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## ORIGINAL RESEARCH

# A Facility-based Study of Preauricular Sinus Among Adults in Southwest Nigeria

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## Abstract

**Background:** Preauricular sinus (PAS) is common among the blacks and it presents in different manners. The perceptions of adult patients with PAS may influence the management strategies deployed.

**Objectives:** To determine the perceptions of adult patients who have PAS about the disorder and describe the modalities of presentation.

**Methods:** A descriptive study was conducted at the ENT Clinics over nine months. Using a questionnaire, socio-demographic information, perception of patients on the cause, concerns, modality of treatment, inclination to surgery and perceived complications of surgery were explored. Examination findings of the sinuses, including results of audiological investigations were recorded.

**Results:** The prevalence of PAS was 7.4% (56/754) and the mean age of the subjects was 43.9±10.6 years. Multiple causes of PAS were perceived by 58.5% of the patients, 13.2% were not concerned but intermittent symptom was experienced by 43.3% of the patients. More than half (52.8%) believed the sinus could be treated medically with drugs, 86.8% perceived complications from surgical excision while 73.6% abhorred surgical excision. Half of the PAS was in the left ear while 11.3% were bilateral. Audiological evaluation revealed 46.4% of patients had evidence of sensorineural hearing loss while 56.5% had abnormal middle ear functions.

**Conclusions:** Some adult patients had wrong perceptions of PAS and most patients abhorred surgical excision. The clinical presentations were mostly of the classical type, with almost two-thirds violated. There may be a need for genetic studies on PAS in future research.

**Keywords:** Abscess, Ear, Perceptions, Preauricular sinus, Pure Tone Audiometry, Tympanometry.

## Introduction

Preauricular Sinus (PAS) is a congenital anomaly resulting from the poor fusion of the branchial arches 1 and 2 during embryological development. During embryogenesis, three auricular hillocks (of His) arise from the caudal

border of the first branchial arch, and three arise from the cephalic border of the second branchial arch. These hillocks should unite during the next few weeks of embryogenesis. Preauricular sinus occurs when there is incomplete fusion of

these hillocks. [1] PAS can occur sporadically or may be genetic, exhibiting incomplete autosomal dominant genetic pattern. [2] It presents mainly as a small hole anterior to the upper limb of the helix, although some may not present in this classical manner. PAS occasionally occurs with other congenital malformations in the branchial apparatus and the renal system as a component of a syndrome. [2] Therefore, the characterization of PAS is important.

The incidence of PAS in the general population has been estimated to be 0.1% to 0.9% in the United States of America, 0.9% in England, 1.6% to 2.5% in Taiwan, and 4% to 10% in some parts of Africa. [3] PAS is arguably more common in the local population of blacks of African ancestry. While many cases of PAS are asymptomatic, a few presents with symptoms such as watery or offensive effluents from the sinus, either spontaneously or on expression. PAS sometimes develop complications, especially recurrent episodes of infection and may become swollen, painful, and exude pus. [4] Others leave ulcers and scars over the surface of the sinus. The mode of presentation may influence the choice of management.

Provided PAS remains asymptomatic or with minor symptoms, interventions may not be required while symptomatic cases are treated as feasible. Sometimes an abscess may develop after recurrent infections. PAS abscess is commonly mistaken for pimples, furunculosis, chronic infection such as tuberculosis and mycosis, and congenital conditions such as dermoids and sebaceous cysts. [4] The management of recurrent PAS infections and abscess may be problematic, and it is the main indication for surgical drainage and excision. However, some patients will prefer excision of the sinus for cosmetic reasons, despite being asymptomatic. Consequently, the decision of the modality of treatment is primarily patient-centred. Such decisions may be influenced by the perception of the patients concerning the sinus as

well as some myths. One of such myths is that PAS is caused by some offensive actions of a woman during pregnancy or by a spiritual attack. [5]

Most studies on PAS had focused on the problems associated with the surgical approaches, and complications accompanying infections and abscess. [6 - 8] Few studies had reported the prevalence and methods of presentation of PAS in the Nigerian population. [9,10] However, the perceptions of the patients with PAS concerning the lesion have scarcely been explored. This study aimed to determine the perceptions of adult patients who have the PAS concerning the lesion. The study also described the various modalities of clinical presentation. The information gathered could give an insight into the patients' reasons behind their decisions concerning the management of PAS.

## **Methods**

This was a descriptive study involving adult patients attending the Ear Nose and Throat Clinic of Olabisi Onabanjo University Teaching Hospital (OOUTH) from 1<sup>st</sup> January to 30<sup>th</sup> September 2020. The eligible patients were those with the preauricular sinus.

