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The Complete Second Branchial cleft fistula –

A case report:

Abstract.

Branchial anomalies compose approximately 30% of congenital neck mass and present as cyst, sinus or fistula and among them second branchial cleft and pouch anomalie are commonest, but complete second branchial cleft anomalie with external and internal opening is rare. We present three case report of complete branchial fistula, which was confirmed by fistulogram and was managed by complete excision of fistula through transcervical and transoral approaches. We present technique of excision of fistula with help of use of intraoperative catheter and dye.

Introduction.

The Branchial apparatus was first described by Von Baer (1827).The term branchial fistula had been applied by Simpson (1969) [1]. Branchial fistula is formed due to persistence of the embryonic second branchial cleft. Branchial anomalies compose approximately 30% of congenital neck mass and present as cyst, sinus or fistula[2, 3]. They are equally common in males and females and usually present in childhood or early adulthood [2]. Cysts are remnant of the cervical sinus without an external opening. Sinuses are persistence of the cervical sinus with external opening, whereas fistula also involves persistence of the branchial groove with breakdown of the branchial membrane resulting in a pharyngocutaneous fistula [2].

Second branchial cleft anomalies are the most common, representing 95% of all branchial cleft malformation [2]. The evaluation of this lesion begins with complete history and physical

examination which mainly reveals a congenital opening on the lower neck, anterior to sternomastoid muscle with recurrent clear to mucoid discharge from opening, tract may end blindly forming sinus or may extend up to tonsillar fossa forming complete fistula [3]. Sinogram can be performed to define the track. The most accepted modality of treatment is complete excision of fistulous tract by combine transoral and transcervical approach [4]

Case report.

17 year old male patient came with chief complaints of opening in lower part of neck (fig:1) since birth and recurrent mucoid to mucopurulent discharge form opening since age of 6 year which was mainly associated with upper respiratory tract infection. Discharge from opening get relieved after oral antibiotics. On physical examination there was small punctum in the skin at junction of middle two third and lower one third of anterior border of right sternocleidomastoid muscle. When patient presented there was no active discharge from punctum but on pressure above the opening few drops of mucoid discharge were milked out.

Ear nose throat and general examination was normal. There were no other congenital anomalies. Full blood count and urine examination was normal. On the basis of clinical history and physical examination clinical differential diagnosis were made as 1) second branchial cleft sinus or fistula, 2) third branchial cleft fistula.

Sinogram was performed which demonstrated that sinus tract extending up to oropharynx and there was spilling of dye in tonsillar fossa, suggestive of complete second branchial cleft fistula (fig: 2).

Anesthetic fitness was taken and patient was posted for total excision of fistula under general anesthesia. Surgical plan was made which included transcervical and transoral approach with stepladder technique for complete excision of tract. To facilitate the dissection of fistula, cannula was passed through fistula and then methylene blue dye was injected through cannula (fig: 3). Elliptical incision was taken around the fistula over neck, and blunt dissection was carried out through subplatysmal plane along the carotid sheath up to level of carotid bifurcation (fig: 4). Then second incision was made at level of hyoid bone, then tract with cannula were pulled through second incision to facilitate further higher dissection (fig: 5). The tract was separated from the surrounding soft tissues superiorly beneath the stylohyoid muscle

and digastric muscle; tract was going medially in the parapharyngeal space toward the tonsillar fossa between the internal carotid and external carotid artery after crossing the loop of hypoglossal nerve. Then Boyle-Davis mouth gag was applied to visualize Oropharynx, there was leak of dye from right lower pole of tonsillar fossa. As there was no tonsillar tissue, dissection was done around the fistulous tract in the tonsillar fossa and parapharyngeal space (fig: 6). Complete fistulous tract with cannula was pulled through neck, which was of 11.5 cm in length (fig: 7). Oropharyngeal defect was closed with help of 3.0 catgut and both neck wound was closed with help of 3.0 mersilk suture and 3.0 catgut suture. Drain was kept in neck. Postoperative period was uneventful. For initial three days patient was kept nil by mouth and feeding was done through nasogastric feeding tube according to weight of patient. Antibiotics and mouth gargles (betadine and hydrogen peroxide) was given for seven postoperative day.

