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INFRASELLAR CRANIOPHARYNGIOMA

Case report

Asdrubal Falavigna[1], Jorge Luiz Kraemer[2]

ABSTRACT - We report a case of infrasellar craniopharyngioma in a 34 year-old woman who presented with progressive headache and diplopia. Computed tomographic and magnetic resonance images showed a heterogeneous tumor originating from the sphenoid bone with ethmoid sinus and sella turcica extension. A sublabial rhinoseptal transsphenoidal surgery was performed. Craniopharyngiomas with infrasellar development are very rare. Infrasellar craniopharyngioma is uncommon, thirty-five cases has been reported in literature. The embryology, clinical features and radiographic investigation of these tumors are discussed.

KEY WORDS: craniopharyngioma, sphenoid sinus, skull base tumor.

Craniofaringioma infra-selar: relato de caso

RESUMO – Relatamos um caso de craniofaringioma infra-selar em uma paciente de 34 anos com sintomas de cefaléia e diplopia. A investigação radiológica com tomografia computadorizada e ressonância magnética de encéfalo demonstrou um tumor heterogêneo localizado no osso esfenoidal e com extensão para o seio etmoidal e sela turcica. Realizada cirurgia pela via transesfenoidal. A ocorrência de craniofaringioma com topografia infra-selar é incomum, havendo relato na literatura de 35 casos. A literatura é revisada, sendo discutidas a embriologia, a apresentação clínica e características radiológicas do tumor.

PALAVRAS-CHAVE: craniofaringioma, seio esfenoidal, tumor de base de crânio.

Craniopharyngioma is a rare tumor[1]. It arises within the sella turcica and expands mainly into the suprasellar region[2]. Occasionally, the tumor can occur without sellar involvement. These tumors rarely extend below the sellar floor into the sphenoid sinus or invade the pharynx and the nasal cavities[3-34]. The infrasellar craniopharyngioma may then originate anywhere along the tract of the obliterated craniopharyngeal duct, which would include the sphenoid bone, vomer, and nasopharynx[35,36].

In this study we report a case of infrasellar craniopharyngioma with the epicenter situated in the sphenoid sinus and review the 35 other reported cases in the literature of this unusual localization since 1924.

CASE

A 34 years-old woman presented with frontal headache with progression of 7 months and diplopia that appeared in the last month. Clinical evaluation revealed left abducens nerve palsy. No signs and symptoms of pituitary dysfunction were evident. The laboratory studies, including hypophyseal function were normal.

Coronal computed tomographic (CT) scans after intravenous contrast showed a large sphenoid sinus heterogeneous mass with destruction of the ethmoid sinus and sella turcica (Fig 1). T1-weighted (plain and gadolinium-enhanced 600/11/2 [repetion-time/echo time/excitations]) and T2-weighted (3000/108/1) magnetic resonance showed isointense image (Fig 2). Post-contrast the brightly enhancing, heterogeneous mass was clearly visible in the sphenoid bone (Fig 3). There was extension of the tumor to ethmoid sinus and to the sella turcica. Carotid and vertebral angiography revealed that the tumor was avascular.

Sublabial rhinoseptal transsphenoidal surgery was performed. After removal of the anterior wall of the sphenoid sinus, a solid firm mass was filled the sphenous sinus. The circumferential wall of the tumor was firmly attached to the cavernous sinus, with profuse bleeding during the maneuver. A subtotal removal was performed. At the end of the tumor resection, posterior pharyngeal packing was left in place. Her postoperative course was good

[1]Professor Assistente da Disciplina de Neurologia da Faculdade de Medicina da Universidade de Caxias do Sul, Pós-Graduando em Neurocirurgia da Universidade Federal de São Paulo - Escola Paulista de Medicina, São Paulo SP, Brasil (UNIFESP-EPM); [2]Professor do curso de Pós-Graduação em Cirurgia da Universidade Federal do Rio Grande do Sul (UFRGS) e da Fundação Faculdade de Ciências Médicas de Porto Alegre (FFCMPA-ISCMPA), Mestre pela UFRGS e Doutor em Neurocirurgia pela UNIFESP- EPM.

