The Surgical Treatment of Choroid Plexus Papillomas

The results of 27 years experience

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Choroid plexus papillomas are uncommon tumours, their incidence ranging from 0.4 to 0.6% of brain neoplasms (1, 6, 8, 27, 36, 42). They are usually histologically benign. Therefore operation appears to be the most appropriate treatment for these lesions.

A considerable number of publications have been devoted to these neoplasms. Nevertheless, because of their rarity, only a few surgical series include a sufficient number of operated cases to permit analysis of the results (1, 6, 17, 23, 27, 30, 37, 39). According to these, the results of surgical treatment seem to be unsatisfactory, with a high mortality – about 30% on average. On the other hand, follow-up results remain a less clear point, since in most of the reported series these are either not given or they refer to a rather short period of observation.

In this paper we consider the long-term results of the 27 operated cases, out of the 28 choroid plexus papillomas observed in the Institute of Neurosurgery of the Rome University School of Medicine during the period 1952–1978. Most of the operations were performed by the senior author (B. G.).

The first 17 cases (1952–1968) have already been reported in a previous paper (26). The aim of the present study is to compare the results of surgical treatment in this first series with those of the more recent cases, usually operated on with the aid of the operating microscope and microsurgical techniques.

Material and method

In the above-mentioned period 28 patients with histologically proven choroid plexus papillomas were admitted to our Institute. Histological specimens were all reviewed by our team of pathologists and reclassified if necessary. These cases account for 0.65% of the 4299 brain tumours observed during the same interval.

The sex and age of the patients at the time of admission, as well as the location of the tumours, are summarized in Table 1. Clinical and diagnostic features are outside the scope of this paper.

Our material was divided into two series, according to when the operation was performed. In the first series (1952–1968) there are 17 cases. These have been previously analysed as regards their clinical diagnostic and surgical features (26). The second series (1969–1978) consists of 12 cases. One case (case 14) is included in both series because two operations were performed, the first in 1966 and the second in 1977 for a recurrence. The second operation was performed with the aid of the operating microscope, whereas the first was not.

Follow-up data were obtained from all the patients who survived the operation, and the results were classified as follows:

a) Excellent – the patient is free from any neurological deficit.

b) Good – a mild neurological deficit is present, but does not prevent the patient leading a normal life.

c) Fair – there is a significant neurological deficit, epileptic sequelae and/or mental retardation, but otherwise the patient is able to look after himself.

d) Poor – the patient is unable to look after himself.

e) Death.

We also considered as late tumoral deaths – or deaths from recurrence – those which occured after operation for a recurrent papilloma.

The patients operated on more recently were usually checked by CT scan examination, in order to confirm the clinical impressions and to offset the shortness of the follow-up observation period.

Surgical technique

Our policy in each case was to attempt a radical removal, whenever possible. This was achieved in 14

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Tab. 1. Sex and age of patients at the admission, location of tumours, follow-up period and results.

Case 1	M 40	R lat. ven. (malignant)	4 months	Death from unknown cause
Case 2	F 37	R lat. ven.	27 years	Excellent
Case 3	M 39	L lat. ven.	26 years	Excellent
Case 4	M 50	R lat. ven.	19 years	Death from M. I. (excellent)
Case 5	M 35	IV ven.		Postoperative death
Case 6	F 12	R lat. ven.	20 years	Excellent
Case 7	M 36	L lat. ven. (+ colloid cyst)		Postoperative death
Case 8	F 1½	L lat. ven.	17 years	Good
Case 9	F 41	IV ven.		Postoperative death
Case 10	M 48	IV ven.		Preop. death following ventriculography
Case 11	M 13	IV ven.	14 years	Excellent
Case 12	F 3½	IV ven.	14 years	Excellent
Case 13	M 53	IV ven. (malignant)	9 months	Death from recurrence
Case 14	M 35	IV ven.	11 years	Late non-fatal recurrence
			2 years	Excellent (after reoperation)
Case 15	M 36	C. P. A.		Postoperative death
Case 16	M 4	L lat. ven.		Postoperative death
Case 17	M ½	R lat. ven.	10 years	Fair
Case 18	M 33	IV ven.	10 years	Excellent
Case 19	M 1	R lat. ven.		Postoperative death
Case 20	F 17	IV ven.	9 years	Excellent
Case 21	F 16	R lat. ven.	9 years	Excellent
Case 22	F 19	IV ven.	8 years	Good
Case 23	M 2	L lat. ven. (+ vermian cyst)	8 years	Fair
Case 24	F 15	IV ven.	8 years	Excellent
Case 25	F 38	IV ven. (malignant)	7 years	Excellent
Case 26	F 7	IV ven.	4 years	Excellent
Case 27	F 3	III ven.	3 years	Fair
Case 28	M 56	IV ven. (malignant?)		Postoperative death