All patients who presented in the clinics for a first-time consultation and diagnosed to have PAS were recruited into the study. Purposive sampling was used. Excluded from the study were patients with significant hearing impairment who found it difficult to communicate, patients with mental retardation, and those with Down's syndrome. Patients who had other previous otologic surgery involving the external or the middle ear cavities were also excluded.

The study instrument was an interviewer-administered, purpose-designed questionnaire which was administered separately from the

consultation of the subjects. The questionnaire was pre-tested at the Sacred Heart Hospital, Abeokuta, on similar subjects. The questionnaire was used to obtain information on socio-demography (age, sex and religion), and perception of the subjects on the cause, concerns and symptoms experienced, possible modalities of treatment, inclination towards surgical excision and perceived complications that may arise from surgical excision of the sinus. The subjects had their ears examined as part of routine examination irrespective of their complaints. Particular details were made of the pinna, the preauricular sinus location, and physical condition, the external auditory canal, the tympanic membrane and the functioning of the facial nerve.

Some patients had basic audiological investigations, namely Pure Tone Audiometry (PTA) and Tympanometry as necessary, and the results were recorded. PTA was conducted on patients who had no otological symptoms and no clinical evidence of hearing loss while tympanometry was conducted only on patients that had intact (unviolated) tympanic membranes.

*Data analysis*

The data were entered into a computer programme. Data analysis was done using the Statistical Package for Social Sciences, SPSS version 21 (Chicago, IL), and the data was presented in descriptive formats in tabular forms.

*Ethical considerations*

Ethical approval to conduct the study was obtained from OOUTH Health Research Ethics Committee (HREC) with approval number OOUTH/HREC/340/2020AP. Informed consent was taken from each patient before the administration of the questionnaire.

**Results**

Seven hundred and fifty-four adult patients attended the clinics during the study period; 56 of these were diagnosed with PAS, giving a prevalence rate of 7.4%. Three of the 56 patients were excluded, leaving a final sample size of 53 patients. The socio-demographic characteristics of the patients are shown in Table I. The age range was 19-59 years, with a mean age of 43.9±10.6 years. There were 21 males and 32 females giving a male-to-female ratio of 1:1.5. Equal proportions of the patients practised Christianity and Islam.

**Table I: Socio-demographic characteristics of the 53 patients**

<i>Characteristics</i>	<i>Frequency</i>	<i>Percentage</i>
<b>Age range (Years)</b>		
18-30	22	41.5
31-45	23	43.4
46-60	8	15.1
<b>Sex</b>		
Male	21	39.6
Female	32	60.4
<b>Religion</b>		
Christianity	27	50.9
Islam	26	49.1

Table II depicts the perceptions of the patients concerning PAS. More than half (58.5%) of the patients felt there was more than one cause of PAS; notable perceived causes included spiritual forces (24; 45.3%) and birth injury (18; 34.0%). Notable myths in the responses of the patients included the tendency of individuals with PAS to be unruly or difficult (26; 49.1%) and the sinus could cause hearing impairment (20; 37.7%).

While 13.2% of the patients were not bothered nor concerned about the sinus, others were concerned about symptoms such as intermittent swelling, discharges and pain at the sinus. More than half (52.8%) perceived that PAS could be cured with medications while 86.8% perceived complications could arise from surgical excision of the sinus. Almost three-quarter (73.6%) of the patients were negatively inclined to surgical excision of the preauricular sinus.

**Table II: Perceptions of patients concerning Preauricular sinus**

<i>Parameters</i>	<i>Frequency</i>	<i>Percentage</i>
<b>Causes</b>		
Not known	2	3.8
Birth injury	18	34.0
Spiritual forces	24	45.3
Multiple causes	31	58.5
<b>Concerns and symptoms</b>		
Intermittent swelling	18	34.0
Discharge	14	26.4
Intermittent pain	8	15.1
None	7	13.2
Shameful	6	11.3
<b>Perceived modality of treatment</b>		
Medications	28	52.8
Surgical excision	25	47.2
<b>Perceived complication of Surgery</b>		
Not known	7	13.2
Hypertrophic scar/Keloid	9	17.0
Non-healing wound	11	20.8
Recurrent discharge	12	22.6
Recurrence	14	26.4
<b>Inclined to surgical excision</b>		
Yes	14	26.4
No	39	73.6

The spectrum of the clinical presentation of PAS is shown in Table III. Thirty (56.7%) patients had not noticed any symptom attributable to the sinus, while 23 (43.4%) patients claimed they had previous symptoms, notably swelling and discharge of the sinus. Physical examination revealed majority 47/53 (88.7%) had unilateral PAS with half of the sinuses (50.9%) being on the left side while 11.3% had it in both ears. More

than a third (35.6%) had the sinus intact and not violated, while others appeared violated in one form or the other. Three of the sinuses had developed abscesses. Some of the patients had other abnormalities in the external ear including preauricular skin tags on the pinna in 3, nodular attachment around the sinus in 2, multiple sinus openings in one ear, bifid tragus and lobule, and

undue angulation and stenosis of the external auditory canals.

