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OTOLOGY

The diagnosis and treatment of a variant type of auricular sinus: postauricular sinus

Jiaqi Pang[1,2] · Hao Xiong[1,2,3] · Qingyun Liao[4] · Yaodong Xu[1,2,3] · Qiuhong Huang[1,2] · Yongkang Ou[1,2,3]

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Abstract

Objectives To make otolaryngologists aware of the variant types of auricular sinus, we have performed a systematic review of patient diagnoses and presented our operative experiences.

Methods From 2009 to 2013 in Sun Yat-Sen Memorial Hospital, there was a total of 20 children with the variant type of auricular sinuses including the comprehensive group. Postauricular sinuses have pits located posterior to the imaginary vertical line that is tangent to the external auditory canal. Sinuses that penetrate the cartilage and cause postauricular swelling or skin defects characterize type 1 of the variant type, while sinuses that adhere to the cartilage and cause preauricular or auricular swelling or skin defects characterize type 2. Patients with pits both anterior to and posterior to the imaginary vertical line comprise the comprehensive group. The patients who had infected underwent auricular sinusectomy using a dual approach, with accurate fistula tracing and proper cartilage removal.

Results Sixteen children who had infected sinus underwent surgery, while the other four were asymptomatic. Ten children (62.5%) of 16 patients were diagnosed as type 1 of the variant type, 2 (12.5%) as type 2. Four children (25%) were diagnosis as the comprehensive group. The asymptomatic could not be defined as the sinuses location were unknown. Sixteen children (100%) of 16 patients who underwent surgery had a history of misdiagnosis and treatment. These patients did not experience recurrence over a 5-year follow-up period.

Conclusion The locations of pits and sinuses help to categorize the different types of auricular sinus. The effective method that we have described should be considered a viable way to reduce recurrence.

Keywords Anomalies · Auricular sinus · Postauricular sinus · Diagnosis · Excision

Introduction

Preauricular sinus is often first diagnosed during childhood because it is a common congenital malformation that easily becomes infected in children. The incidence ranges from 0.1 to 10%, depending on the area and population type. Preauricular sinuses are more common among Africans and Asians than among Caucasians, depending on their health conditions [1–3]. The formation of a preauricular sinus occurs during embryogenesis and is closely associated with the development of the auricle; it commonly contributes to the variability in the location of the anterior margin of the ascending limb of the helix. However, some fistular pits are open along the superior–posterior margin of the helix, the tragus, and even the lobule [4]. Choi et al. [1] divided patients with preauricular sinuses into a classical group and a variant group based on the location of the pits relative to an imaginary line connecting the tragus and the posterior

Jiaqi Pang and Hao Xiong contributed equally to this work.

✉ Yongkang Ou
ouyk@mail.sysu.edu.cn

1 Department of Otolaryngology, Sun Yat-Sen Memorial Hospital, Sun Yat-Sen University, 107 West Yan Jiang Road, Guangzhou 510120, China

2 Institute of Hearing and Speech-Language Science, Sun Yat-Sen University, Guangzhou 510120, China

3 Department of Hearing and Speech Science, Xinhua College, Sun Yat-Sen University, Guangzhou, China

4 Department of Otolaryngology, The First Affiliated Hospital of the University of South China, Hengyang 421000, China

margin of the ascending limb of the helix. The recurrence rate of the classical group can reach 42% [5], and the variant type has a greater recurrence rate.

Infection of the preauricular sinus occurs more often in children than in any other age group. Pain and recurrence have a deep psychological effect on children. Numerous operations on the auricle can to some degree affect the appearance of the ears, which is important to children. Currently, there is lack of information on the postauricular variant of auricular sinus in children. In this study, we summarize the clinical manifestations of different types of auricular sinuses and present our experiences in the management of postauricular sinuses in children, including a systematic discussion of their diagnosis and a summary of the surgical techniques used for the variant type.