L	left
R	right
M. I.	myocardial infarction
lat. ven.	lateral ventricle
III ven.	third ventricle
IV ven.	fourth ventricle
C.P.A.	cerebellopontine angle

cases of the first series (87%) and in 11 of the second (92%). In the remaining cases an extensive subtotal removal of the infiltrating mass was performed.

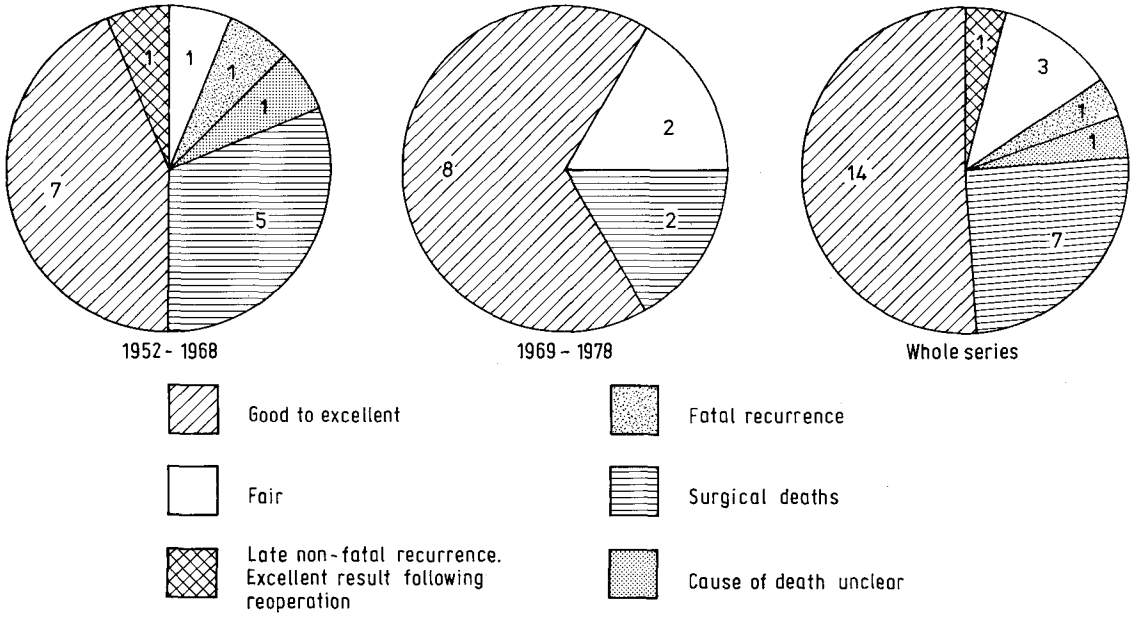
Neither ventricular shunting nor radiotherapy were used preoperatively in these cases. As a rule, an external ventricular drain was employed during the first two days after the operation.

Lateral ventricle papillomas were exposed in the conventional manner, through a reasonably small middle temporal (20, 22, 23, 30) or posterior parietal (20, 21, 23) corticotomy. Tumours in the fourth ventricle were approached by splitting the vermis. The case of cerebellopontine angle papilloma required a paramedian retromastoid incision, whilst the third ventricle papilloma was approached through the right lateral ventricle via a right frontal parasagittal bone flap (13). As mentioned above, the operating microscope and microtechniques were routinely used in the cases of the second series (1969–1979).

The most important step in the operation was the identification and ligation of the feeding vessels. After this, it was usually possible to remove the collapsed tumour, often "en bloc". During the whole operation great care was taken to isolate the tumour from the rest of the operative field by means of cottonoid packs, in order to minimize the risk of seeding of the tumour.