At seventh postoperative day skin sutures was removed and oropharynx was examined for any defect, as skin sutures and Oropharynx mucosa was healthy, patient was discharged on seventh postoperative day. Patient was kept on follow up on 15^{th}, 45^{th} day and after 3 months. The postoperative follow up was uneventful.

Histopathological report confirmed branchial fistula tract lined with pseudostratified ciliated squamous epithelium.

Discussion.

Pharyngeal arches are arches of mesnchyme derived from paraxial and lateral plate mesoderm and neural cell appear in 4^{th} and 5^{th} week of development. They are covered externally by ectoderm, which forms cleft between successive arches and internally by endoderm which forms pouches between arches. The branchial apparatus consists of six paired mesodermal arches, separated by invagination of endoderm on the inside and ectoderm on the outside known an as pharyngeal pouches and branchial cleft respectively [5]. Their anomaly of development result into cyst, sinuses and fistulae. Pharyngeal arches play a role in the formation of face, ear and neck.

Branchial anomalies can be lined with either respiratory or squamous epithelium. Cyst often lined by squamous epithelium, whereas sinus and fistula are more likely to be lined with ciliated columnar epithelium [2.] Branchial cleft cysts occur three times more often than branchial fistulas [4].

Second arch anomalies are classified into 4 types. Type1. Lesion lies anterior to the sternocleidomastoid muscle and do not contact the carotid sheath. Type2. Lesion are the most common and pass deep to the sternocleidomastoid and either anterior or posterior to the carotid sheath. Type3. Lesion passes between internal and external carotid arteries and are adjacent to pharynx. Type4. Lesion lies medial to carotid sheath close to the pharynx adjacent to the tonsillar fossa [2].

The second branchial cleft and sinuses are encountered along the anterior border of the sternocleidomastoid muscle in its lower third and may be bilateral [3, 6]. The complete branchial fistula with external and internal opening is rare [7]. Preoperative imaging of tract with contrast material demonstrate the entire course of the tract, aid into surgical planning, differentiate between sinus and fistula, and minimise the chance of recurrence [4]. Some time complete tract cannot be demonstrated because it may be blocked by secretion and granulation. Anatomically fistulous tract passes deep to Platysma muscle between second and third pharyngeal arch structures by ascending along the carotid sheath and passing medially between internal and external carotid arteries above the glossopharyngeal nerve and below the stylohyoid ligament [5]. The fistula may open into pharynx, usually into tonsillar fossa.

It can present at any stage, more commonly in the first and second decade of life. Two to ten percent of them can be bilateral. 6% of the patient with complete fistula can have a family history of branchial fistula anomalies [7]. Branchial cysts are more common (80.8%) than branchial fistulae. Some patient may also have conductive or sensory neural deafness as well as other anomalies of the first and second arch derivatives.

The definite treatment of branchial anomalie is complete surgical excision of tract, most suitable age for surgery is 2 to 3 year or as early as possible if it is already delayed and among surgical techniques stepladder incision is most accepted method [7, 4], as it provide better visualisation of tract near pharynx which is combined with transoral approach for complete excision of tract, other modality of treatment includes a) Sclerosing agents which is seldom used today due to the associated inflammatory reaction and the risk of necrosis with perforation into the pharynx, b) stripping method was described by Taylor and Bicknell in 1977 [8], but this has not been widely used due to great risk of damage to adjacent structures.

Complication of surgery includes recurrence, which could be 3% in fresh cases [7] and up to 20% in second surgical attempts. Other complication include secondary infection, injury to facial, hypoglossal, glossopharyngeal, spinal accessory nerves, injury to internal jugular vein, and hematoma formation.

The recurrence rate following surgery vary up to 3% been reported, this is due to incomplete surgical excision.

In our case 17 year old patient presented with classical history of opening in lower part of neck since birth and recurrent discharge, provisional diagnosis was made as sinus in lower part of neck of branchial anomalie which was further evaluated with sinogram which confirm diagnosis of complete branchial fistula, before performing sinogram intravenous antibiotic was given for three days to reduce any infection, adhesion, or granulation around the tract to demonstrate entire course of fistula. Complete excision of fistula tract was performed under general anaesthesia via transcervical and transoral approach, in our case we cannulated the fistula and injected methylene blue dye through the cannula, as cannula provide firm base for dissection and dye helps in differentiating fistula from surrounding tissue. Postoperative period was uneventful and patient was asymptomatic in his successive follow-ups.