Twenty-eight patients who had no otological symptoms had PTA, and 13 (46.4%) of these had evidence of sensorineural hearing loss in the

affected ear. Tympanometry was performed to assess the middle ear function in 23 patients, 10 (43.5%) of which had normal middle ear functions.

**Table III: Presentation of Preauricular sinus**

<i>Characteristics</i>	<i>Frequency</i>	<i>Percentage</i>
<b>Symptoms</b>		
Absent	30	56.7
Present	23	43.3
<b>Laterality of sinus</b>		
Left side	27	50.9
Right side	20	39.7
Bilateral	6	11.3
<b>Physical findings (n = 59)</b>		
Intact/Not violated	21	35.6
Scarred/Fibrosed	18	30.6
Infected	13	22.0
Ulcerated	7	11.9
<b>Other abnormalities (n=16)</b>		
Pinna	10	62.5
External auditory canal	6	37.5
<b>Pure Tone Audiometry (n = 28)</b>		
Normal hearing	15	53.6
Sensorineural hearing loss	13	46.4
<b>Tympanometry (n = 23)</b>		
Type A (Normal)	10	43.5
Type B (Flat)	3	13.0
Type C	5	21.7
Type As	5	21.7

**Discussion**

PAS has possibly remained in the medical literature for the clinical implications of its syndromic links and the management problems when it becomes complicated. The management problems may be confounded by the perceptions of individuals with PAS. Exploration of the perceptions of those who have the sinus may give a better picture of its overall clinical significance. The outcome of management may also be influenced by the perceptions and attitudes of individuals with the lesion. Most of the patients

perceived PAS as a minor congenital abnormality which is generally innocuous but may become noticeable with occasional swellings and discharges. Some (58.5%) believed there were other causes of PAS aside from being a congenital abnormality. The notable beliefs included association with the delivery of the child such as an accidental needle prick during a caesarean section while others assumed that there were extraneous or spiritual forces associated with the sinus. Few myths associated with the PAS included that children with the sinus tend to be unruly, and others perceive it to be a cause rather

than an association with hearing impairment. Lasisi and Ajuwon, [11] had previously reported some beliefs and myths of ear, nose and throat-related conditions among residents in Ibadan, Nigeria.

A large proportion (86.8%) of the respondents were concerned or disturbed with the intermittent swelling and discharge of the PAS, regarding these as ominous signs, which require aggressive intervention or treatment. The perception held by 52.8% of the patients that PAS could be cured with medications was wrong and needs to be corrected. Although all the respondents agreed that surgery was treatment modality, most of them had a negative inclination and attitude to have their sinuses excised surgically, in 73.6% of the respondents. This aversion to surgery could be predicated upon the finding that the majority of the respondents were aware of the common complications that could arise from surgical excision. However, none of the respondents was aware of the possibility of facial paresis or palsy complicating excision. The suggestion of facial deviation elicited responses ranging from disbelief to fear in many of the patients. Therefore, there were some wrong perceptions among adults concerning the cause and treatment of PAS

The prevalence rate of PAS among adults (7.4%) in this study was within the general range reported in the population of Nigerians. [9] The relatively high prevalence rate of PAS in the native African population had been previously noted. [3] The rate could have been higher if children were included in this study. Children were excluded from the study because it would be difficult to obtain their correct perceptions of the subject. Most of the patients in the present study were mature adults with the mean age in this study being 34.9 years. The mean age of persons with PAS reported in Ekiti, South-western Nigeria was 36.2 years. [12]

PAS could occur either as a sporadic or an inherited disorder. Inheritance is characterized by an incomplete autosomal dominant pattern with reduced penetrance and variable expression. [13] In bilateral occurrence, the likelihood of being inherited is increased in 25-50% of cases. About 3-10% of cases of PAS occurs as part of syndromic features, primarily in association with deafness and branchio-oto-renal (BOR) syndrome. [13] Sampath *et al.*, [14] reported a novel autosomal dominant disorder with variable phenotypic expression in a three-generation family; this distinctive clinical phenotype and linkage to a novel locus at 14q31 supported the existence of a new syndrome of the branchial cleft comprising hypertelorism, preauricular sinus, deafness, and punctal pits with lacrimal-duct obstruction. Therefore, there is a need for genomic studies of PAS, especially in the African population where the anomaly is disproportionately common.