Postoperative care of these patients included great attention to the vital functions and to metabolic balance. Hyperthermia occurred after operation in most cases. It usually subsided quite soon afterwards. Gastrointestinal haemorrhage occurred in four cases. In case 5 this was the probable cause of death.

Tab. 2.



Results

First series (1952–1968)

One of the 17 cases died soon after a ventriculography. This case is obviously excluded from the analysis of surgical results. Seventeen operations were performed on the remaining 16 patients.

One patient underwent two operations. He showed a definite improvement after removal of a malignant papilloma infiltrating the floor of the fourth ventricle and the left tonsil. Post-operative radiotherapy was not given.

The patient was operated on for a recurrence eight months later. A few days after the second operation paraparesis and signs of spinal subarachnoid block supervened. These were thought to indicate spinal tumoral seeding. The patient's condition gradually worsened and he died one month later. Autopsy was not permitted.

As regards the other 15 patients, a total macroscopical removal was accomplished in all but one case (case 7), in which a left ventricle papilloma was incidentally found during an operation for a colloid cyst of the third ventricle. Troublesome bleeding complicated removal of the papilloma, which was not radical. The patient died in the early postoperative phase.

In case 1 a choroid carcinoma was removed from the right lateral ventricle. The operation was thought to be radical, and symptoms rapidly improved. The patient died four months later. No information could be obtained concerning the cause of death.

Among the remaining cases there were four early postoperative deaths. One patient – case 5 – died from massive gastrointestinal haemorrhage six days after successful removal of a fourth ventricle papilloma. In the other four cases (7, 9, 15 and 16), death was probably related directly to the surgical operation. In these patients, one tumour was located in the lateral ventricle, one in the cerebellopontine angle, and the remaining two cases were in the fourth ventricle. In case 14, mentioned above, the tumour recurred 11 years after the first operation.

Case 17 had a total removal of a huge right lateral ventricle papilloma. There was a stormy postoperative course, complicated by meningeal infection. Later the patient underwent a V-P shunt, after which he slowly but progressively improved. He had several episodes of epileptic fits, which completely subsided within five years of the operation. Anticonvulsant therapy was discontinued four years ago, without any further recurrence of epilepsy. At present this patient has a minimal left-sided pyramidal weakness, and is mentally retarded.

The remaining 7 patients are, in all but two cases, in excellent health, (Table 1), respectively 27, 26, 20, 14, and 14 years after the operation. Tumours were located in the lateral ventricles in three cases and in the fourth ventricle in two cases. Case 4 died from

myocardial infarction 19 years after successful removal of a right ventricle papilloma, followed by prompt regression of symptoms. Case 8, operated on 17 years ago for a right lateral ventricle papilloma, suffers from a very mild left hemiparesis, which does not prevent her from leading an active life. Her weakness was in fact worse prior to operation.

Second series (1969–1978)

We performed 12 operations in 11 new cases and one recurrence after 10 years.

A total removal was accomplished in all but one case. In this latter (case 28), the surgeon, an experienced member of the senior staff, reported that the tumour had infiltrated the floor of the fourth ventricle. He performed a gross subtotal removal of the mass, which looked macroscopically like an ependymoma. After an apparently uneventful operation the patient had a cardiac arrest when the incision was being closed. Resuscitation was attempted without success. Autopsy was not permitted. Histological examination of the surgical specimen revealed a choroid plexus papilloma without malignant features, with the possible exception of hyperchromatic nuclei. However, since the infiltrating portion of the tumour could not be examined histologically, the possibility of this showing definite malignant changes could not be excluded.

Among the other ten cases in which total removal was performed, one postoperative death occurred. This patient was an 11-months-old boy (case 19), who was admitted in very poor condition from another medical centre. He was operated on as he became decerebrate. During the operation a 7 × 6 cm. right lateral ventricle papilloma was uneventfully removed. However, the patient did not regain consciousness and died soon thereafter. The other nine patients all showed clinical improvement following the operation.

Case 23 was operated at the age of nine months for a huge midline cerebellar cyst. Sixteen months later he was readmitted with disturbances of gait and signs of increased ICP. Neuroradiological investigations surprisingly revealed a left intraventricular tumour. The previous ventriculography was carefully reviewed, but it failed to show any sign of this tumour. At operation this proved to be a left lateral ventricle papilloma, and it was totally removed. After this he had scattered episodes of epileptic fits. He was readmitted one and a half years later for hydrocephalus. After a V–A shunt he definitely improved. At a follow-up examination eight years after removal of the papilloma, he showed mild pyramidal weakness in the right leg and reduced visual acuity in the right eye. He was slightly mentally retarded; however, he was attending normal schools. He has not suffered from any fits during the last six years, although anticonvulsant therapy was discontinued four years ago. The CT scan showed only the signs of the previous operations.