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Dr. Asdrubal Falavigna - Rua Dr. Moreira César 271/sala.1 - 95034-000 Caxias do Sul RS - Brasil. E-mail: asdrubal@doctor.com

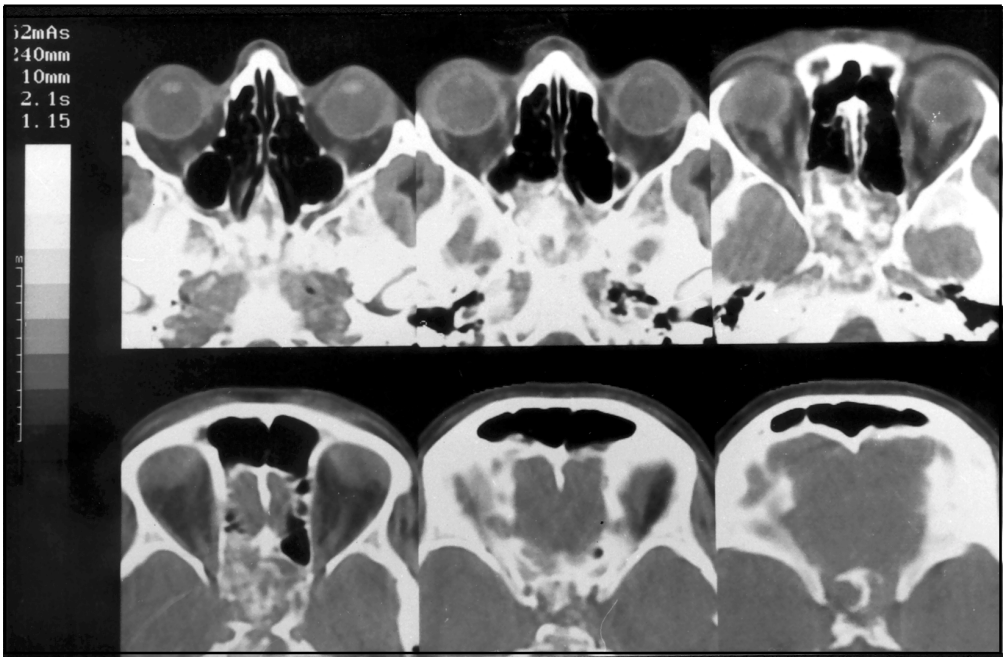

Fig 1. CT enhanced axial view showing a large heterogeneous mass arising from the sphenoid bone with destruction of the ethmoid sinus and sella turcica.

