# Preauricular fistulae: not always an obvious diagnosis

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**Abstract**

Preauricular fistulae (PAF) are the most common congenital fistulae. Diagnosis could be challenging. They could be isolated or be a part of malformative syndromes. We report a 3-case series of PAF.

**Key words:** preauricular, fistula, congenital

# Introduction:

Congenital cysts and fistulae of the face are poorly understood embryological malformations. Dermatologists, pediatricians and otolaryngologists must recognize these lesions early for proper management. They can be explained by the persistence of an anatomical component, normally transitory during the formation of the embryo in the second month of life in utero.

Preauricular fistulae (PAF) are the most common congenital fistulae. They are located a few millimeters in front of the root of the helix. They could be diagnosed at birth by the presence of a swelling or an external opening, or later in childhood at the occasion of repeated superinfections or in the presence of an isolated swelling (1). Surgical treatment is indicated in case of symptomatic cases, remote from the infectious episode.

We describe three cases of PAF and we emphasize on its clinical aspects with a literature review concerning associated anomalies and therapeutic modalities.

# Observations :

**Case 1:** M, 8 years old, with a family history of cervical lymph node tuberculosis (TB) in the mother, presented with painful tumor-like lesion of the upper right preauricular region over the 6 previous months which had gradually increased in size. On examination, we have noted a 3cm-diameter non-infiltrating exophytic formation with an erythematous-crusty surface and pus drainage under pressure. It was located above the right pretragal region (Figure 1). Further examination showed a small and firm 3 mm-diameter flesh coloured nodule on the pretragal region suggestive of an accessory tragus. ENT examination and facial MRI did not show a fistula. Based on family history, consumption of raw milk and histology findings (epithelioid and gigantocellular granulomas without caseous necrosis), we made an initial diagnosis of primary cutaneous TB. The patient was put on anti-TB treatment for 6 months. There was clinical improvement but a fistula was noted after swelling disappearance (Figure 2). We finally made diagnosis of PAF complicated by a pyogenic granuloma. The patient underwent fistula removal surgery with good outcome (Figure 3).

**Case 2:** F, 6 years old, with no particular history, was referred to our department for a 4-year history swelling of the upper preauricular region. On examination, there was a 1cm-nodule with erythemato-telangiectatic surface and viscous liquid drainage under pressure. It was located 1 cm anterior to a small opening of the left helix root (Figure 4). Physical examination was normal. We made diagnosis of PAF and the patient was referred to ENT department for further care.

**Case 3:** B, 2 years old, presented with a congenital malformation of the right upper preauricular region. On examination, there was a purple nodular lesion 1.5 cm in size with impetiginized surface and an opening leaking pus under pressure. It was located in front of the right helix root (Figure 5). Physical examination was normal. We made diagnosis of PAF complicated by superinfection. The patient was put on antibiotic therapy and then referred to ENT department for surgery.

# Discussion :

PAF are common congenital fistulae. Their prevalence varies according to ethnic origin from 0.1% to 0.9% in the United States, 0.9% in England and from 4% to 10% in Africa (2,3). Their prevalence remains unknown in Tunisia.

PAF formation occurs during embryogenesis when the pinna of the ear (auricle) is being developed at the sixth week of gestation. The auricle is formed from six mesenchymal buds (colliculi of His) which develop from the first two branchial arches, on the edges of the first branchial cleft. PAF would result from an abnormal fusion of the three anterior buds (4).

PAF is located almost exclusively anterior to the root of the helix, above the tragus superior edge. It is most often unilateral with a predominance on the right side according to the literature, as was the case in two of our patients (3). It can be bilateral (25 to 50%) and, in this case, more frequently familial in autosomal dominant pattern of inheritance with variable penetrance and expressiveness (2,3).

PAF is symptomatic in only 25% of cases. Diagnosis is made at birth if the fistulous orifice is noted but can be made late in childhood or in young adults with oozing of yellowish liquid and epidermal debris or during superinfection with purulent discharge or pre-auricular abscess. When obstructed, an inflammatory retention nodule (cases 2 and 3) or an extensive granulomatous or even pseudotumoral lesion, simulating actinomycosis or cutaneous TB (case 1) could be noted. Hence the importance of monitoring and re-examining patients consulting for lesions of the preauricular region after disappearance of inflammatory signs.

This malformation should not be confused with first branchial cleft fistulae, typically located on the anterior neck near the angle of the mandible.

PAF is most often isolated (cases 2 and 3). However, it can also be part of malformative syndromes including branchio-oto-renal syndrome with structural anomalies of the ear (external, middle, internal), loss of hearing, renal malformations, laterocervical cysts and fistulae and/or tear duct fistulae. The association with abnormalities of the branchial arches can reach up to 60% of cases in the literature (6). In our series, only one patient (case 1) had a PAF associated with an accessory tragus. It should be noted that in its isolated form, the risk of hearing abnormalities or kidney malformation does not seem to be higher than in the general population (1.7% and 2.6% respectively according to a Chinese study) (2). It is classic to recommend ENT examination including otoscopy and audiogram to look for minor aplasia of the ear or deafness. Renal ultrasound is only recommended in the presence of one or more abnormalities: 1- other malformation or dysmorphic character; 2- family history of deafness; 3- family history of ear or kidney malformations; 4- maternal history of gestational diabetes (2).

If no associated anomaly is identified and in asymptomatic forms, no treatment is recommended (3). Surgery is relatively not difficult but must be rigorous because recurrence is not exceptional, especially if the reference technique is not respected. It is only indicated in the presence of secondary infections or abscesses. In case of abscess, we must first perform a puncture- aspiration with antibiotic therapy and perform surgery ideally in the following 2 to 3 weeks (7). The recurrence rate is high (up to 40%) if surgery is limited to an incision around the fistula and contact dissection. The recommended technique is to make an elliptical incision around the fistula, dissect the soft tissues to excise an ellipse of cartilage, deep to the apex of the fistula, where the fistula tissue adheres to the cartilage. Cartilaginous suture is then made to minimize the dead space responsible for postoperative complications and long-term recurrences (8,9).

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**Legends**

Figure 1: Budding non-infiltrating exophytic formation of the upper right preauricular region

Figure 2: Outcome after 6 months of anti-tuberculosis treatment. Note the preauricular fistula

Figure 3: Outcome after surgery

Figure 4: Nodular lesion with erythemato-telangiectatic surface located 1 cm anterior to a small opening of the left helix root

Figure 5: Impetiginized purple nodular lesion centered by a small opening releasing pus under pressure located in front of the root of the helix

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