Patients and methods

Patients

The institutional review board of Sun Yat-Sen Memorial Hospital approved this study. Between January 2009 and September 2013, a total of 20 children were diagnosed with postauricular sinuses. The clinical charts of these children were retrospectively reviewed to obtain common information and clinical manifestations.

Patients classification

The classical type of postauricular sinuses are both pits and sinuses located anterior to the imaginary vertical line that is tangent to the external auditory canal and characterize the preauricular sinuses. Patients with this single type were excluded in our study. The variant type of auricular sinuses can be clarified by the location of the pits and the sinuses. The variant type are pits posterior to the imaginary vertical line and characterize the postauricular sinuses. According to the location of the sinuses, the postauricular sinuses could be classified into two types. Sinuses that penetrate the cartilage and cause postauricular swelling or skin defects characterize type 1 of the postauricular sinuses, while sinuses that adhere to the cartilage and cause preauricular or auricular swelling or skin defects characterize type 2 of the postauricular sinuses. Patients with pits both anterior to and posterior to the imaginary vertical line comprise the comprehensive group (Fig. 1). All patients underwent ear endoscopy before surgery to exclude fistulas inside the external auditory canal.

Surgical approaches for the postauricular sinuses

After anesthesia, the auricle, especially the area surrounding the sinus, was infiltrated with 2% xylocaine/epinephrine (1:100,000) to reduce bleeding. A perpendicular oval-shaped incision that included the opening of the sinus was made (Fig. 2a). For the type 1 of the postauricular sinuses, the route of the sinus tract to the cartilage was determined. Removed the sinuses and the cartilage around them. For the type 2 of the postauricular sinuses, sinus tissue that may be adherent to the cartilage. We scratched the cartilage or removed some of it. The swelling or skin defects that remained after treatment during the acute infection period were identified (Fig. 2b). The historical incision could be used as a marker for the operative incision in the retroauricular or preauricular area. A perpendicular oval-shaped incision (Fig. 2c) that included the area of swelling was made. The excision was preceded by identifying the inflamed and granulated tissue in the sinus. Usually, the sinus tract communicated between the pit and the swelling or skin defect and could be identified using a lacrimal probe (Fig. 2d). The tract and the cyst were resected as a whole and removed through the postauricular incision (Fig. 2e). Primary closure was performed and examined the completeness of the sinuses (Fig. 2f). The excision of the comprehensive group depended on the region of the infection to remove each sinuses.

Histopathologic studies confirmed the diagnosis of auricular sinuses because the sinus tracts were lined with chronic inflammatory squamous epithelium. The children were followed up yearly after their surgeries.

Ethical considerations

This body of work was approved by the authors of this study. As a retrospective study of case series, when they followed up, we asked the agreement from the patients' parents of the variant type and the comprehensive type to publish the data and some photos. This study did not influence patient follow-up and treatment.

Results

There are 20 children who were diagnosed with postauricular sinuses including the comprehensive group. Sixteen children who had infected underwent surgery, while the other four were asymptomatic. Ten children (62.5%) of sixteen patients were diagnosed as type 1 of the variant type, two (12.5%) as type 2. Four children (25%) were diagnosis as the comprehensive group. The asymptomatic could not be defined as the sinuses location were unknown. The male-to-female ratios of the variant and the comprehensive groups were 8:8 and 2:2, respectively. The average age of the children was 9.7 years (range 3.5–13 years) in the type 1 variant group and 9.2 years (range 7–12 years) in the type 2 variant

