Dermoid Cyst of the Parotid Gland: Report of a Rare Entity with Literature Review

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Received: 7 July 2014 / Accepted: 4 August 2014 / Published online: 13 August 2014
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Abstract Dermoid cysts (DCs) are benign lesions and histologically composed of tissues originating from ectoderm and mesoderm, but not endoderm. Approximately 7 % of all DCs are seen in head and neck area. However, parotid gland is an extremely rare localization in which DCs develop, and only 17 cases have been reported in the literature to date. Correct preoperative diagnosis is difficult to be established due to the rarity and ambiguous radiological findings. We report a case of a 21-year old man. All previous reports reviewed and the pathogenesis as well as the histopathologic and radiologic features are discussed.

Keywords Dermoid cyst · Parotid gland cyst · Head and neck cyst

Introduction

Dermoid cysts (DCs) are benign lesions, lined by stratified squamous epithelium with variable amount of pilosebaceous units and sweat glands supported by a fibrous

connective tissue wall. They are histologically derived from ectoderm and mesoderm. Tissues from endodermal origin, on the other hand, are never found in these cysts [1]. The lumen usually contains sebaceous material and occasionally hairs. The head and neck area is a common site of occurrence for DCs and they are located most often in superficial subcutaneous tissues [2–4]. DCs can occur in any age with no gender predilection. More than 50 % of all head and neck region cases are detected by the age of 6, though approximately one-third are present at birth [2–4]. These cysts are generally asymptomatic unless they grow enough to be conspicuous, causing cosmetic problems or compressive effects. Although DCs are common in the head and neck area, they are extremely rare in the parotid gland. To our knowledge, 17 DCs in total were reported in 15 prior case reports to date [5–19].

We herein present a rare case of parotid DC and review of data in light of the whole accessible cases in the English and non-English literatures, published between the years of 1982 and 2014.

Case Report

A 21-year-old man presented with a 2 month history of a painless expanding mass of the left facial region. He had no history of any prior disease, surgery or trauma related to this area. Physical examination showed a mobile, soft and somewhat fluctuant mass near the angle of mandible. There was no evidence of palpable lymphadenopathy or any neurologic deficit. Ultrasonography (US) showed a well-defined cystic lesion measuring 2.8×2.7 cm in the tail of the parotid gland. Subsequently, the computed tomography (CT) scan revealed a $3 \times 3 \times 4$ cm mass with round, well-delineated, homogenous and hypodense appearance with

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Fig. 1 **a** Coronal CT imaging shows a well-defined lesion in the left parotid gland. **b** Axial sections show subtle linear structures, which were thought to be hair shafts, near the periphery of this fat containing cyst (arrows)

fat density in the superficial lobe of the left parotid gland (Fig. 1a). Additionally, a 0.4 cm nodular area was identified within the posteroinferior localization of the lesion. By close inspection, multiple thin linear reflections in the lesion suggesting calcified crystalline material or hair structures were observed (Fig. 1b). There was no other accompanying lesion in the head and neck region. Radiologically, it was considered a benign lesion suggestive of lipoma or DC. Fine needle aspiration cytology (FNAC) was not performed preoperatively.

The patient underwent a superficial parotidectomy. Intraoperative examination showed a well-defined and fluctuant lesion which was occupying almost the entire of the superficial lobe of the parotid gland. Only a small portion of the lesion caused a bulge on the parotid gland and the cyst wall seemed to adhere to the buccal and zygomatic branches of the facial nerve. The nerve branches were spared by using a nerve stimulator. Postoperative follow-up period was uneventful with normal functioning.

Pathologic Examination

Macroscopically, the excised superficial lobe of the parotid gland measured $5.5 \times 4.5 \times 4.5$ cm. On cut surface, a unilocular cyst measuring $3.5 \times 3.5 \times 3$ cm was observed. The cyst had a thick white-yellow wall, and its inner surface showed focal irregularities, grossly. The lumen of the cyst contained amorphous, yellowish-pink colored and soft material with a few hair shafts. The lesion was submitted totally and processed routinely. The remaining pathology specimen was consistent with normal parotid gland.

Microscopic examination revealed a well-circumscribed and partially collapsed unilocular cystic lesion in the

parotid gland parenchyma. The cyst wall was lined by mature keratinized stratified squamous epithelium with pilosebaceous units and occasional apocrine sweat glands in the underlying collagenized stroma (Fig. 2a). In many areas, the cyst wall showed foreign body type granulomatous inflammation with denudation of the cyst lining. This finding indicates that the cyst was partially ruptured. The epithelial lining was mostly denuded and the underlying fibrous tissue with or without remaining pilorector smooth muscle bundles was exposed in some areas as well (Fig. 2b). The lumen was filled with squamous cells, macrophages, and hair shaft fragments. Any other component, which was supposedly derived from the embryonic germ layers, rendering the diagnosis teratoid cyst instead of DC was not detected. A mild lymphocyte infiltration was observed in the adjacent parotid tissue. The remaining parotid gland was otherwise normal and contained one intraparotid lymph node.

