

# Accessory parotid gland with ectopic fistulous duct - Diagnosis by ultrasonography, digital fistulography, digital sialography and CT fistulography. A case report and review of current literature.

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## ABSTRACT

Accessory parotid glands are a common clinical occurrence and usually drain into the main Stenson's duct by small ductules and thereby, into the buccal cavity. Presence of an accessory parotid gland with an ectopic fistulous duct is a rare occurrence. We present the imaging findings in a case of right accessory parotid gland with ectopic fistulous duct associated with bilateral pre-aural appendages. Diagnostic workup was done by ultrasonography, sono-fistulography, contrast digital fistulography, contrast digital sialography and computed tomography fistulography. Imaging showed a right accessory parotid gland lying anterior to and separate from the main parotid gland draining via an ectopic fistulous duct opening over the right cheek. The child was managed surgically by internalisation of the duct to open into the buccal mucosa and excision of pre-aural appendages.

## CASE REPORT

### CASE REPORT

A 10 year old male child presented with history of intermittent discharge of watery fluid from an opening in the right cheek since birth. The discharge increased on eating and was not associated with any swelling or pain. There was no previous history of swelling, redness, pain, or injury to the region. He was otherwise healthy and had normal developmental milestones.

Patient had normal general examination with normal vital parameters. Systemic examination was within normal limits. Local examination revealed a small punctum on the right side of the face, at a distance of about 2.5cm from the angle of the mouth, and 1cm inferior to a line joining the tragus and angle of mouth (Fig. 1a.). Expression of clear serous fluid on local

massage was seen. No associated palpable mass or tenderness was noted. Oral examination was normal with normal Stenson's ducts opening bilaterally.

Bilateral pre-aural appendages were noted on regional examination, two on the right side- anterior and anteroinferior to the tragus, and one appendage on the left side just anterior to the tragus (Fig. 1a, b.).

As a part of the diagnostic workup, Ultrasonography, Sono-fistulography, contrast Digital Fistulography, contrast Digital Sialography and contrast CT fistulography were done.

On ultrasonography, (SIEMENS SONOLINE G50, 10-5 MHz Linear Probe), a well defined, hypochoic, ovoid, area with no internal vascularity was seen in the subcutaneous plane, anterior to the right Masseter muscle, and separate from

the main parotid gland (Fig. 2). A tubular hypoechoic structure was also visualised coursing from it anteriorly. On sono-fistulography, injection of normal saline into the fistulous opening demonstrated moving echoes within the duct and consequent palpable swelling in the region (Fig. 3). The main parotid gland was normal in position and extent.

Contrast digital fistulography and digital sialography were performed, one after another and later simultaneously (Prognosys Medical Systems, PRORAD 3N Digital Radiography). The external punctum was cannulated by a 24 gauge i.v cannula and 2 ml of water soluble radiographic contrast (Urograffin, ZydusCadila- German Remedies) in 70% dilution was injected. Contrast fistulography clearly demonstrated a smooth walled duct directed posteriorly and slightly superiorly. Contrast pooling was seen at the end of the duct in the soft tissue plane lateral to mandible on frontal (Fig. 3) and lateral projection (Fig. 4). Palpation revealed a localised swelling at the anterior margin of the right masseter muscle.

Digital contrast sialography was done by cannulating the normal orifice of right stenson's duct by 24 gauge i.v cannula, and injecting 2ml of contrast. The normal right Stenson's duct and fine ductules in the region of the right parotid gland were outlined. No passage of contrast was seen into the external fistulous duct (Fig. 5).

Fistulography and sialography were then done simultaneously. Findings revealed the two separate ducts, the prominent accessory duct coursing above and laterally to the main Stenson's duct. Two separate areas of contrast collection were seen, one in the region of the right parotid and another more antero-superior to the first. Thus, two separate gland systems were seen. (Fig. 6)

Plain CT (SIEMENS, SOMATOM, dual slice CT) revealed an area of soft tissue attenuation antero-lateral to the right masseter with relatively reduced bulk of the muscle (Fig. 7a.). On CT fistulography, with 2% diluted contrast, an opacified tubular shaped density was visualised with opacification of soft tissue area noted on the plain image (Fig. 7b.). No intra oral spillage of contrast was seen.