Case 22 was readmitted in 1977, eight years after removal of a fourth ventricle papilloma, on account of an epileptic fit. The CT scan showed no recurrence of tumour. The patient is now asymptomatic except for a right amaurosis, which was already present before the operation.

Case 27, reported in detail in a previous paper (13), was readmitted for hydrocephalus six months after operation. After a V-P shunt the patient definitely improved. At a follow-up examination three years after the operation the patient showed mental retardation and signs of increased ICP. The valve was not functioning satisfactorily. A CT scan revealed a fourth ventricle tumour, possibly a papilloma, but there was no recurrence of the previously removed third ventricle papilloma. We are of the opinion that this was the result of tumoral seeding, although cases of multiple supra- and subtentorial occurrence of choroid plexus papilloma have been recorded (39, 41). The patient's parents emphatically refused permission to operate.

Radiotherapy was not used except in the case of one patient. In case 25 a fourth ventricle papilloma was found to have infiltrated the vermis. However, after a very careful and gentle dissection of the adhesions of the tumour to the floor of the fourth ventricle, removal was thought to be complete. On account of the atypical histological appearance, radiation treatment was recommended and a dose of 4500 R was given. The patient is now asymptomatic, seven years after operation. A CT scan, performed elsewhere, confirmed the absence of recurrence.

All the remaining patients are now asymptomatic, respectively 10, 9½ , 9, 8 and 4 years after the operation. In all but one case, a fourth ventricle papilloma was removed. The remaining patient had a papilloma in the right lateral ventricle.

Overall results

Table 2 summarizes the results for the entire series of 27 operated cases.

The series included nine children – i. e. below 12 years of age, as defined by Matson and Crofton (23) – and eighteen adults. Tumours were located in the lateral ventricles in 12 cases (44%), in the third ventricle in one case (4%), in the fourth ventricle in

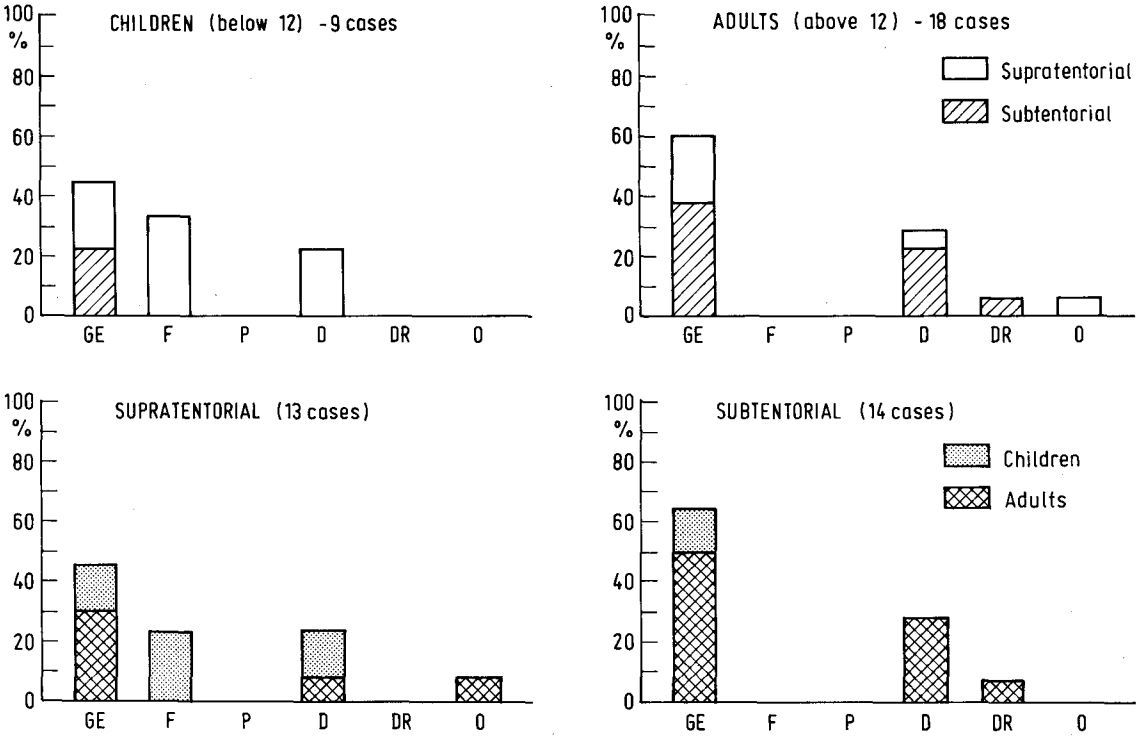
13 cases (48%), in the cerebellopontine angle in one case (4%).
The results of operation in children and in adults, as well as in supratentorial and in subtentorial cases, are compared in Table 3.