Discussion

DCs are generally regarded as benign entities. More than half are detected in children around 6 years of age, while approximately a third of DCs are conspicuous at birth [2–4]. Rare cases of malignant transformation of DCs have been reported in the oral cavity with a probability of 5 % [20, 21]. It is known that 7 % of all DCs are seen in head and neck region and 80 % of them predominantly occur in the orbit, floor of mouth and nose [22–24]. The head and neck is the third most common site, after the coccyx and the ovary, with a rate of 44.5 and 42.1 %, respectively [25]. Although rare the parotid gland can harbor DCs. In general, cystic lesions account for 2–5 % of all parotid gland lesions [6]. A total of 17 DCs in the 15 prior case reports

Fig. 2 **a, b** Intraparotid cyst is lined by stratified squamous epithelium with pilosebaceous units. H&E, $\times 20$ and $\times 40$. **c, d** Squamous epithelium was replaced with a foreign body type

inflammatory granulomatous reaction in most areas. Note the hair shaft fragment in the lumen (*arrow*). H&E, $\times 40$ and $\times 100$

have been published in the literature up to date, to our best knowledge (Table 1) [5–19]. Unfortunately, we could not obtain the very first case report reported in the literature, despite all our efforts. Therefore, we were able to summarize 17 cases, in addition to ours, in Table 1. Of these 17 patients, 13 were male (76 %) and 4 were female (24 %). Patients' age ranged from 9 to 69, with median of 31. Twelve of these 17 lesions were in right parotid gland (71 %), and the remaining 5 were in left (29 %). Lesions' greatest dimensions varied, from 2 to 5.5 cm.

Patient complaints were usually related from slow-growing painless mass recognized as early as 2 months to a 20 year period. Physical examination generally shows a mobile and non-tender lesion with or without fluctuation. Preoperatively, the differential diagnoses include lipoma, branchial cleft cyst, lymphoepithelial cyst, mucous retention cyst, suppurative infections, pleomorphic adenoma, fibroma or neurofibroma, and blockage of the parotid duct.

Batsakis described three histological types for such benign head and neck cysts: Epidermoid, dermoid and teratoid [26]. Epidermoid cysts are composed of epidermal type squamous epithelium without any adnexal structures derived from the lining epithelium. DCs have the same squamous epithelium with pilosebaceous units and sweat glands. Teratoid cysts, also known as mature cystic teratomas, contain variable admixtures of mature tissues

originating from all three germ layers and harbor additional components to those of DC structures, like glioneuronal elements from ectoderm, cartilage from the mesoderm or respiratory epithelium from the endoderm.

Some authors, on the other hand, preferred to classify these cysts simply in two categories: Congenital and acquired. Congenital cysts arise from a rest of embryonic epithelium such as branchial cleft cyst. Acquired cysts are generally results of traumatic or iatrogenic displacement of the skin into the deeper layers. New and Erich classified these cysts into three following categories [23];

1. Congenital DCs of teratoma type: They arise from the embryonic germinal layers. Depending on the dormant layer, they can contain skin, hair or teeth etc. They are almost always limited to the ovaries and testes.
2. Acquired DCs: They are inclusion cysts as a result of traumatically implanted skin in the deeper tissues. They occur on the hands and the other exposed parts of the body.
3. Congenital inclusion DCs: These develop from inclusions of displaced dermal cells along the lines of embryologic fusion. This category is subdivided into four subgroups:
 - (a) Cysts around the eyes and orbits, originating along the nasooptic groove,

