REVIEW

Glomus coccygeum: a review

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Key words

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Summary

With limited information about the coccygeal glomus found in classic textbooks, we deemed it necessary to review the subject. The illustrations presented in this article derive from four coccygeal glomera incidentally encountered during examination of pilonidal disease specimens. Familiarization with its microanatomical features may help to avoid inappropriate interpretation of this enigmatic structure.

Introduction

A glomus body is a spheroidal to ovoid dermal microanatomical structure that constitutes an arteriovenular glomeriformis anastomosis. Each glomus consists of one or more arterial segments that branch into a number of glomic arterioles surrounded by rows of round, uniform, epithelioid contractile cells, that drain into a collecting venous plexus ¹. These sphincteric anastomoses, the essential parts of the glomus, are eponymously known as the Sucquet-Hoyer canals after J.P. Sucquet (1840-1870), a French anatomist, and H. Hoyer (1864-1947), a Polish anatomist, who first described them.

Endowed with a high-sympathetic tone, the glomus body is involved in skin thermoregulation acting as a sphincter to control the flow of blood by locally mediated axon reflex responses, to bypass or enable the capillary bed, and prevent loss or dissipate heat.

Knowledge of the function of cutaneous arteriovenous anastomoses in man dates back to the early 1930s when the work² of the British physiologists and cardiologists Thomas Lewis (1881-1945) and George W. Pickering (1904-1980) as well as the work³ of the British cardiologist Ronald T. Grant (1892-1989) and the American cardiologist Edward F. Bland (1901-1992) were published. These arteriovenous anastomoses are scarce in the newborn, but develop rapidly during infancy and early childhood. In the elderly, the glomera regress, sclerose, and diminish in number, thus explaining the increase susceptibility to thermoregulatory disturbances affecting the extremes of life¹.

Glomera are frequently encountered in the hands and feet, chiefly in the deep dermis of the digital pads and nail beds, but they also occur in the ears, nasal and alimentary mucosa (where the function is related to absorption), thyroid, erectile tissue, and many other sites including the coccygeal region ¹.

In 1860, Hubert von Luschka (1820-1875), a German surgeon and anatomist, first identified the glomus coccygeum at the ventral tip of the coccyx in his dissection studies of the pelvis, naming it "glandula coccygea"⁴. Luschka compared glomus coccygeum with the carotid body ("glomus caroticum"), a chemoreceptor, thinking that both were glands. In 1865, the Swiss-born Julius Arnold (1835-1915) described the "glandulae coccygeae" even on the ventral surface of sacrum, recognizing their vascular origin along the median sacral artery, and naming them "glomeruli arteriosi coccigei" ⁵. He considered both glomus coccygeum and "glomus" caroticum as true glomeruli ⁶. In 1902, these microanatomical structures were included in the paraganglion system ⁷ by the Austrian histologist Alfred Kohn (1867-1959). In 1907, the non-paraganglionic (non-chromaffin) nature of the glomus coccygeum was demonstrated histochemically⁸

by another Austrian *Mediziner*, the anatomic pathologist Oskar Stoerk (1870-1926). However, credit is due to William H. Hollinshead (1906-1986), an American anatomist, who in 1942 categorically established anatomical and physiological distinctions between the "glomus" coccygeum and "glomus" caroticum (carotid body) ⁹.

Several terms have been used in the past, referring to the same glomus coccygeum, such as coccygeal body, glomus coccygicum, pericoccygeal glomus, Luschka's body, gland of Luschka, coccygeal gland, and "glandula coccygea".

Glomus coccygeum is homologous to the "caudal glomeruli" (also called "glomerula caudalia"), which are non-nutrient arteriovenous anastomoses present in variable number (up to 15) – according to species – in tailed mammals, from rodents to monkeys ⁹⁻¹⁶. However, while there is evidence that caudal arteriovenous anastomoses in animals play an important role in thermoregulation ¹⁵⁻ ¹⁷, glomus coccygeum in humans (and in other tailless primates as well) represents a phylogenetic vestigial rest, that is an atavic organ loosing the normal thermoregulatory function as the dermal glomera of other sites. In fact, the subcutaneous panniculus, where the glomus coccygeum resides, serves as an insulator, not as a dissipator or absorber of heat.

However, the precise function of this structure in humans remains speculative ¹⁸. A presumptive hematopoietic function via an immunomodulatory activity of the sympathetic nervous system has been recently proposed ¹⁹.

There is limited data, if any, regarding glomus coccygeum in classic textbooks of anatomy and physiology, and therefore in this article we review the world literature on the subject.