Discussion

The incidence of these tumours in relation to other brain tumours in the present series was similar to that reported in the literature (1, 6, 22, 27, 36, 41). There was a predominance of adult patients – 50% were above 30 – and a slight excess of males – 16 as against 12. Other authors (9, 23, 27, 29, 34) have found choroid plexus papillomas to occur predominantly in childhood.
In all but two of our cases, tumours were located in the fourth ventricle (50%) and in the lateral ventricles (43%). The remaining two cases (7%) were located in the third ventricle and in the cerebellopontine angle respectively. We did not find the higher incidence of left ventricle tumours reported by others (16, 28, 29, 38) amongst the choroid plexus papillomas situated in the lateral ventricles.

In all but three cases, the tumours proved to be histologically benign. In another case, as is explained above, histological malignancy could not be ruled out, on account of the incompleteness of the examination. Moreover, the difficulty of classifying case 1 as a choroid plexus carcinoma has already been clearly described in a previous paper (26). Indeed the presence of a small pulmonary carcinoma as primary lesion can never be excluded except on the basis of clinical and/or autopsy results (10).
This patient died four months after operation, after having shown good clinical improvement in the first weeks. Such a short interval between operation and death from recurrence was observed in other cases of choroid plexus carcinoma, in which, however, a total tumour removal had not been performed (22, 23). In any case the lack of details about the postoperative clinical course does not allow any conclusions to be drawn.
Generally speaking, histological malignancy corresponded to a fair degree with the infiltrating character of the mass and with tumour recurrences. On the other hand, tumour seeding in the present series did not seem to show a similar correlation with histological characteristics. This is well in keeping with the

Tab. 3.



findings indicated in literature (3, 12, 15, 18, 19, 32, 33, 38, 41, 42). The sole recurrence observed in the benign cases can, in our opinion, be explained by an unrecognized technical failure. The fact that in this case the tumour recurred 11 years after operation is remarkable. In view of this we would again stress the necessity for long-term follow-up in all surgically treated choroid plexus papilloma cases. CT scan examination should be helpful in checking the patients at a short interval after the operation.

In tables 4 and 5 we have summarized data collected from the largest surgical series reported in the literature. Because of the considerable difference in tumour location in children and in adults, and to the possible prognostic influence of this factor, we considered it advisable to analyse the results relative to patients of all ages separately from those regarding children only. The location of tumours indicated in the quoted series thus proved to be very similar to that found in our series.

Table 4, which illustrates the results for the series including all age-groups, indicates that the number of totally removed tumours is usually far from being satisfactory, and that the long-term results, when given, are not particularly encouraging. It appears reasonable, in our opinion, to admit a correlation between these two facts.

Surgical mortality in the other reported series ranged from 18% to 40%, with an average of 29%, while late deaths related to tumours occurred in 10% to 35% of cases, with an average incidence of 16%. Early mortality in the whole present series was 26%. Our surgical policy was rather aggressive. This could perhaps explain the low incidence – 4% – of late tumour deaths. We obtained 55% good-to-excellent long-term outcomes in our operated cases, which were followed up from 2 to 27 years. We did not find any indication in the literature that more conservative therapeutic approaches, such as radiotherapy and/or shunting procedures combined with either partial or subtotal surgery, could provide equally good long-term results.