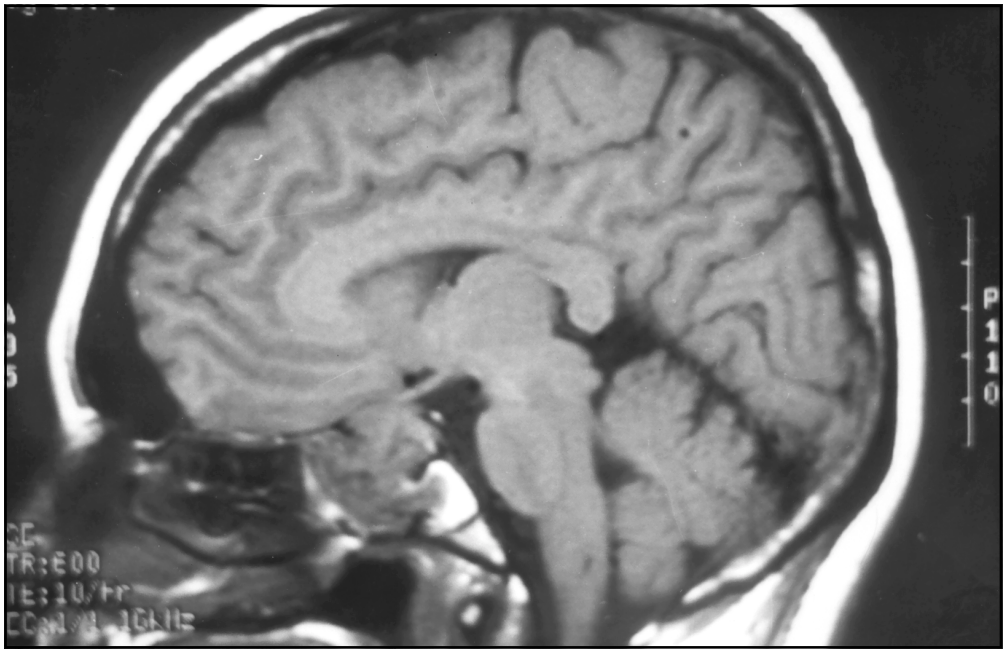

Fig 2. Sagittal magnetic resonance in T1-weighted image without gadolinium administration showing an isointense tumor in the sphenoid bone with extension to the sella turcica.

and the nasal packing removed on the third postoperative day. The patient was discharged from the hospital on the eight postoperative day with recovery of the abducens nerve palsy and headache-free. Histologic examination revealed an adamantonomatous craniopharyngioma (Fig 4).

The patient was referred for radiotherapy. Conformational radiotherapy consisting of a total of 55Gy (30 fractions) was administered 1 month later. At clinical follow-up, 2 year after irradiation, her neurological examination and hormonal studies were normal.

DISCUSSION

Craniopharyngiomas are benign but aggressive epithelial neoplasms, which comprises approximately 3% of all intracranial tumors[1]. They are generally found intracranially with a similar frequency in children and adults but with a slight preponderance in children between 5 and 15 years of age[2]. Equal sex incidence has been noted in groups of children[2]. Craniopharyngiomas are most commonly located extraaxially in the sellar or suprasellar area in 90% of cases[2]. They can extend to the anterior (2-5% of cases), middle (2%), or posterior (1-4%) cranial fossa, and infrasellar extension is found in about 5% of cases[3,4]. Rarely, craniopharyngiomas arise primarily in unusual locations, such as the nasopharynx, sphenoid bone, third ventricle, pineal gland, sylvian fis-