Anatomic and clinical findings

There is no recorded evidence of this type of arteriovenous shunt in fetuses between 10-18 weeks-gestation, when a presumptive anlage for the coccygeal body may be an arterial plexus derived from a tortuos median sacral artery admixed with nerve fibers and sympathetic ganglion cells ²⁰. However, well-formed coccygeal glomera have been observed for decades in previable fetuses (145-170 mm crown rump length) ²¹.

Postnatally, the glomus coccygeum lies deeply buried in the adipose tissue immediately below or just ventral to the tip of the coccyx, near the center of the natal cleft, in the vicinity of the anococcygeal ligament, between the branches of the median sacral artery and vein, innervated by the pelvic sympathetic plexus ¹⁸⁻²⁰ ²²⁻²⁶ (Fig. 1). Glomus coccygeum, which is endowed of up to 4 arterial segments may be either single ²², binodal, or multiple ²⁴²⁷, or present as a network of glomus bodies in the coccygeal region ¹² ²⁴, with a few of them occasionally located even in coccygeal vertebrae ^{24 25}.

It is one of the largest glomera in humans ^{18 25}, measuring between 1 and 5 mm (mean diameter: 3.5 mm in a large study) ^{22 25}.

Fig. 1. Scanning view of skin and subcutaneous tissue from the sacroccygeal region of a surgical specimen in a 25-year-old woman with pilonidal cyst. Top and center: Natal cleft and cutaneous sinus tract, typical of mostly inactive pilonidal disease, and a 1 mm glomus coccygeum (arrow) with associated vein lying just under the apex of the sinus. Inset: the glomus coccygeum at higher magnification.



The prevalence of glomus coccygeum is uncertain. Its presence was nearly constantly documented in three anatomic autopsy studies as it was found by different investigators in 5 out of 5 ²⁸, in 29 of 32 ²⁶, and in 17 of 20 ²⁴ coccygectomy specimens either from adults ²⁸, elderly ²⁶, or individuals of pediatric and adult ages ²⁴.

Instead, in anatomic studies of specimens submitted for surgical pathology, glomus coccygeum was identified in 13 of 40 (32.5%) coccygectomy specimens resected in individuals with coccygodinia ²⁵ and in 18 of 37 (48.6%) coccygeal bones removed during rectal resection for advanced rectal and uterine carcinomas and for various other reasons ²⁷.

As a normal structure, the coccygeal body is inapparent. In five cases of coccygodynia which had been ascribed to pericoccygeal glomus tumors ²⁹⁻³², which were most likely normal coccygeal glomera, the symptomatology subsided after coccygectomy, but in 3 cases there were radiographic abnormalities of the coccyx or intraoperatively proven fractures. And there are large series of patients treated with coccygectomy for both traumatic and idiopathic coccygodynia in which no remarkable features involving glomus coccygeum were encountered in most of the cases, except for histological degenerate changes in the sacrococcygeal or intercoccygeal discs in some ^{25 32 34}. In clinical practice glomus coccygeum is usually an incidental finding in excised specimens from sacrococcygeal areas for diverse causes, such as pilonidal dis-

ease ³⁵⁻³⁹, coccygeal dimple ⁴⁰, coccygeal pad ^{41 42}, myxopapillary ependymoma ⁴³, tailgut cyst ⁴⁴, sacrococcygeal teratoma ⁴⁵, as well as in surgical specimens from coccygectomies for idiopathic or secondary coccygodinia ^{27 29-} ³², and in perirectal resections including coccyx for rectal or advanced uterine cancers ²⁷. Two cases were observed in infants, in surgical resection specimens performed for pilonidal sinus ³⁷ in a case, including the apex of coccyx, and for coccygeal dimple ⁴⁰ in the other one, respectively. Of interest, in a study based on archival pathology material, glomus coccygeum was found in only 2 cases among 382 pilonidal sinus excision specimens ³⁸.

We incidentally observed several cases of glomus coccygeum during examinations of surgical specimens excised for pilonidal disease as well as for other causes (see illustrations), and the pictures herein presented are from 5 such coccygeal bodies found in 4 young and one elderly individuals.

Histological features

Glomus coccygeum may exhibit a well-circumscribed or multinodular appearance, mainly comprised of small arterioles surrounded by a mantle of epithelioid myoid cells, rich of unmyelinated nerve fibres and embedded in loose to dense fibrous connective tissue.