Table 1 Case specifications in previous reports

Authors	Age	Gender	Side	Location in the gland	Radiologic appearance	Dimensions	Treatment	Postoperative nerve weakness	Recurrence
1. Princ et al. [5]	NA	NA	NA	NA	NA	NA	NA	NA	NA
2. Choi et al. [6] ^a	22	Male	Right	Superficial lobe	Fatty	4 × 4 cm	Superficial parotidectomy	NA	Following previous simple excision, which was performed 1 year earlier
	22	Male	Left	Inferior portion of superficial lobe	Fat containing cyst	4 × 5 cm	Superficial parotidectomy	NA	
3. Moody et al. [7]	37	Male	Right	Superficial lobe	Fatty	2.5 × 1.5 × 0.5 cm	Superficial parotidectomy	Facial nerve weakness which resolved completely in 5 months	No
4. Yutaka et al. [8]	38	Female	Right	Deep lobe	Cyst	4 cm	Total parotidectomy	Facial nerve weakness which resolved completely in 3 months	No
5. Baschinsky et al. [9] ^a	38	Male	Left	Tail of the parotid gland	Cyst	3.0 × 1.9 × 1.7 cm	Superficial parotidectomy	NA	No
	26	Male	Right	Superficial lobe	Cyst	2.5 × 2.0 × 1.5 cm	Superficial parotidectomy	NA	No
6. Naujoks et al. [10]	46	Male	Right	Superficial lobe	Soft tissue mass	3 × 2.3 × 2.7 cm	Superficial parotidectomy	Weakness of the forehead-branch of the facial nerve, which improved in weeks	No
7. Lee [11]	15	Female	Right	Superficial lobe	Fatty	3 × 2.7 × 3 cm	Superficial parotidectomy	No	No
8. Islam and Hoffman [12]	69	Male	Left	Superficial lobe	Cyst	5.5 × 2.6 × 1.4 cm	Superficial parotidectomy	No	No
9. Saylam et al. [13]	42	Female	Right	Superficial lobe	Fatty	3 × 3 cm	Superficial parotidectomy	NA	No
10. Aynehchi et al. [14]	18	Male	Right	Deep lobe	Fat containing cyst	4.5 × 2.0 × 0.5 cm	Total parotidectomy	No	No
11. Seo and Yoon [15]	35	Male	Right	Superficial lobe, extending to the deep lobe	Cyst	4 × 3 cm	Total parotidectomy	No	No
12. Tas et al. [16]	9	Male	Right	Superior portion of the superficial lobe	Soft tissue mass	2.4 × 1.8 × 2.5	Superficial parotidectomy	Weakness of the eye and buccal branches of the facial nerve, which resolved in 4 weeks	No
13. Trandafir et al. [17]	21	Male	Right	Superficial lobe	Soft tissue mass	2 × 2 × 2.5 cm	Superficial parotidectomy	Facial paralysis disappeared in 2 months	No
14. Birsan et al. [18]	20	Female	Right	Superficial lobe	Fat containing cyst	2 × 2 cm	Superficial parotidectomy	Mild residual weakness in the lower lip, which improved subsequently	NA

Table 1 continued

Authors	Age	Gender	Side	Location in the gland	Radiologic appearance	Dimensions	Treatment	Postoperative nerve weakness	Recurrence
15. Gonzalez-Perez and Crespo-Torres [19]	43	Male	Left	Superior portion of the superficial lobe	Soft tissue mass	4.8 × 4.5 × 4 cm	Superficial parotidectomy	No	No
16. Our case	21	Male	Left	Tail of the parotid gland	Fat containing cyst	3.5 × 3.5 × 3 cm	Superficial parotidectomy	No	Follow-up period, which is 2 month only, is not enough to evaluate any recurrences

NA Not Available

^a Choi et al. and Baschinsky et al. presented two cases in their reports

- (b) Those around the nose, resulting from intrusion of the frontonasal plate,
- (c) Those around the floor of the mouth, and in submental and submaxillary regions, originating from the 1st and 2nd branchial arches,
- (d) Those around the suprasternal, suboccipital, thyroïdal, lower lip and palate, most of which occur along the midventral or middorsal lines of the body.

Because of the rarity and the absence of consensus, DCs of the parotid gland are difficult to classify. Choi et al. [6] hypothesized that these lesions may be considered in the third subgroup of the third category described above. We think it might also be logical that the second category may clarify the etiology in the light of the following issues;

- Although we came across DCs of the submandibular and sublingual region, areas which are closer to the midline when compared to the parotid gland, in the literature, none of them was seen within the submandibular or sublingual gland [27–29]. This observation does not support the theory put forward in the third subgroup. Also, these glands are more sheltered than parotid gland for external traumatic injury due to their anatomic locations. Hence, the theory of trauma as a possible etiologic factor sounds reasonable. Furthermore, the DCs are usually seen within the superficial lobe of the parotid gland. It might also reflect the relative vulnerability of parotid gland, supporting trauma hypothesis. Accordingly, Gonzalez–Perez and Crespo–Torres suggested a relationship between recurrent trauma and DC development in the parotid gland for their presented case [19].
- Traumatic dermoepidermal tissue implantations by surgery or needle sticking during lumbar puncture was blamed for spinal DCs [30, 31]. Hence, the similar mechanism seems applicable for parotid DCs.
- There were some published case reports of epidermoid cyst of the parotid gland, nearly half of which developed after otologic surgery [32–34]. This relationship of prior surgery in cases of parotid epidermoid cysts further strengthens the possibility of a similar etiology for DCs.
- Although most of the DCs of head and neck region are seen among children, parotid DCs are almost always reported in adults, except for one case reported in a 9-year old child [16]. Hence, the higher frequency of DCs in adult age group support the possibility of the non-congenital theory.

The clinical differential diagnosis is broad and should include epidermoid cyst, branchial cleft cyst, parotid duct cysts, infectious diseases, benign mesenchymal tumors,

and salivary gland neoplasms such as pleomorphic adenoma or mucoepidermoid carcinoma [6–12].