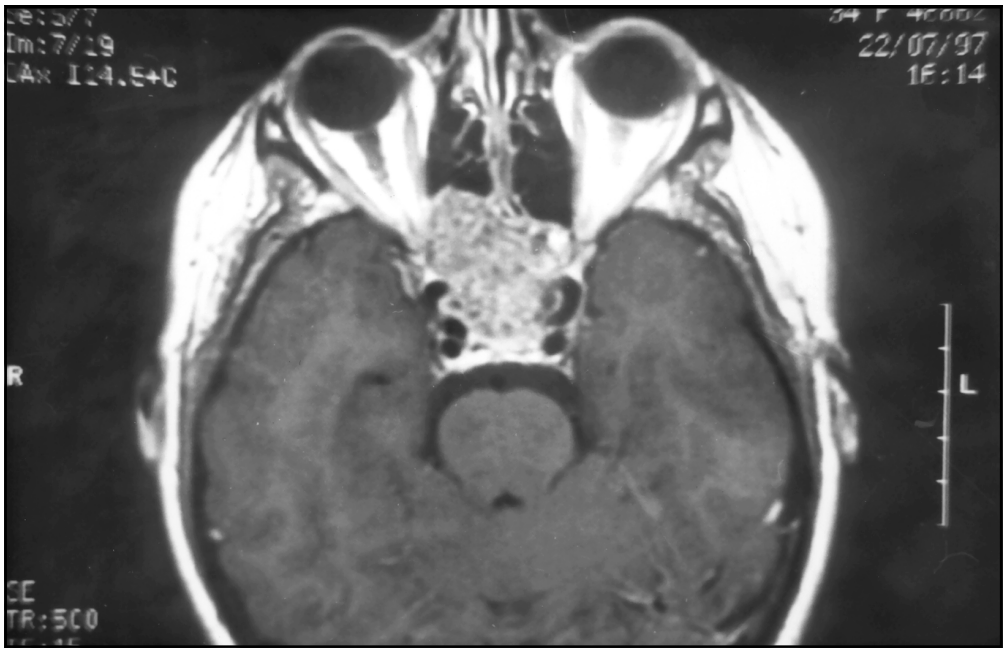

Fig 3. T1-weighted gadolinium enhanced axial MRI scans, showing a heterogeneous enhancing sphenoid sinus mass with extension into the ethmoid sinus.

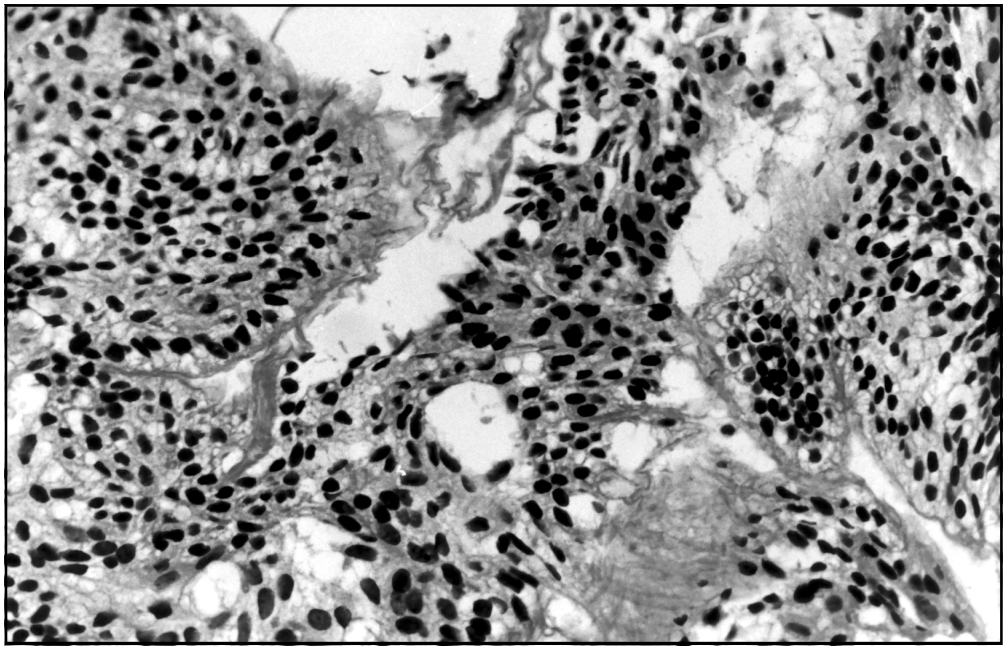

Fig 4. Photomicrography of adamantinomatous craniopharyngioma showing a epithelial component and peripheral palisading cells (hematoxylin and eosin, original magnification x 125)

sure and cerebellopontine angle[3-34,37-43]. The growth of a craniopharyngioma in the infrasellar region is extremely rare, only 36 cases were found in the literature (Table 1).

The infrasellar craniopharyngioma reported in literature has a similar sex distribution, being 3 times more frequent above 15 years of age. The microscopic appearance of most craniopharyngiomas shows an external layer of high columnar epithelium, a variable amount of polygonal cell, and a central network of epithelial cells. Two clinicopathologic varieties have been delineated microscopically: the papillary squamous type, which is seen almost exclusively in adults and carries a better prognosis; and the classical adamantinomatous variant, which develops mainly in children and has a worse overall outcome[41].