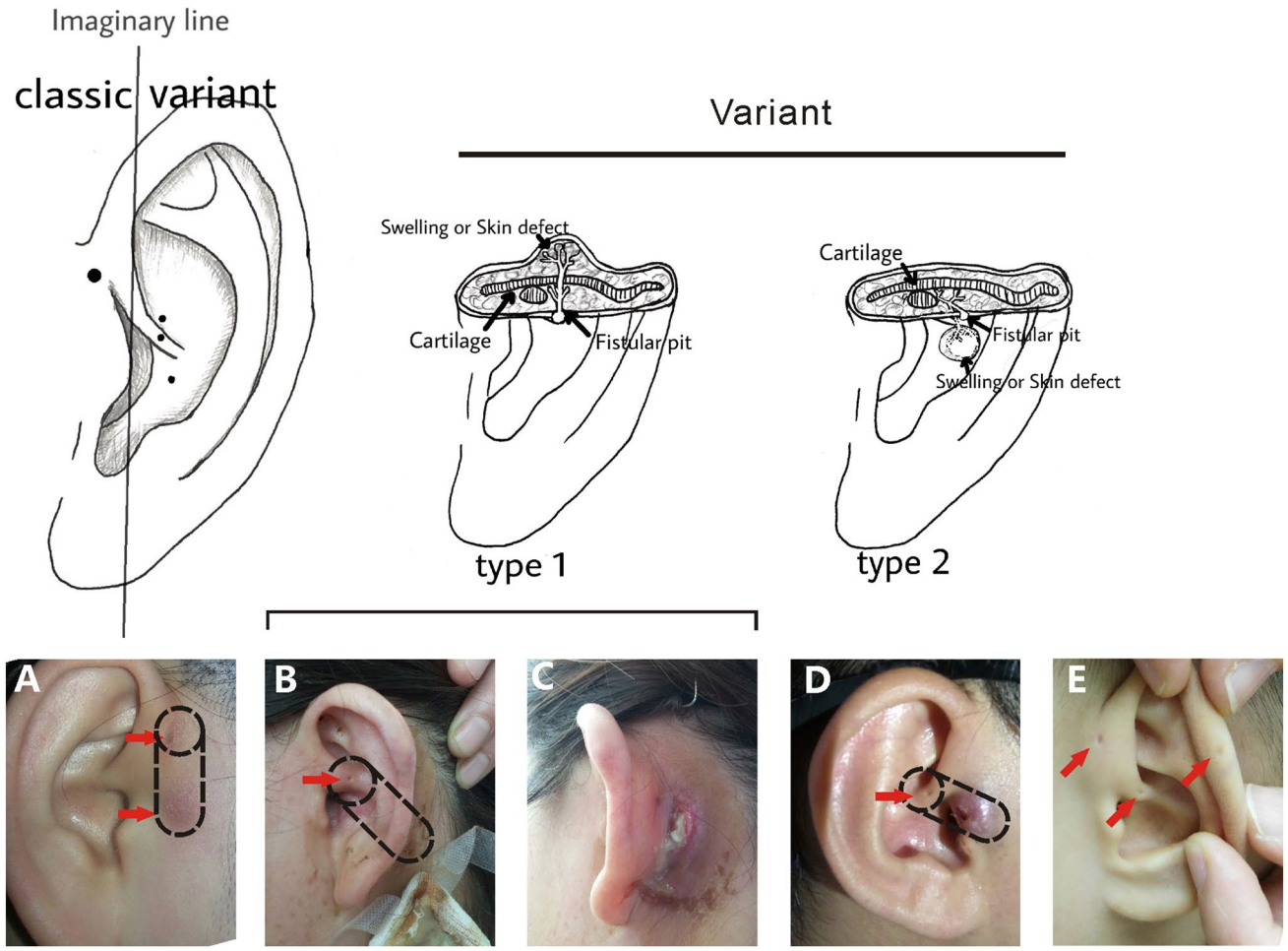

Fig. 1 The schematic drawing shows the differences among the classical, variant and comprehensive types of auricular sinus. The line is an imaginary vertical line that is tangent to the external auditory canal. Classical auricular sinus is characterized pits and sinuses that are anterior to the imaginary vertical line (**a**). Variant auricular sinus is characterized by pits behind the imaginary vertical line. Sinuses penetrating the cartilage and causing postauricular swelling or skin defects characterize type 1 of the variant group (**b**, **c**), while sinuses that adhere to the cartilage and cause preauricular or auricular swelling or skin defects characterize type 2 of the variant group (**d**). The comprehensive group is characterized by the presence of pits both anterior to and behind the imaginary vertical line (**e**)

group, and 8.6 years (range 2–14 years) in the comprehensive group. Sixteen children with the postauricular sinuses reported complaints, which included postauricular swelling (50%), postauricular skin defects (25%) or no symptoms (25%). Postauricular swelling (100%) was the main complaint of the comprehensive group.