The imaging findings helped in arriving at a diagnosis of Accessory parotid gland with ectopic fistulous duct. The accessory gland and its ectopic duct were demonstrated on USG, sono-fistulography, contrast fistulography, contrast sialography and CT fistulography.

Patient was subsequently operated upon, and the ectopic fistulous duct was internalised to open in the buccal cavity. Although the pre aural appendages would not have lead to any impairment, they were excised for cosmetic purposes (Fig. 1c.).

## DISCUSSION

Parotid glands are paired serous salivary glands located lateral and posterior to the masseter muscle. They extend from the zygoma superiorly, to the angle of the mandible inferiorly, and are limited posteriorly by the external auditory meatus. Anteriorly, the gland extends to near half of the masseter width. The Facial nerve divides the parotid into the superficial and deep lobes. The gland is drained by the main parotid duct or Stenson's duct into the buccal cavity. The Stenson's duct arises from the anterior margin of the gland, courses anteriorly, slightly inferiorly and superficial to the masseter muscle where it is palpable, then turns medially at the anterior margin of masseter to pierce the buccinator muscle and open into the mouth. The internal opening is a slightly elevated punctum at the level of the upper second molar tooth.

Accessory parotid glands have been described in about 21% of general population [1]. These glands, mostly pea sized, unilateral, are located superior to the main parotid duct, anterior to the masseter, and separate from the parotid gland. They drain via one or two channels into the main parotid duct. They may consist of both serous and mucinous acini [2].

Frommer et al first described the incidence of accessory parotid gland as 21 % in a study of 96 human cadavers [1]. However, no record exists of the incidence of accessory gland with an ectopic duct. Accessory parotid gland having a duct of its own is exceedingly rare [1], [2]. The duct courses anteriorly, inferiorly and opens externally to the skin behind the angle of the mouth. An association has been observed between co-existent pre aural skin appendages. However, this association is yet to be proven conclusively as very few cases have been reported till date.

Naguru H et al reported the first case of salivary fistula associated with aural appendage in 1972 from Japan [3]. Yamasaki et al used the Delore's method of surgical translocation of the ectopic fistulous duct to the buccal cavity in a similar case in 1986 [4]. Moon et al reported a case in a 5 year old girl with an ectopic accessory parotid gland with a fistulous duct from Korea. They accurately demonstrated the ectopic gland by CT sialography and CT fistulography. However, they were unable to demonstrate the normal ipsilateral parotid gland, thereby, justifying the use of their term Ectopic Accessory parotid gland [5].

Gadodia et al reported the imaging findings in an 8 year old boy with congenital salivary fistula from an accessory parotid gland in 2008, from India [6].

Hun Hah et al reported a surgical management method for a similar case, also from Korea by chemo cauterization using tri-chloro-acetic acid (TCA) in 2008. They suggested that most surgical incisions for operating fistulae in the face leave a scar; hence chemo cauterization by TCA is a better option [7]. However in our case, we would like to point that if the accessory parotid gland has no other drainage tract then it would be unwise to cauterize the fistula leaving the gland intact, which can get infected later on. If cauterization is deemed necessary then the accessory gland must also be

excised. A better option would be to internalise the ectopic duct opening and leaving the accessory gland in situ to drain into the mouth.

In our case, the ten year old male child presented with a fistula on the right cheek with bilateral pre auricular appendages. Acquired causes like traumatic, infective, or post surgical could be ruled out at the outset as history was from birth. Imaging workup as described previously provided conclusive evidence to arrive at a diagnosis.