Treatment of these tumors is mostly excisional, through a craniotomy or transsphenoid approach. Complete removal of the tumor is the preferred procedure[2,29]. When vital structures are involved and excision is compromised by significant risks of morbidity and mortality, subtotal removal of the tumor followed by supplemental radiotherapy is the pre-

ferred treatment[29]. Radiotherapy, once considered ineffective, is currently the most often used adjuvant treatment. Craniopharyngiomas have been found to be radiosensitive, and radiotherapy plays a major role in preventing recurrence and improving survival[42].

Embryology - According to Warwick and Williams[43], during the fourth week of gestation an ectodermally lined diverticulum (Rathke's pouch) develops in the roof of the stomodeum just anterior to the oropharyngeal membrane. This pouch ascends cranially traversing the mesenchyme to meet the neuroectoderm of the infundibulum (neurohypophysis), which descends as a neural outgrowth from the floor of the third ventricle (diencephalon) of the embryonic brain. Rathke's pouch differentiates into the anterior lobe of the pituitary, the adenohypophysis. This course traversed by Rathke's pouch forms a cord of cells joining the stomodeal ectoderm to the future adenohypophysis in embryo. Later in embryonic life this cord disintegrates, leaving an obliterated craniopharyngeal canal. It is a tract that runs from the anterior part of the hypophyseal fossa of the sphenoid bone to the junction of the posterior septum of the nose with the palate, which is the stomodeal end of the recess. The pharyngeal hypophysis (functioning adenohypophyseal tissue), which remains in adults, is a caudal remnant of this cord.

Types of origin - There are several theories regarding the origin of infrasellar craniopharyngioma. Most of them are based on the embryologic development of the adenohypophysis. Mott and Barret[35] in 1899 were the first to postulate that these tumors might arise from the remnants of the pharyngeal hypophysis. Erdheim[36] proposed in 1904 that the craniopharyngioma originated from the remnants of the obliterated craniopharyngeal duct, suggesting that these tumors can arise anywhere along the tract of migration of Rathke's pouch from the vomer, the roof of the nasopharynx, through the midline sphenoid bone beneath the floor of the sella turcica.

Clinical presentation - Symptoms and clinical findings are related to the craniopharyngioma's localization and mass effect with compression of the surrounding structures (Table 1). The symptomatology of suprasellar tumor is often characterized by defects in the visual fields (bitemporal or homonimus hemianopsia), varying signs of pituitary insufficiency (diabetes insipidus, amenorrhoea, diminished libido and cachexia), and the symptoms of increased intracranial pressure (headache, vertigo and cranial nerve deficit) that occur relatively late in the course according to Carmel et al.[2] and Sato quoted by Fujitani et al.[16]. The signs of pituitary dysfunction appear early in the cases in which the tumor expands within the sella. The tumor located at the sphenoid sinus usually presents with headache and cavernous sinus syndrome[6-8,11,12,15,17,20,23,24,27]. The craniopharyngiomas located in the nasopharyngeal region usually present with frontal headache, nasal obstruction, epistaxis, nasopharyngeal and/or nasal fossa masses[3,5,9,10,13,15,18,19,22,25,26,29,32,34]. Our case had symptoms of headache, diplopia due to left abducens nerve palsy, without pituitary insufficiency, which suggests that the tumor had appeared in the sphenoid bone and, later, extended to the sella turcica region.