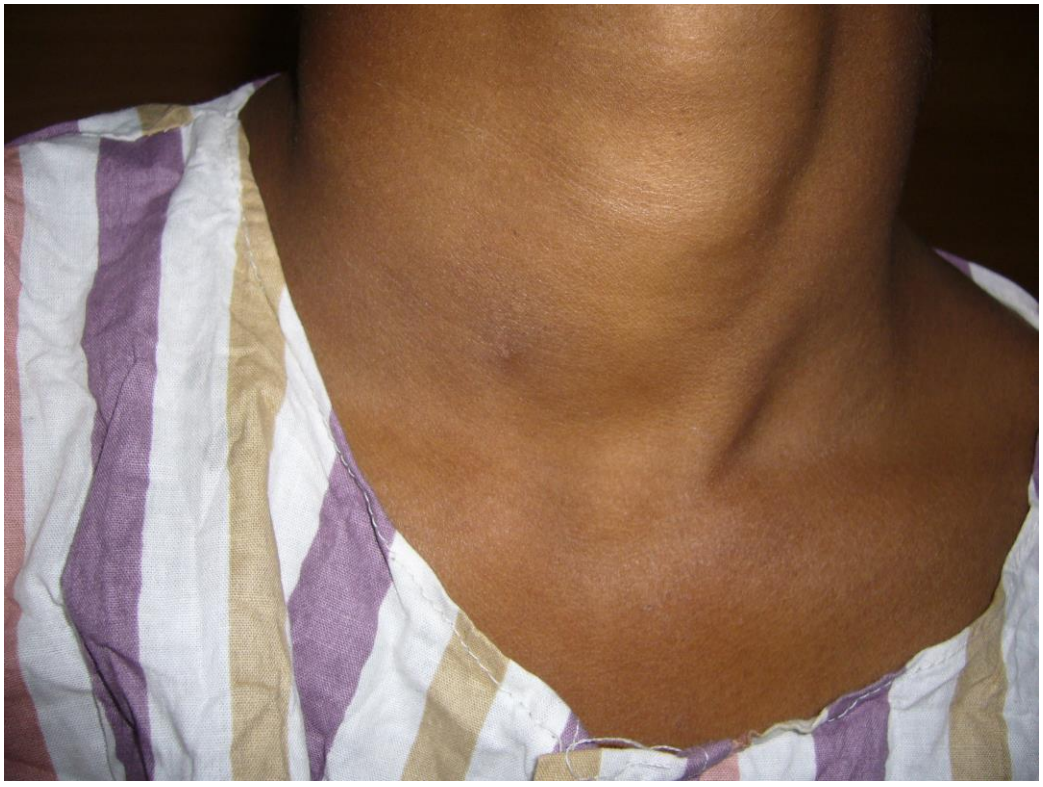

Figure 1 showing clinical photograph of the patient

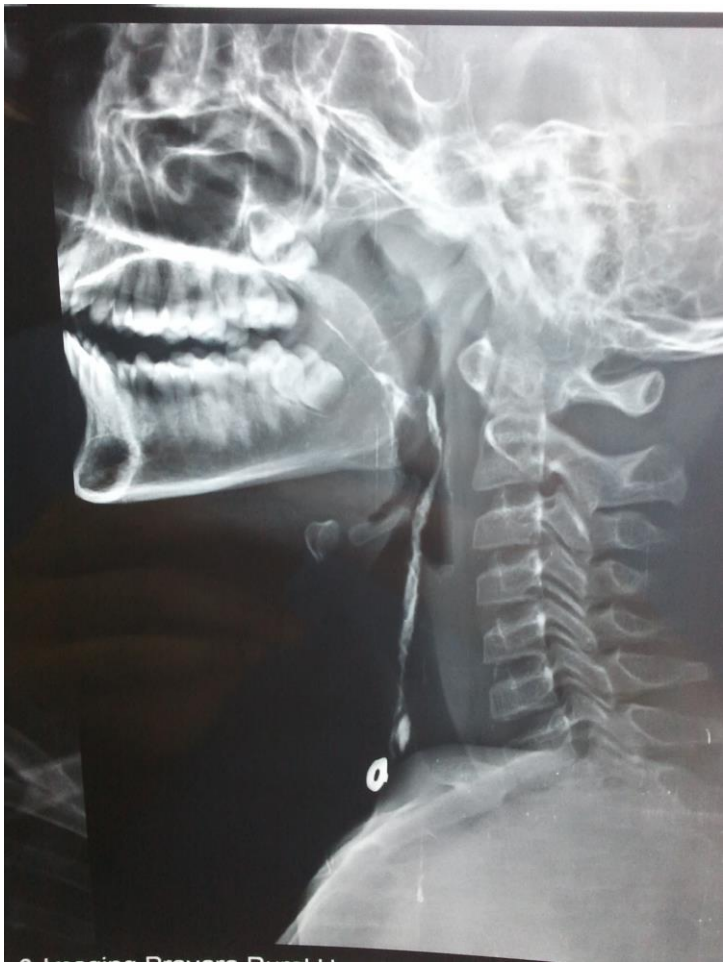

Figure 2 Showing Sinogram