USG demonstrated a well defined soft tissue structure in the region anterior to the echogenic main parotid gland with a tubular extension coursing anteriorly. This pointed towards the possibility of an accessory parotid gland. Moving echoes in the tubular structure on sono-fistulography proved that the soft tissue structure was communicating to the external fistula via a tract. Sono-fistulography with normal saline is particularly useful as a safe method to determine the course of fistulae in soft tissues by observing the dilatation of tract or duct and moving echoes inside. Contrast digital fistulography clearly defined the ectopic fistulous duct, with no communication with the stenson's duct. Contrast sialography done in immediate succession demonstrated the normal parotid gland and its normal duct, entirely separate from the fistulous duct. Contrast CT fistulography supported the sialography and fistulography findings by outlining the relationship of the ectopic duct, and of the accessory parotid gland with the other tissues of the cheek region in axial scans. Opacification was seen in the ectopic duct and in the accessory parotid gland on contrast injection.

These findings along with history and examination findings of serous discharge on massage and eating proved that the fistulous duct originated from an accessory right parotid gland.

"Accessory parotid gland with ectopic fistulous duct" would be the appropriate nomenclature, as there was a normal parotid gland with its normal duct, and an accessory gland with an ectopic duct which did not communicate with the stenson's duct, but opened as a fistula over the cheek.

Fistulae in the parotid region generally arise secondary to trauma, surgery, malignancy, or infection and are seen more often in adults [8]. They can be differentiated from congenital fistulae based on history. Fistulae in paediatric age are mostly congenital. The congenital fistulae in the region of the face and neck result from the partial failure in normal involution of the branchial arches during embryonic development. Important differential would be pre-auricular sinuses, which are seen more posteriorly near the tragus, end blindly and may be visualised on ultrasound. Local lesions like epidermal inclusion cysts or basal cell carcinoma which can present as discharging sinus can be excluded by clinical examination and histology.

The main parotid duct marks the landmark between the closing mandibular process and the maxillary process of the face during embryogenesis. The parotid gland can be recognized in human embryos at Stage 15 as an elongated

furrow running dorsally from the angle of the mouth between the mandibular and maxillary prominences. The groove, which is converted into a tube, loses its connection with the epithelium of the mouth except at its ventral end and grows dorsally into the substance of the cheek. The tube persists as the parotid duct and its blind end proliferates in the local mesenchyme to form the gland. Subsequently, the size of the oral fissure is reduced by partial fusion between the maxillary and mandibular prominences, and the duct opens thereafter on the inside of the cheek at some distance from the angle of the mouth. The parenchymal part of the parotid gland is formed by branching of the blind end of the parotid tube and by the development of structures of ectodermal (secretory parenchymal) and mesodermal (stromal) nature. The accessory parotid is derived from a similar branching and a similar glandular proliferation, arising from the parotid tube more anteriorly and clearly separate from the main parotid tissue [9], [10].

However, in our case we postulate that around the same time as the main duct develops from the furrow between the maxillary and mandibular processes, at about 4 weeks of development, the primeval groove duplicates and grows cranially and posteriorly to form two ducts and subsequently, the normal and accessory parotid separately. During fusion of the maxillary and mandibular processes, the normal parotid duct remains patent from inside the mouth, whereas the duct of the accessory gland opens as a cutaneous fistula.

The auricle is formed from six hillocks, out of which the tragal hillock along with two other arises from the first branchial arch. Pre-aural appendages are remnants of additional tragal hillocks.

Thus, coexistence of accessory parotid gland with ectopic fistulous duct and pre-aural appendages can be explained as an example of malformations of the First Branchial arch derivatives.

Excluding our case, only five such cases have been reported in English literature, and all from Asian countries. As a result, the exact nomenclature of this condition is yet to be concluded upon. There could be a regional or racial predilection to this condition. Unless a substantial number of cases are studied, the epidemiology and pathogenesis remain controversial.

#### TEACHING POINT

Children presenting with a congenital fistulous opening on the side of the cheek, with history of serous discharge should be suspected as a case of Accessory parotid gland with ectopic fistulous duct. Although a benign condition with good prognosis, its surgical management depends upon exact delineation of duct and gland by imaging modalities such as Ultrasonography, Sono-fistulography, contrast sialography, contrast fistulography, and CT fistulography.