Localization - Craniopharyngiomas usually originate intracranially, grow along the pituitary stalk, on the axis of the sella-infundibulum and located below the brain, above the pituitary, behind the optic chiasma, and within the circle of Willis. Occasionally there is an intrasellar one and, rarely, one within the body of the sphenoid or nasopharynx in the tract of the former craniopharyngeal canal proper. Since 1924, 35 cases of infrasellar involvement by craniopharyngioma have been described in the medical literature. In these cases the tumors are situated at the nasopharyngeal region, the sphenoid sinus, the maxillary sinus and usually involved the sella turcica and the supra sellar region. The most common location of infrasellar craniopharyngioma has been the sphenoid sinus either alone, 4 cases, or combined with other sites, 28 cases (Table 1). The tumor location in the case presented could have been extensions from the sella turcica or derivatives of pharyngeal canal remnants. In the former situation, the tumor could be a typical craniopharyngioma that arose in the sella turcica and then went downwards into the sphenoid sinus. In the latter, the tumor originated in the infrasellar region, sphenoid sinus, and then grew extradurally and superiorly. We suspect that this tumor was a craniopharyngioma that originated in the infrasellar region because it is situated mainly in the sphenoid sinus, the pituitary hormones were always normal and we could not find the dura mater of the sellar floor during the transsphenoidal approach.

Diagnostic evaluation - Skull x-ray study, tomograms, and CT were the most common radiographic techniques employed in evaluation. Infrasellar craniopharyngioma cases had the plain skull x-rays and

Table 1. Infrasellar craniopharyngiom.

Author	Year	Patient Age/Sex	Tumor Location	Symtoms & Signs	Radiographic Investigation
Boch (3)	1924	51/M	NPX	Headache, nasal obstruction	Skull x-ray (Necropsy)
Zeitlin (4)	1935	40/M	SS, ST, middle fossa (L), petrous bone (L)	Difficulties in phonation, face pain (L), anisocoria, internal rectus muscle palsy (R), ptosis (R), loss or decreased vision, facial paralysis (L), tongue deviated to left side	Necropsy
Drummond (5)	1938	14/F	NPX, SS, ST	Nasal obstruction, headache, loss or decreased vision, NPX and NF mass	Skull x-ray
Sato acc. Futitani et al. (16) 6)	1944	56/F	NPX, SS, ST, S	Visual loss	Skull x-ray
Northfield (6)	1957	54/F	SS	Headache, vomiting, homonymous, hemianopia (L)	Skull x-ray
Hamberger et al. (7)	1960	25/M	SS	Headaches, face hypoesthesia in territory of infraorbital nerve (R)	Skull x-ray
Johnson (8)	1962	39/M	SS, ST, S	Headache, diplopia, vision loss (R)	Skull x-ray, tomogram
Podoshin et al. (9)	1970	15/F	NPX, SS, ST	Nasal obstruction, NPX and NF mass	Skull x-ray
Isayama (11)	1970	62/F	SS, ST, S	Facial pain, diplopia	Skull x-ray
Trible (13)	1970	71/M	NPX, SS, ES, ST	Headaches, diplopia, nasal obstruction, epistaxis	Skull x-ray
Cooper & Ransohoff (14)	1972	16/M	NPX, SS, S (middle fossa)	Headache, epistaxis, CSF rhinorrhea, diplopia, lack of hearing (L), palpable mass in the zygomatic area (L)	Skull x-ray
Prasad & Kwi (10)	1975	55/F 48/M	NPX, SS NPX, SS, ST	Nasal obstruction, NPX and NF mass Nasal obstruction, headache, diplopia with lateral rectus weakness (L), facial hypoesthesia in territory of infraorbital nerve (L), NPX and NF mass	Skull x-ray, tomogram, angiogram Skull x-ray, tomogram
Ishiyama (12)	1977	41/F 25/M	SS, ST, S SS	Facial pain Headache, diplopia	Skull x-ray, tomogram Skull x-ray, tomogram
Illum et al. (15)	1977	14/F	NPX, SS, ST	Headache, diplopia with abducens nerve palsy (L), temporal hemianopsia (L)	Tomogram, angiogram, pneumo-encephalography
Majlessi et al/ (18)	1978	17/M	NPX, SS, ST, S	Headache, bitemporal hemianopsia, exophthalmos, hypopituitarism, NPX mass	Skull x-ray