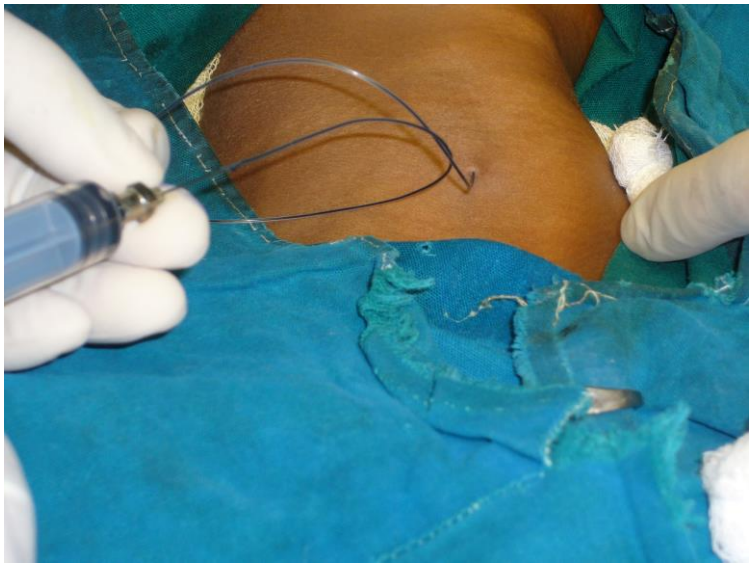

Figure 3 showing methylene blue being injected into the fistula

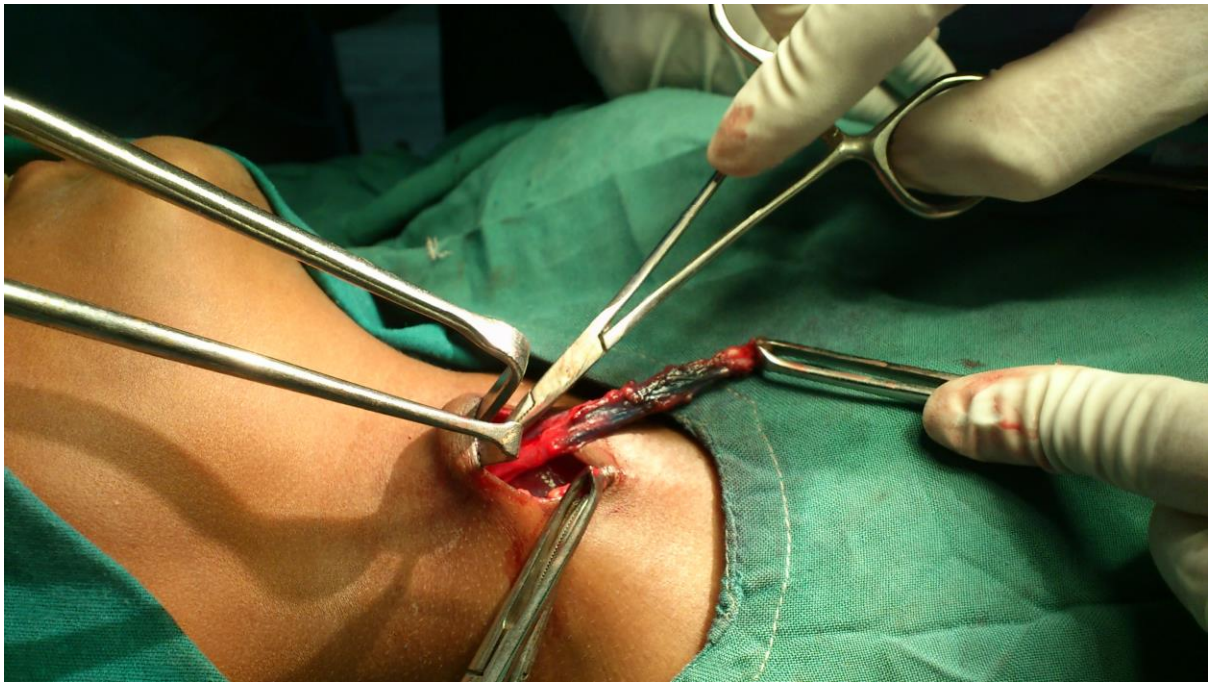

Figure 4 showing exposure of the fistulous tract