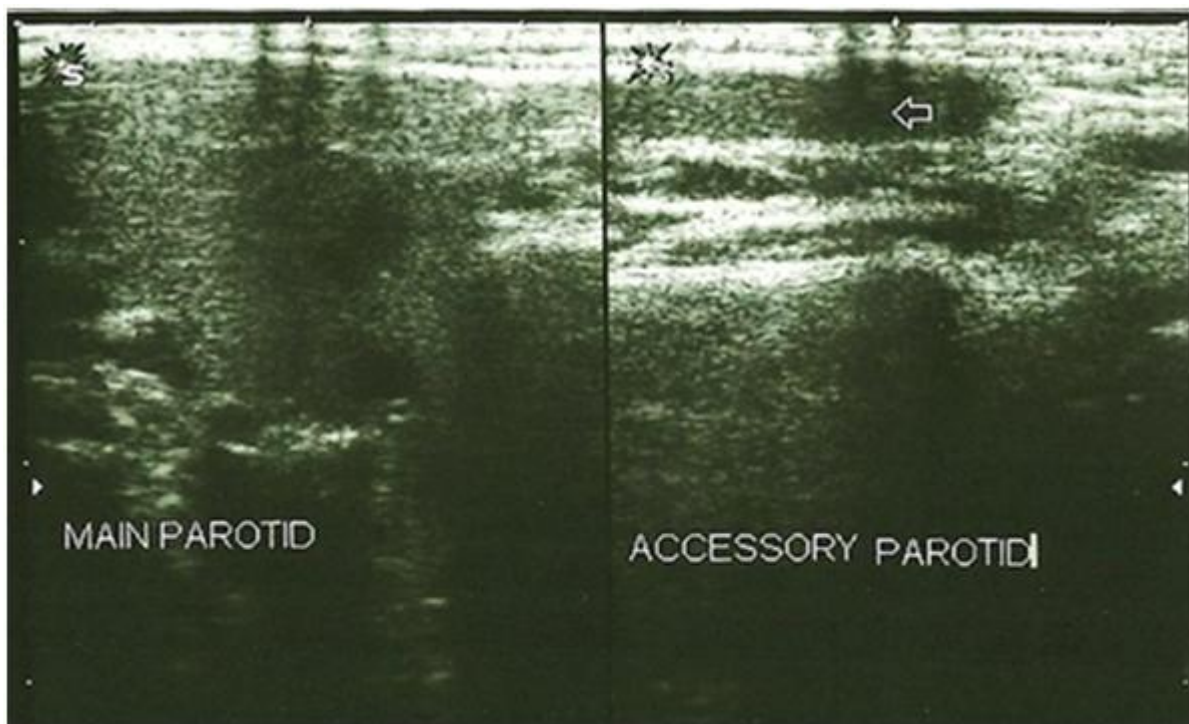
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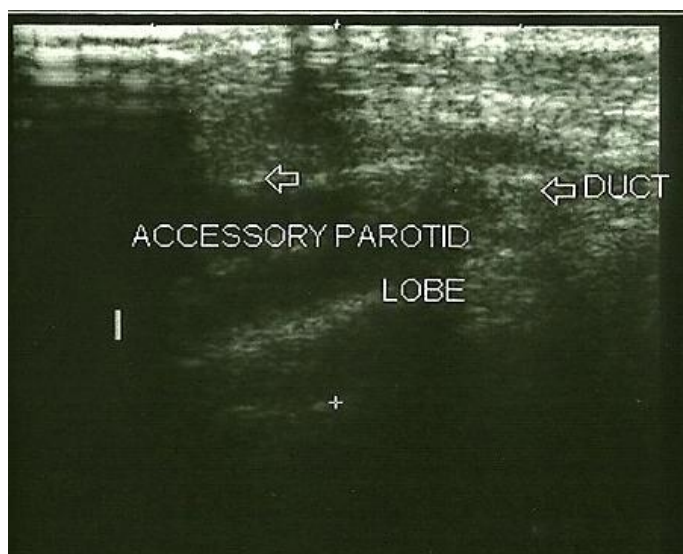
FIGURES