Fujitani et al . (16)	1979	18/F	NPX, SS, ES, MS, orbit (R)	Exophthalmos, visual disturbance	Skull x-ray, angiogram, scintigraphy, cystography CT
Pheline et al. (20)	1981	12/M	SS	Headache, diplopia with abducent nerve palsy	Skull x-ray, angiogram pneumogram
Mukada et al. (21)	1984	13/M	NPX, SS, ST, S	Visual disturbances, panhypopituitarism	Skull x-ray, tomograms, CT, angiogram, pneumogram
Lewin et al (20)	1984	27/F	NPX	Epistaxis, NPX mass	Skull x-ray, tomogram, angiogram, CT
Maier (23)	1985	77/M	NPX, SS, ST	Headache, CSF rhinorrhea	Skull x-ray, tomogram
Maiuri et al. (22)	1987	25/F	NPX, SS, ST	Headache, nasal obstruction	Skull x-ray, tomograms, CT
Hillman et al. (24)	1988	64/M	SS, ST, S	Visual disturbances, abducent nerve palsy (L)	CT, MRI
Benitez et al. (26)	1998	29/M	NPX, SS, ES	Nasal obstruction, pressure in the nose and behind the eyes	Skull x-ray, CT, MRI, angiograms
Ortiz et al. (25)	1998	20/M	NPX	Epistaxis	CT
Pharaboz et al. (26)	1989	27/F	NPX, SS	Nasal obstruction	CT, MRI
Akimura et al. (28)	1989	12/F	NPX, SS, ES, ST S, MS	Visual disturbance	CT, MRI, angiogram
Byrne & Sessions (29)	1990	29/M	NPX, SS, ES, MS	Nasal obstruction, NPX na NF mass	Skull x-ray, CT, MRI
Gili & Garcia (17)	1991	9/M	NPX, SS	Nasal obstruction, diplopia with abducent nerve palsy (R)	Skull x-ray, CT, MRI
Bret & Beziat (30)	1993	16/F	NPX, SS	Nasal obstruction	CT, MRI
Sener ((34)	1994	8/M	NPX, SS, ES, S	Headache, decreased vision, nasal obstruction	CT, MRI
Kanungo et. al (32)	1995	40/F	NPX, SS, S	Headache, nasal obstruction	Skull x-ray, CT, MRI
Cheddadi et al. (31)	1996	Premature/F	NPX	Respiratory insufficiency	Nasal endoscopy, CT
Jiang et al. (33)	1998	7/M	ES	Epistaxis	CT
Chakrabarty et al. (40)	1998	46/M	NPX, SS, ST	Nasal obstruction, double vision	CT, MRI

NPX, nasopharynx; SS, sphenoid sinus; ST, sella turcica; S, suprasellar; ES, ethmoid sinuses; MS, maxillary sinus; NF, nasal fossa; CSF, cerebrospinal fluid; MRI, magnetic resonance imaging; CT, computed tomography; [L], left; [R], right.

tomograms that showed sinus opacification, tumor expansion, enlarged sella turcica, cystic lesions, fluid level, bony erosions, and calcifications[6-10,13-22,27,30].

Computed tomography in the cranial base craniopharyngioma cases reported usually reveals the heterogeneous nature of the tumor with its solid and cystic components, calcification, multicysts, lyctic lesions, irregular enhancement. Multiple calcifications are observed within the tumor, especially in younger patients. In adults, craniopharyngiomas are often not calcified[17,19,21-23,26,27,29,30,32,35,40]. Magnetic resonance image clearly showed the tumor extension, cystic portions, mixed intensity signal, inhomogeneous or heterogeneous enhancement[17,23,25-30,32,34,40].

CONCLUSION

Craniopharyngiomas with infrasellar development are very rare. Up to now, thirty-five cases has been reported in literature since 1924. The most common location of infrasellar craniopharyngioma has been the sphenoid sinus either alone or combined with other sites. The infrasellar craniopharyngioma may then originate anywhere along the tract of the obliterated craniopharyngeal duct, which would include the sphenoid bone, vomer, and nasopharynx.

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