Due to the variable proportion of the constitutive elements of the glomus body, some authors distinguish the following variants: the glomus cell nodule-dominant type, an intermediate mixed morphology with a mixture of nodules and vessels, and the vascular-dominant type with scattered individual glomic cells²⁶ (Figs. 2-6).

Immunohistochemistry and electron microscopy

Immunohistochemically, the glomic cells of the glomus

body in general are immunoreactive for vimentin, collagen type IV (external basal lamina), muscle specific actin, and alpha-smooth muscle actin (Fig. 2B), and negative for epithelial markers, EMA, endothelial markers (CD31 and FVIII-RA), and neuroendocrine markers, such as chromogranin and synaptophysin, with low proliferative activity ^{25-27 36 38 39}. Conflicting findings have been reported for neuron specific enolase (mostly positive), desmin, and S-100 protein (mostly negative) ²⁵. CD34 was expressed in 3 cases, focally in 2 ³⁸, and diffusely in one ³².

Ultrastructurally, pericellular external lamina, subplasmalemmal vesicles, bundles of actin microfilaments, groups of membrane bound organelles including mitochondria and endoplasmic reticulum can be seen in glomus cells, but no membrane bound endocrine granules have ever been observed ^{27 31 46}.

Histological differential diagnosis

In a compilation of tumoral and pseudotumoral entities of the soft tissues that surfaced over the preceding quarter of a century published in 2006, two of the authors (MB; CAG) made reference to the potential pitfall for the "glomus coccygeum of Luschka"⁴⁷.

In fact, because of its considerable size and multiple anastomotic channels, coccygeal glomus mimics glomus tumor, that is a tumor mostly occuring in distal extremities which was firstly described in 1924⁴⁸ by the French-born, Canadian, histopathologist Pierre Masson (1880-1959). As a matter of facts, P. Masson applied it the name "glomus" ("neuromyoarterial glomus tumor", also called "arterial angioneuromyoma") ⁴⁸ to depict its similarity to the glomus coccygeum of Luschka, hence the term glomus tumor has occasionally been used synonymymously (but erroneously) to refer to a normal glomus coccygeum ³⁷. Parenthetically, glomus tumor was also alternatively

named "glomangioma" in 1935 by the American pa-

Fig. 2. Glomus coccygeum of the so-called "glomus cell nodule-dominant type" (same case as Fig.1). (A) A well circumscribed, unencapsulated glomus body with densely packed clusters of glomic cells, embedded in fibrous connective tissue. While some cell aggregates exhibit slit-like lumina lined by flattened endothelial cells, others are seemingly devoid of vascular lumina. (B) Immunoperoxidase for smooth muscle actin highlighting the glomus cells that surround vessels of the Sucquet-Hoyer canals.



Fig. 3. Glomus coccygeum of the so-called "vascular-dominant type with scattered individual glomic cells", in a 28-year-old man with pilonidal cyst. **(A)** The glomus coccygeum is seen at the level of its arterial pole with branches of the Sucquet-Hoyer canals. The media of the arterioles show in this case a poorly development of the epithelioid cell component. **(B)** Higher magnification. Several unmyelinated nerve fibres are in the immediate vicinity of arterioles and glomic cells.



Fig. 4. Glomus coccygeum of the so-called "intermediate mixed type (nodular and vascular)", in a 15-year-old girl with pilonidal cyst. **(A)** Low power view of a binodal variant of glomus coccygeum with central draining veins. **(B)** Higher magnification of the lower left-sided part of the glomus.



thologist Orville T. Bailey (1909-1998), who believed it represented a subgroup of (hem)angiomas⁴⁹.

Importantly, glomus coccygeum may be easily misinterpreted as a coccygeal glomus tumor by the unwary histopathologist, and in fact a series of glomus coccygeal tumors, presumed to be normal coccygeal glomera, have been reported by several authors ^{29 30 31 50 51 [1 of the 3} cases reported in the last reference of the ones quoted herein]. Bell and collaborators first exposed the potential for confusion ²⁸, and Albrecht and Zbieranowski emphasized the risk of this misinterpretation ³⁵. No additional coccygeal glomus tumors have been published since, and subsequent publications all warned against misdiagnosis and overdiagnosis of this normal microanatomical structure ^{27 36 38 39 43 52}. Therefore, it seems that the glomus body in this specific location is not particularly susceptible to neoplastic transformation.