Sixteen children with the variant and comprehensive types of sixteen patients who underwent surgery had a history of misdiagnosis and treatment, including eight patients who had been misdiagnosed with lymphadenectasis and underwent drainage for abscess formation, four patients who had been misdiagnosed with sebaceous cysts and underwent an incision, three patients who had been misdiagnosed with mastoiditis and recurrence, and one patient who had been misdiagnosed with classical preauricular sinus and had undergone excisions twice because the surgeon had not paid attention to the branches adhering to the cartilage. Two children experienced suppurative auricular perichondritis (Fig. 1b). One patient had undergone excisions twice and developed serious auricular perichondritis that spread to the entire auricle. Antibiotics, incision, and drainage, which had lasted for two weeks, proved to be ineffective. Thus, after communication with the parents, we decided to excise the infected sinus. Antibiotics and drainage continued three weeks after complete excision. Fortunately, the auricle survived. The other patient underwent excision after the infection was controlled. The demographic data are shown in Table 1.

The children in the variant and comprehensive groups are currently without recurrence at least 5 years after their surgeries (Fig. 3).

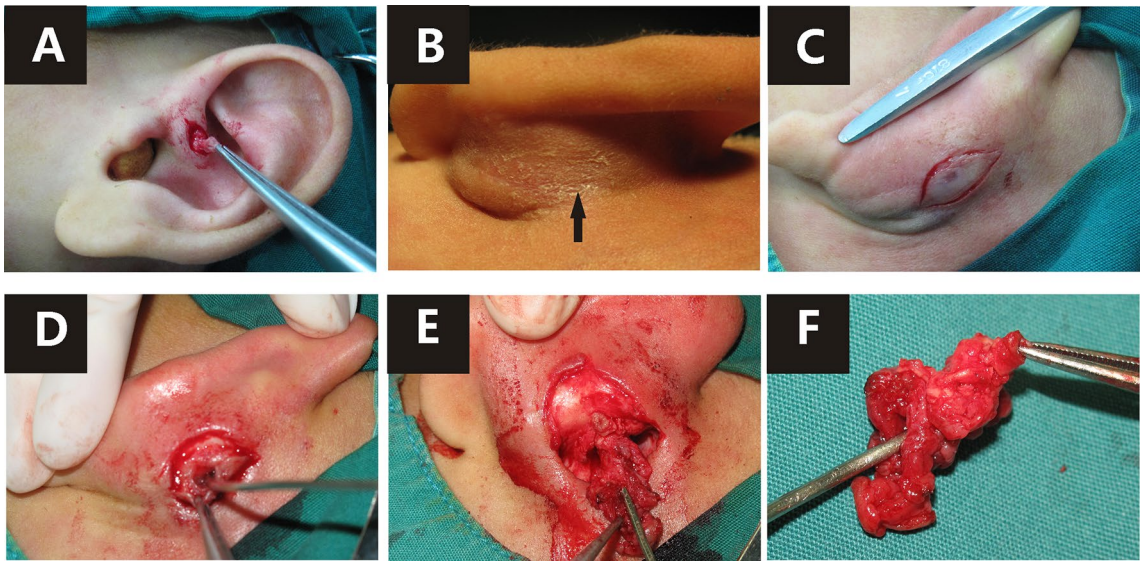

Fig. 2 Surgery. Perpendicular oval-shaped incision including the opening of the sinus (**a**). Skin defects in the retroauricular area, indicated by arrows (**b**). Perpendicular oval-shaped incision in the retroauricular area (**c**). A lacrimal probe in the sinus tract (**d**). Removal of the entire sinus from the retroauricular area (**e**). Examine the completeness of the sinuses (**f**)

