib cartilage simultaneously and manufacture both the auricles in the first setting. Rib harvesting is the most painful part of the operation, postoperatively. The advantage of simultaneous harvesting for the children is that is only once, instead of twice, this postoperative pain is endured. However, we reconstruct one ear at a time so that the patient can lie down on one side, except only on their back during the recovery period. One of the reconstructed auricles is positioned at the correct location on one side. The other reconstructed auricle is placed back under the skin near the rib, under the skin of the thorax, for being transplanted to the correct location during the second operation. During the third and fourth operation, the second modeling phase of each auricle separately takes place.

It is important to share our experience in treating patients with an absent auricle per different end point. Hopefully this study will enable other hospitals to review their way of organizing healthcare around patients with an absent auricle, and creating awareness, centralization of care; thus, enhancing the quality of healthcare for these patients.

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