However, although any coccygeal glomus tumor should be viewed with scepticism, 4 cases of solitary "true" coccygeal benign glomus tumors have been reportd so far in the literature: 2 of the 3 cases by Nutz and Stelzner ⁵¹, and the cases by Llombart et al. ⁵³ and Kim et al. ⁵⁴. These tumors, which were clinically a "palpable lesion", and of 1 to 2 cm in size, exhibited the same morphologic features of glomus tumors occurring in the usual extracoccygeal sites. Benign glomus tumors are expansile lesions, which cause the classic triad of pain, tenderness, and cold sensitivity, sometimes eroding the adjacent bone surface.

There is no record of inherited glomus vascular malformations, malignant glomus tumor, or glomus tumors of uncertain biologic behaviour involving the glomus coccygeum.

Extracoccygeal glomus vascular malformation or glomuvenous malformation (multiple "familial glomangiomas") are either localized (glomangiomatosis) or widespread, with an autosomal dominant pattern of inheritance ^{55 56}. **Fig. 5.** Clomus coccygeum of the "intermediate mixed type", buried within dense fibrous connective tissue. This is an incidental finding from a carcinoma of the lower rectum specimen of a 74-year-old man. **(A)** Sucquet-Hoyer canals with collarettes of glomic cells, emptying in thin-walled venous channels. **(B)** Magnification of the left section in "A" detailing a glomus arteriole and its relevant capaceous thin-walled venule. **(C)** Magnification of the right section in "A" featuring a longitudinally oriented Sucquet-Hoyer canal, connecting a spheroid of epithelioid contractile cells in the media of an arteriole of the glomus body with its relevant venule (visible in "A").



Fig. 6. Clomus coccygeum in a 19-year-old male with trisomy 21, who presented with a longstanding, gradually enlarging "mass" in the intergluteal (natal) cleft region, clinically felt to be quiescent pilonidal disease, but histologically diagnosed as coccygeal pad (juvenile coccygeal fibrosis). (A) Skin showing orthokeratotic epidermis and massively expanded dermis with haphazardly oriented, variably thick collagen bundles. (B) Deep dermis with increased dermal mucin, and fibrotic subcutaneous tissue. (C) Buried in a collagenous matrix is a glomus coccygeum of the "glomus cell nodule-dominant type", incidentally found very close to the deep margin of resection.



Generally, malignant glomus tumors ⁵⁵ are deeply seated, measure 2 cm, exhibit atypical mitotic figures, moderate to high nuclear grade, and at least 5 mitotic figures/50 high power fields. Glomus tumors of uncertain biologic behaviour ⁵⁷ exhibit high-mitotic activity and are superficially located, or of large size only, or deep seated only. No histological variants of glomus tumors, either with oncocytic features ⁵⁸ or symplastic high grade nuclei ⁵⁷, have been reported in the coccygeal region.

Other entities which may possibly enter the differential diagnosis with normal glomus coccygeum are the following: intradermal melanocytic nevus, paraganglion and paraganglioma, neuroendocrine tumor, adnexal skin tumor, and metastatic carcinoma. Intradermal melanocytic nevus is less organoid and immunohistochemically is strongly positive for S-100 protein and negative for alpha-smooth-muscle actin.

Paraganglioma (paraganglioma of the filum terminale might be in point) is a tumor arising from the paraganglion system. If carefully examined, it looks morphologically different, is positive for argyrophilic stains and immunoreactive for neuroendocrine markers, and shows typical neuroendocrine granules when ultrastructurally examined. Although the glomus coccygeum is not related to the paraganglia, with which it may be confused, we would like to alert the reader about the incorrect use, in the older literature ⁵⁹ as well as among clinicians of some discipline even today, of the name "glomus" for other dif-

ferent microanatomical chemoreceptorial structures of the paraganglion system (glomus caroticum, glomus jugulare, glomus tympanicum, ...), which erroneously glomus coccygeum one and a half century ago was ascribed to. Adnexal skin tumors (mainly eccrine acrospiroma), neuroendocrine tumors, and metastatic carcinomas have different morphologies and can be easily excluded with the support of immunohistochemistry, mainly evidencing their consistent cytokeratin immunoreactivity.

Conclusions

TAKE-HOME-MESSAGES:

- It is surprising how little attention is devoted to glomus coccygeum in standard textbooks of human anatomy and histology.
- It is a normal, likely phylogenetic vestigial anatomical structure with similar cytoarchitectural constituents as the dermal Sucquet-Hoyer canals.
- Its functional significance is uncertain, even enigmatic, given its ectopic location in the hypodermis.
- Its constituents seem to be disinclined to pathological alterations such as hyperplastic and neoplastic proliferation.
- In summary, familiarization with this seldom observed microanatomical structure of the coccygeal region is necessary to avoid overinterpretation as a pathological one.

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