US imaging often reveals a well-circumscribed cystic lesion which may have mixed internal echoes because of its fat-resembling content. Additionally, due to the presence of cellular material within the cyst, it may exhibit a pseudo-solid appearance on US with uniform homogeneous internal echoes. Although most of the truly solid tumors could be recognized and differentiated from dermoid or epidermoid cysts with US, lipomas might still be indistinguishable from these cysts [35]. On CT scans, the lumen is usually filled with a homogenous, hypodense fluid-quality material. This material may be reminiscent of fat as well. Magnetic resonance imaging (MRI) with its better soft tissue contrast and improved imaging capacity has advantage over US and CT. MRI of a DC typically depicts a cystic mass, hypointense on T1-weighted images and hyperintense on T2-weighted ones with peripheral enhancement on contrast. Some DCs have variable signal intensity on T1 weighted images which may be hyperintense because of the presence of sebaceous lipid [36]. It might be useful to keep in mind that in all imaging techniques, namely US, CT and MRI, globules of fat floating within the lumen may produce a characteristic “sack of marbles” appearance which is considered nearly pathognomonic for a DC. Rarely, fat and/or fluid levels within the lumen may also be identified [1]. Sometimes, DCs can be identified by interlacing linear and punctuate reflections corresponding to crossing strands of hair within the cyst, a feature that is most characteristic of hair embedded in sebum as in the ovarian counterparts [37]. This finding was not observed on radiologic investigations of the prior case reports of parotid DCs, except our case. Briefly, when a partial or total fatty appearance suggestive of the lipoma, “sack of marbles” appearance, fluid levels and/or thin linear or punctate structures are detected radiologically, DCs should be kept in mind. Radiologically, 5 of the previously reported 17 cases in the literature were seen as purely cystic, 4 of them were reported as cysts with fatty appearance, 4 of them had purely fatty appearance, and another 4 of them were considered soft tissue mass without any specific diagnosis.

Cytological evaluation revealed benign appearing squamous cells, anucleated squames, and amorphous cellular debris [9, 18, 38, 39]. When foreign body type granulomatous reaction was intense, multinucleated giant cells and macrophages might be the dominant cell population in the aspiration cytology smears. FNAC may also show squamous cells with reactive atypia when the cyst was superimposed with acute or chronic inflammation. FNAC can be used to identify a cystic lesion in nature when radiologic findings are compatible with lipoma. A total of 8 among these 17 cases were also examined by

FNAC, preoperatively. In one of the case reports, Birsan et al. [18] observed hair shaft fragments in aspiration cytology which was a strongly suggestive finding for DC. However, the possibility of hair fragments that were artifactually pushed into the cyst lumen from the overlying skin should not be forgotten. The significance of FNAC for preoperative diagnosis was highlighted in some reports [9–12]. Similarly, Cramer et al. [40] recommended FNAC for DCs in the mouth floor as the first-choice diagnostic procedure. But some authors did not consider the FNAC as a definitive diagnostic modality [16, 19]. In summary, FNAC may provide reliable information on cystic lesions if there are supporting radiologic findings, but can also be deceiving because some well-differentiated squamous cell carcinomas and malignant lesions showing squamous metaplasia may share the benign-looking cytologic features with DCs. Hence the definitive diagnosis should always be made by histopathologic examination [7, 10].

Surgically, simple excision of the cyst in the parotid gland was not recommended to avoid the possible remnants of the lesion which may be responsible for later recurrences. In one case report, superficial parotidectomy had to be done after 1 year from the initial simple excision because of the recurrent lesion [6]. Superficial parotidectomy is the standard intervention for the lesions in the superficial lobe, which seems the most frequently encountered location of DCs (14 cases). Total parotidectomy, on the other hand, is recommended for deep-seated DCs, which is seen very rarely (3 cases only). In all kinds of surgical intervention, careful dissection of the nerves from the cyst wall is paramount to prevent damage which may potentially cause neurologic deficits. In postoperative course, neurologic weakness of facial nerve or its branches was observed in 6 of the 17 cases which were improved in up to 5 months. None of the cases in the literature showed late recurrences.

Conclusion

DCs of the parotid gland are rare benign lesions. There is no agreement on their mechanisms of development. Radiologic investigations generally reveal a well-defined lesion with various appearance which might be cystic, fatty, fat containing cyst or soft tissue mass. FNAC can provide clues of a benign cystic lesion lining with squamous epithelium. However, a definitive diagnosis cannot always be made preoperatively. Instead of simple excision, nerve-sparing superficial or total parotidectomy has been suggested for the curative treatment. Postoperatively, temporary short term facial nerve dysfunctions observed in nearly half of the cases.

Conflict of interest The authors declare no conflict of interest.

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