While PAS has classically been described to present as an opening at the anterior end of the ascending limb of the helix, variations in the ways and manners of presentation sometimes occur. Some variants had been reported as case presentations or case series in the medical literature. These include cases of PAS with postauricular extension, [15] fistula opening below the intertragic notch, [16] and infra-auricular sinus. [17] Three of the patients in the present study had preauricular skin tags, while two had nodular attachment around the sinus which were regarded as variants of PAS in this study. Deshpande *et al.*, [18] had previously reported that preauricular skin tags are the most common variants associated with the PAS in the United Kingdom.

Varying reports concerning sex distribution in PAS had been noted. The finding of female preponderance (60.4%) in the present study is similar to the finding of Lee *at al.*, [19] in South Korea. Inconsistencies are also noted regarding

the laterality of the sinus. While it has been acknowledged that the unilateral occurrence of the sinus is more common, [20] there is no consensus about the side where it is more common. In the present study, PAS was common in the left ear while 11.3% were bilateral, similar to the report from in Ilorin, north-central Nigeria. [9] Close to two-thirds of the PAS examined (64.4%) had been violated by infections, inflammations, and trauma. While the bulk of these remained asymptomatic, there were three that had developed abscesses. An abscess usually results from recurrent infections within the sinus cavity with the closure of the punctum, such that the pus cannot escape through the sinus opening. Considering that PAS abscess has been reported to occur predominantly in children, [21,22] the prevalence rate of 5.1% for such among adults in the present study is relatively high. The management of PAS abscess is surgical and may be challenging because the sinus must be dissected *en-bloc* to avoid leaving a remnant which can predispose to a recurrence.

In the present study, the syndromic linkage between other congenital anomalies and PAS was speculated. Few pinna abnormalities, including bifid tragus and lobule, nodules on the helix, were the abnormalities found in the pinna of adults with PAS while undue angulation was the main abnormality found in the external auditory canals. These minor abnormalities that had not been previously shown to be associated with PAS and their significance could not be ascertained. Some congenital anomalies reportedly associated with the first brachial arch irregularities include external auditory canal atresia, stenosis, [23] and cholesteatoma. [24]

PAS can occur as part of branchial arch anomalies (branchial cysts, branchial fistulas), with hearing loss and renal anomalies constituting the branchio-oto-renal (BOR) syndrome. [25] Some patients had their hearing level assessment done with pure tone audiometry (PTA) revealing mild

sensorineural hearing loss (SNHL) in 46.4%. These results should be interpreted cautiously because the general level of hearing in the environment is relatively low (high threshold) in adults, and presbycusis starts earlier at about the fifth decade in this setting. [26] Middle ear analyses with tympanometry produced abnormal tympanograms in 56.4% of the ears. Non-specific middle ear malformations have been reported to be associated with the first branchial arch anomaly. [27] However, the middle ear abnormalities found in the present study could not be unequivocally linked with the PAS. Screening for renal abnormalities in branchio-oto-renal (BOR) syndrome carries stringent conditions. A renal ultrasound should be performed in patients with isolated preauricular pits, cup ears, or any other ear anomaly accompanied by one or more of the following: other malformations or dysmorphic features, a family history of deafness, auricular and/or renal malformations, or a maternal history of gestational diabetes. In the absence of these findings, renal ultrasonography is not indicated. [28] Therefore, routine renal imaging is not warranted in patients with minor external ear anomalies unless accompanied by other systemic malformations. [20] None of the patients in the present study met the above criteria for renal ultrasonography so they were technically not screened for this syndrome. This is regarded as a limitation to the study. Another limitation of this study is that by its design, the obtained prevalence rate could not represent the population, nor the community. The perceptions could be different within the community especially with people who have no PAS. Therefore, the necessity of a community-based study which will clarify some of the findings in the present study is entertained.

## Conclusion

The prevalence rate of PAS was 7.4% in adult patients, and most patients abhorred surgical excision of the sinus. The clinical presentations were mostly of the classical PAS type with close to two thirds already violated at presentation. There were some wrong perceptions of the PAS among adults. There is a need for genetic studies in future research on the subject.

**Authors' Contributions:** OAS conceptualized and designed the study, collected and analysed data and drafted the manuscript. EAO participated in the study design, data collection and manuscript review for sound intellectual content. Both authors approved the final version of the manuscript.

**Conflict of Interest:** None.

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