**Figure 1:** 10 year old male child with right accessory parotid gland with ectopic fistulous duct. Clinical photographs showing the external opening of the fistulous duct (a)(black arrow). Associated observations were pre-aural appendages (black arrowheads), two on right side (a), and one on the left side (b). Post operative photograph (c) shows the suture site where the opening of the fistulous duct was internalised (white arrow). And the site of excised pre-aural appendages (white arrowhead)



**Figure 2:** 10 year old male child with right accessory parotid gland with ectopic fistulous duct. Ultrasonography (Siemens Sonoline G50, 10-5 MHz, linear probe) demonstrates the accessory parotid gland (arrow) as a hypoechoic area lying separate from the main parotid gland.



**Figure 3:** 10 year old male child with right accessory parotid gland with ectopic fistulous duct. Sono-fistulography (Siemens Sonoline G50, 10-5 MHz, linear probe) with normal saline demonstrates moving echoes within the hypoechoic duct (arrow).



**Figure 4:** 10 year old male child with right accessory parotid gland with ectopic fistulous duct. Digital fistulography with 2ml of 70 % diluted Urograffin, on frontal projection, demonstrates the fistulous duct (arrowhead) and pooling of the contrast in the accessory parotid gland lateral to the mandible (arrow).



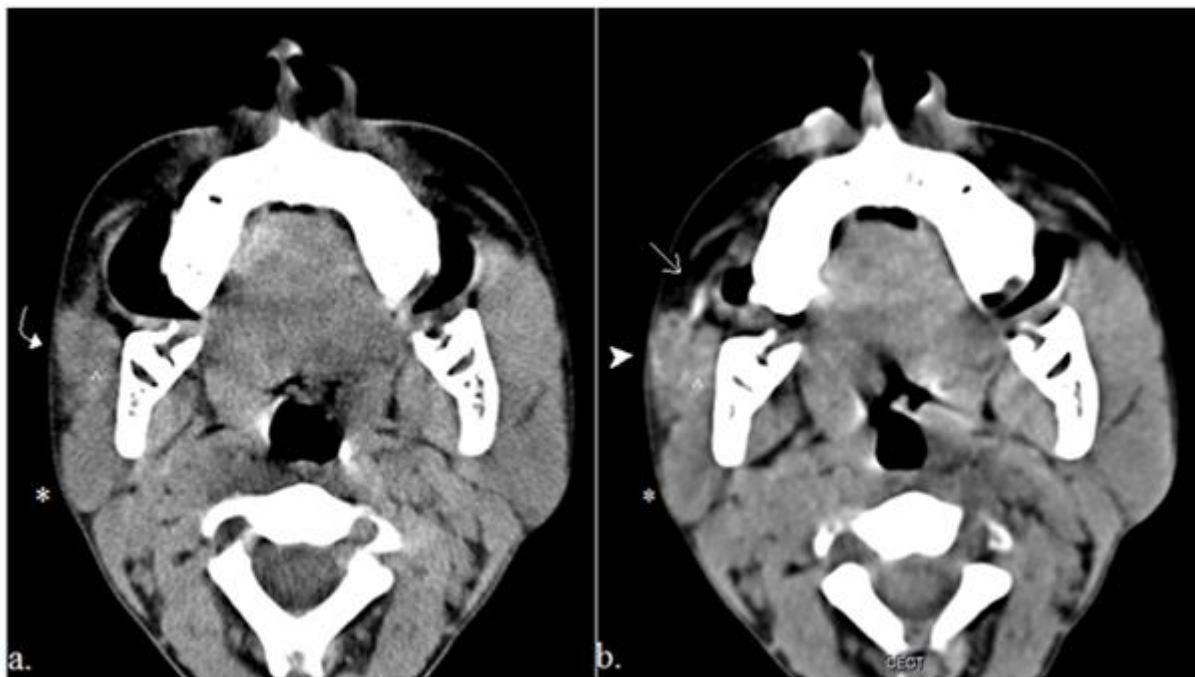
**Figure 5:** 10 year old male child with right accessory parotid gland with ectopic fistulous duct. Digital fistulography with 2ml of 70% diluted Urografin, on lateral projection shows the fistulous duct coursing posteriorly (arrowhead). Pooling of contrast is seen at the end of the duct in the accessory parotid gland (arrow).