Discussion

Preauricular sinuses were first described by Heusinger in 1864. The sinuses are typically first affected during childhood. The openings usually occur along the lateral surface of the helicine crus and the superior–posterior margin of the helix or tragus; sinuses manifest as small pits adjacent to the external ear, usually located at the anterior margin of the ascending limb of the helix. Variant types of auricular sinuses that both pits and sinuses are located posterior to the imaginary vertical line that is tangent to the external auditory canal have rarely been reported [6]. Moreover, in our patients, we found that the sinuses of the variant group may present preauricular, which generally occurs in the classical group. In addition, some patients had sinuses of both the classical and variant type, which has not been mentioned previously. We considered it as the comprehensive group. The complaints of patients with the type 1 variant differ from those of patients with the classical type of preauricular sinuses. Among patients with the variant type, the main complaint is postauricular pain and abscesses. The distance of the pathogenic sites from the pits make them seem irrelevant to the preauricular sinuses, therefore, leading to misdiagnosis. All children with the variant and comprehensive types experienced misdiagnosis, which led to incomplete excisions that had unnecessary psychological effects and increased health costs. Moreover, numerous operations can have a substantial effect on the appearance of the auricle. Two patients even progressed to auricular perichondritis. This progression has not been reported previously. Therefore, otolaryngologists should carefully consider the variant type of preauricular sinus if the patient complains repeatedly of postauricular swelling or infection with a pit on the auricle.

We consider that imaging is not necessarily essential in patients with auricle sinus to exclude other sinuses, however, the aware of the different type of them is. According to careful inquiry and physical examination, we conclude that both the typical and variant auricle sinus are confined around the auricle and they are superficial. Then the patients' complaint helped us to exclude the cervicodynia and sinuses in the neck, as they did not have neck pain and pits. Besides, ear endoscopy is important to figure out whether fistulas inside the external auditory canal or not, and then exclude an anomaly of the first branchial cleft. Although three patients were misdiagnosed as mastoiditis, the swelling of the skin were limited around the auricle instead of on the mastoid zone and finally proved that they were misdiagnosed. Collectively, with care inquiry, physical and ear endoscopy examination, we believe that doctors can diagnose both the typical and variant type and exclude other diseases, hence imaging is not essential.

The variant type is similar with the classic type, moreover, are important. They help you to besides, if the patients complaint about, other sinuses need to be excluded. In our study, the abnormal was around the auricular. The extension of the sinus in the neck have not been reported yet.

Preauricular sinuses require no treatment if they are asymptomatic [7]. However, once infected, antibiotics, incision, and drainage are necessary before the removal of the whole sinus, including its branches. The same treatment principles apply to all three types of preauricular sinuses.

Table 1 Clinical manifestation and management of the variant and comprehensive types of auricular sinus