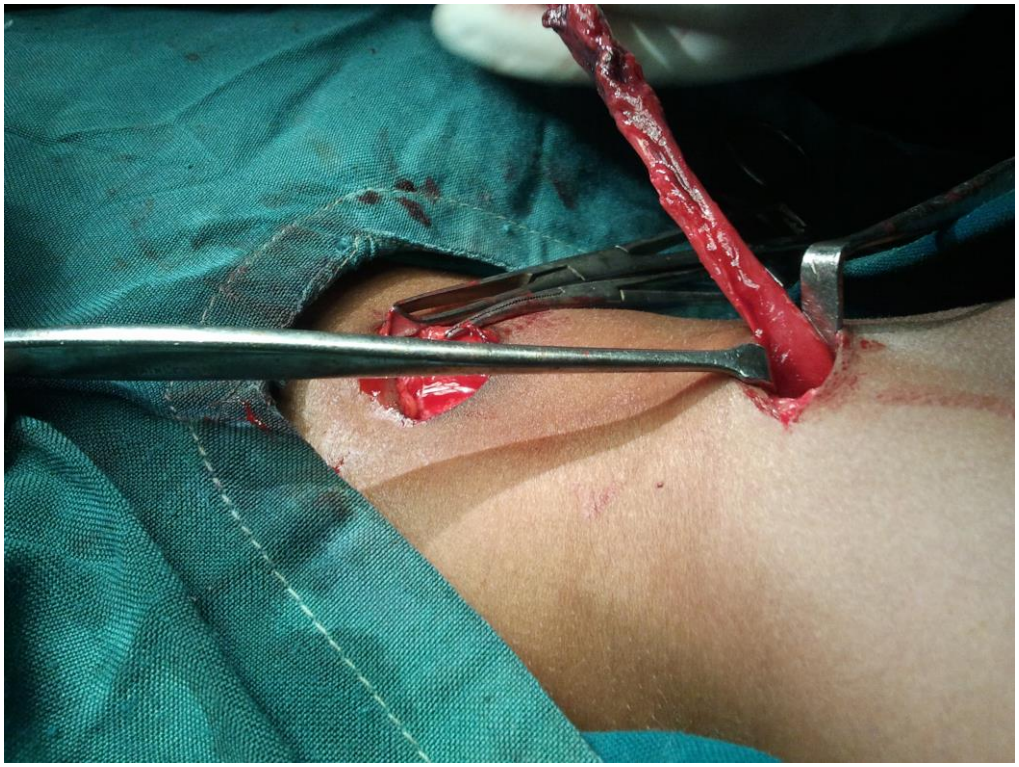

Figure 5 showing the second incision at the level of hyoid bone

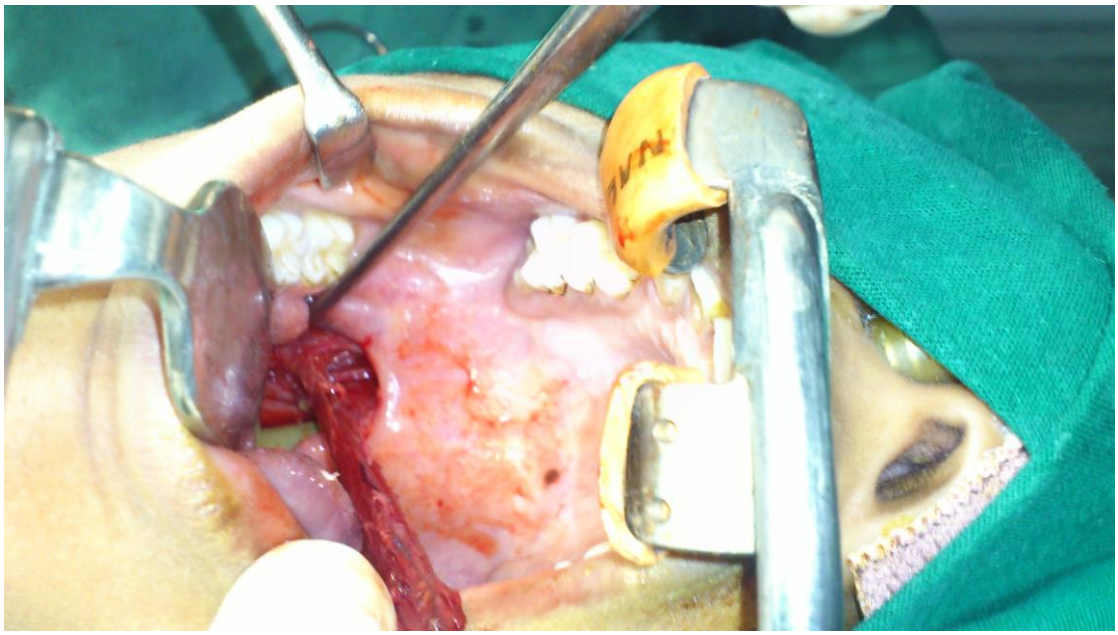