**Figure 7:** 10 year old male child with right accessory parotid gland with ectopic fistulous duct. Digital sialography and fistulography combined, with 70% diluted Urografin, on frontal projection demonstrates two separate duct systems, the normal right Stenson's duct (black arrowhead), and the fistulous duct (curved arrow). The accessory parotid gland is also opacified (white arrow).



**Figure 6 (left):** 10 year old male child with right accessory parotid gland with ectopic fistulous duct. Digital parotid sialography with 2ml of 70% diluted Urografin, on frontal projection demonstrates the Stenson's duct (arrow) and the normal right parotid gland (arrowhead). No communication with fistulous duct is seen.



**Figure 8:** 10 year old male child with right accessory parotid gland with ectopic fistulous duct. Axial CT (Siemens Somatom, Dual slice CT with 75mAs, 130kV, and 5 mm slice thickness), plain (a) and contrast CT fistulography (b) images. Plain image (a) demonstrates the accessory parotid gland (curved arrow), lateral to the mandible, anterior to the masseter (^) and separate from the main parotid gland (\*). Contrast CT fistulography with 2% diluted Urograffin demonstrates increased attenuation in the fistulous duct (straight arrow) and in the accessory parotid gland (arrowhead).

<b>Etiology</b>	Congenital (Probable duplication of primitive Stenson’s duct at about 4 <sup>th</sup> week of intra-uterine life).
<b>Incidence</b>	Five Cases reported till date (Excluding present case in English literature).
<b>Gender ratio</b>	None
<b>Age Predilection</b>	Congenital, Patient usually presents in childhood.
<b>Risk Factors</b>	Unknown
<b>Treatment</b>	Surgical correction. Internalisation of the ectopic duct opening into the buccal mucosa, and excision of pre-aural appendages.
<b>Prognosis</b>	Good
<b>Findings on Imaging</b>	Ultrasound, contrast fistulography, sialography and CT fistulography demonstrate accessory parotid gland with a separate ectopic duct, not communicating with the normal parotid gland and stenson’s duct.

**Table 1:** Summary table for accessory parotid gland with ectopic fistulous duct.

Condition	USG	Sono-fistulography	Contrast fistulography	Contrast sialography	CT fistulography
<b>Accessory parotid gland with ectopic fistulous duct</b>	Hypo- to isoechoic well defined area separate from main parotid gland	Moving echoes in the ectopic duct with increase in size of accessory gland.	Well defined opacification of duct, course of duct identified up to accessory gland, no communication with stenson's duct.	Normal parotid gland with intra-glandular ductules.	Opacification of ectopic duct and enhancement of accessory parotid gland with no change in normal parotid gland.
<b>Acquired parotid fistulae</b>	Only normal echogenic parotid gland seen.	Moving echoes in the fistulous tract	Irregular blind ending tract seen to end in the region of main parotid. May communicate with the Stenson's duct.	Normal stenson's duct opacified. May communicate with fistula.	Opacification of fistulous tract with enhancement in main parotid gland.
<b>Pre-auricular sinus</b>	Sinus anterior to tragus with relationship to superficial temporal artery.	Moving echoes along course of the sinus.	The sinus tract visualised in the soft tissue plane of auricle directed medially.	Normal parotid gland with intra-glandular ductules.	The sinus tract is lateral and superior to the facial nerve and parotid gland.

**Table 2:** Differential diagnosis table for accessory parotid gland with ectopic fistulous duct.

**ABBREVIATIONS**

USG: Ultrasonography  
 CT: Computed Tomography  
 TCA: Tri-Chloro-Acetic Acid

**KEYWORDS**

Accessory parotid gland; ectopic fistulous duct; congenital parotid fistula; pre-aural appendage

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