	Gender	Age (years)	Disease duration (years)	Side	Location of the pit	History of misdiagnosis	Complications		History of treatment	Follow-up
							Postauricular abscesses	Auricular perichondritis		
1	Female	14	5	Left	Type 1 + classical	Sebaceous cyst	−	−	Incision and drainage	No recurrence
2	Male	$10_{4/12}$	1.3	Right	Type 1	Lymphadenectasis	−	−	Incision and drainage	No recurrence (as presented in Fig. 3)
3	Male	$11_{7/12}$	3	Left	Type 1 + classical	Sebaceous cyst	−	−	Incision, drainage and excision twice	No recurrence
4	Female	7	2	Left	Type 2	Sebaceous cyst	−	+	Incision and drainage	No recurrence
5	Male	13	3	Left	Type 1	Lymphadenectasis	+	−	Incision and drainage	No recurrence
6	Female	$10_{3/12}$	4.5	Left	Type 1	Lymphadenectasis	+	−	Incision and drainage	No recurrence
7	Male	14	10	Right	Type 1	Lymphadenectasis	+	+	Incision, drainage and excision twice	No recurrence
8	Female	$3_{6/12}$	0.67	Bilateral	Type 1	Lymphadenectasis	Right −	Right −	Right −	No recurrence
							Left +	Left −	Left: incision and drainage	No recurrence
9	Female	10	0.58	Bilateral	Type 1	Mastoiditis	Right −	Right −	Right −	No recurrence
							Left +	Left −	Incision and drainage	No recurrence
10	Female	$4_{7/12}$	0.75	Right	Type 1	Mastoiditis	+	−	Incision and drainage	No recurrence
11	Male	8	1.5	Right	Type 1	Lymphadenectasis	+	−	Incision and drainage	No recurrence
12	Male	$11_{7/12}$	3 years	Left	Type 2	Sebaceous cyst	+	−	Incision and drainage	No recurrence
13	Female	7	2	Left	Type 1 + classical	Mastoiditis	+	−	Incision and drainage	No recurrence
14	Female	13	3	Bilateral	Type 1	Lymphadenectasis	Right +	Right −	Incision and drainage	No recurrence
15	Female	$10_{3/12}$	4.5	Right	Type 1	Lymphadenectasis	−	−	Incision and drainage	No recurrence
16	Male	2	5	Left	Type 2 + classical	Classical preauricular sinus	−	−	Incision and drainage	No recurrence
	The following patients were not infected									
17	Female	$3_{6/12}$	/	Bilateral	No symptoms	/	/	/	/	/
18	Female	14	/	Left	No symptoms	/	/	/	/	/
19	Male	$4_{7/12}$	/	Right	No symptoms	/	/	/	/	/
20	Male	8	/	Right	No symptoms	/	/	/	/	/

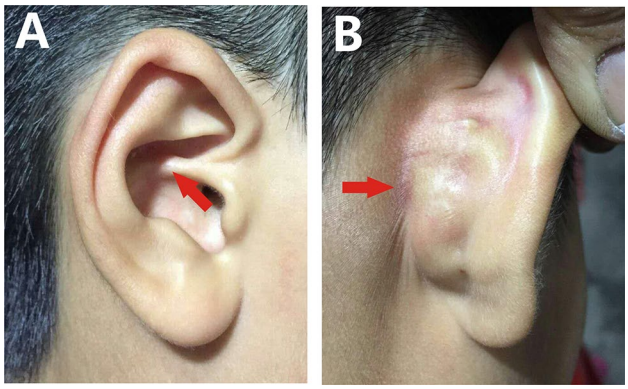

Fig. 3 A child in the variant type 1 group was followed up 3 years after surgery. The arrow shows the opening of the sinus before surgery (**a**). The arrow shows the location of the postauricular sinus (**b**)

If the tract is not completely excised, recurrent acute exacerbations are likely. Some patients have multiple fistulas, i.e., the sinus has multiple branches. According to previous studies, the recurrence rate of the classical type can reach 42%. Therefore, many surgical techniques have evolved for the classical type, all with the aim of reducing the recurrence rate [8–10]. The operation profile of the type 1 variant remains unknown, while surgery for the type 2 variant shares the same principle of cartilage removal using the general technique applied in the classical group, with difference in the treatment of the cartilage. For the comprehensive group, we considered that each pit had its own tract and sinus, although there was no evidence that the tracts were separated. At times, it can be difficult to confirm the origin of the sinus and the tract direction during surgery for the comprehensive group, and this group had a high recurrence of residue. Therefore, we recommend removing all the pits and sinuses once one of them becomes infected.