Figure 6 showing dissection at the level of tonsillar fossa

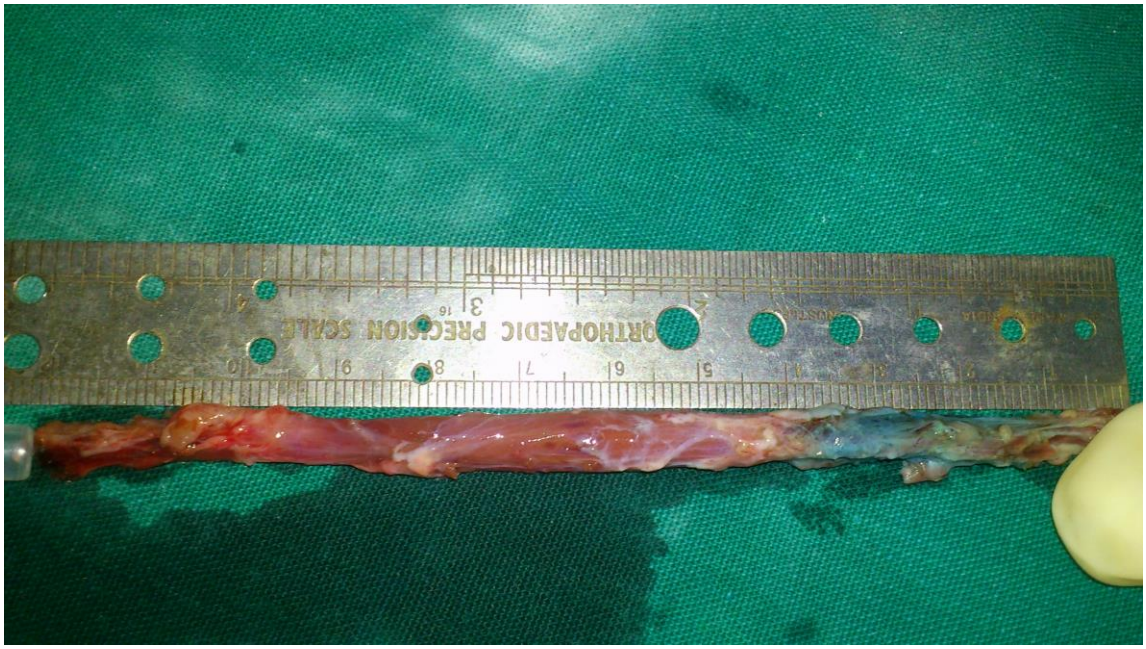

Figure 7 showing the dissected specimen

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