In our experience, three surgical techniques have been used for the complete excision of the type 1 preauricular variant and the comprehensive group including the variant sinus. First, the location of the operative incisions must be determined. If the pits are preauricular and the swelling is postauricular, the general direction of the tract is an imaginary line between them. For a sinus that penetrates the cartilage, the tract that starts at the pit and progresses to the point of cartilage penetration is too narrow to allow the whole tract to be removed from the front unless the preauricular incision is large enough for exposure. Even the smallest probe cannot be inserted into the tract from the pit. Hence, it is better to dissect the sinus using a dual approach through preauricular and postauricular incisions. The preauricular incision should be as small as possible to maintain aesthetics and reduce the possibility of postoperative auricular deformities. After antibiotics, incision, and drainage or excision, swelling, masses, skin defects, or scars on the retroauricular area, which can last for up to six months, are ideal markers for the postauricular surgical incision as these areas are directly linked to the tract and are not obvious, even if they are extended (Fig. 2b, c).

Second, remnant branches are the reason for recurrence. Visualization of the sinus is the key to a complete excision; however, it is difficult to perform during surgery. In previous studies, visualization of the sinus tract has been aided with the use of a microscope [4, 5] or methylene blue dye instillation [11] and probing of the tract [12]. In the variant type, the sinuses are even more narrow than in the classical type because they are attached to the perichondrium of the anterior area and the cartilage. In addition, the cicatrization that occurs in recurrent cases, such as the patient who developed serious auricular perichondritis and required two excisions, blocks the tract. In this case, good sinus tract visualization was impossible because neither methylene blue nor a probe could delineate the operative field. Therefore, the region should be expanded if necessary. The use of methylene blue dye and a probe raises concerns for some surgeons regarding the potential risk of sinus tract perforation and leakage of methylene blue dye into the operative field. Therefore, we did not perform operations using methylene blue. After the fluid is removed from inside of the cyst, a fine lacrimal probe can be used to explore the directions of tracts from the bottom of the cyst to the pierced auricular cartilage and the preauricular region (Fig. 2d). Tracing from the preauricular incision can be helpful in several cases.

Third, cartilage that the sinus has adhered to or penetrated must be removed. In our opinion, the difference between the classical and the variant type is not only the location of the pits and sinuses but also the relationship with the auricular cartilage. The lack of awareness of the importance of the relationship with the auricular cartilage leads to high recurrence. The histological relationship of the preauricular sinuses was determined in a study by Dunham et al. [13], which found that the average sinocartilaginous distance was 425 μm. In more than 50% of the specimens reviewed, the sinocartilaginous distance was less than 0.5 mm, and in nearly all of these cases, the epithelial tract was continuous with the stromal tissue, which was histologically indistinguishable from the perichondrium. In the variant group of preauricular sinuses in which the tract was attached to the perichondrium and cartilage, the close relationship of the sinus tract to the cartilage suggested that it may be difficult to dissect the tract completely from the cartilage without removing some of the cartilage; furthermore, this may be more difficult in variant types in which the tract penetrates or adheres to the cartilage. Therefore, it is important to remove the cartilage that the sinus has adhered to or penetrated.

Although an anomaly of the first branchial cleft and the variant type of preauricular sinuses have a similar clinical presentation, except for the location of the swelling.

Therefore, we underwent ear endoscopy before surgery to exclude fistulas inside the external auditory canal. Surgeons do not need to be concerned about injury to the facial nerve during the incision. Unlike anomalies of the first branchial cleft arising from incomplete closure of the ectodermal portion of the first branchial cleft, preauricular sinuses originate from incomplete fusion of the hillocks or entrapment of the ectodermal epithelium during auricular development. Thus, the sinuses are limited to the area of the auricle and are distant from the facial nerve unless severe inflammation affects the facial nerve area. In such cases, antibiotics should be used.

Conclusion

Otolaryngologists should be aware of the variant type of auricular sinus. Careful examination is obligatory for patients with repeated postauricular swelling or infection and a pit on the auricle, characteristics that indicate the presence of the variant and comprehensive types of auricular sinus. The locations of pits and sinuses help to categorize the different type of auricular sinus. The dual approach through the pit and the sinus incisions, appropriate tracing of the fistula and proper removal of the cartilage substantially aid in the complete excision of the sinuses and reduce the risk of recurrence.

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Compliance with ethical standards

Conflict of interest The authors have no conflict of interests.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from all individual participants included in the study.

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