When comparing the results of our earlier cases with those of our more recent series, we found in the latter an impressive decrease in mortality (16% as against 31%) a lack of tumour recurrences, ascertained by CT scan in most cases, and a higher rate of long-term good results (67% as against 44%), whereas the difference was less striking in the rates of total tumour removal (92% against 87%). A fact to be considered is the higher incidence in the second series of subtentorial cases, which can reasonably be assumed to present a higher operative risk. In view of this, the drop in the surgical death rate is in our opinion even more significant. We consider this a clear improvement, due principally to the use of the operating microscope and microtechniques, in addition to advances in anaesthesia and postoperative care of patients.

Nine of the patients in the present series (33%) were operated below the age of 12 years. The surgical data on these cases are given in Table 5, together with that relative to other large series of papilloma cases occurring in the same age group, collected from the literature. Mortality is usually lower in children than in adults, as confirmed in our series (Table 3), probably due to the lower incidence of subtentorial papillomas.

In the children, in addition to decreased mortality, we observed a lower incidence of late good results. Indeed it can be reasonably assumed that transcortical approach to a deep-seated tumour carries a not

Tab. 4. Results of operation in the largest series of choroid plexus papillomas reported in the literature (patients of all ages)

Authors	n° of operated cases	Lat. vent. tumours	III vent. tumours	IV vent. and CPA tumours	Total removal	Postop. deaths	Deaths from * recurrence	Late good results
Bohm and Strang 1961	25	8	2	15	56%(14)	40% (10)	8% (2)?**	?
Terracciano and Troisi 1961	10	4	0	6	60% (6)	30% (3)	10% (1)	40% (4)*****
Wilkins and Rutledge 1961	19	8 (+1***)	0	10 (+1***)	63% (12)	26% (5)	16% (3)	31% (6)
Nassar and Mount 1968	17	3	5	9	59% (10)	18% (3)	12% (2)	41% (7)
Arseni et al. 1974	38	18	3	17	52% (20)	34% (13)	?	?
Gullotta and Melo 1979	17	11	1	5	?	18% (3)	35% (6)	23% (4)*****
Whole present series	27	12	1	15	89% (24)	26% (7)	4% (1)	55% (15)
Present series 1952–1968	16	9	0	8****	87% (14)	31% (5)	6% (1)	44% (7)
Present series 1969–1978	12	3	1	8****	92% (11)	16% (2)	0	67% (8)

* Referred to all late deaths related to tumour (included fatalities due to operations for recurrent tumour).

** Not clearly reported.

*** Multiple right lateral ventricle and IV ventricle papilloma.

**** Case 14 is considered in both series (see text).

***** Follow-up short in the vast majority of cases.

Tab. 5. Results of surgical treatment of choroid plexus papillomas in children.

Authors	n° of operated cases	Lat. vent. tumours	III vent. tumours	IV vent. and CPA tumours	Total removal	Postop. deaths	Deaths from * recurrence	Late good results
Matson and Crofton (below 12) 1960	15	14	0	1	73% (11)	20% (3)	7% (1)	40% (6)
Raimondi and Gutierrez* (age unspecified) 1975	22	? the vast majority	0	?	?	0	5% (1)	?
Present series (below 12)	9	6	1	2	100%	22% (2)	0	44% (4)

* In this paper follow-up data are not reported.

inconsiderable risk of functional impairment in terms of epileptic sequelae, sensory disturbances and/or pyramidal weakness. Gentle and careful surgery could in our opinion help to lessen this risk. Unfortunately the long-term functional results are not always given clearly in the other series reported.

Long-term epileptic sequelae occurred in two children (22%), whereas none were reported in adult cases, except as isolated episodes. Both these patients, together with another child included in our series, required shunting procedures in the late postoperative course. It is beyond the scope of this paper to discuss the pathophysiological mechanisms of hydrocephalus in choroid plexus papilloma, especially as several recent publications have dealt exhaustively with this subject (11, 14, 16, 24, 25). However, we would like to note that all these patients who subsequently required shunting had a stormy clinical and/or postoperative course. It should perhaps be postulated that the observed complications, such as meningitis and tumour seeding, play some role in the persistence and/or development of hydrocephalus after surgery in some of these cases.

Raimondi and Gutierrez (30) have stressed the routine use of preoperative shunting. Their results are impressively good, at least as regards surgical mortality. Although we lack direct experience of such a procedure in the preoperative treatment of choroid plexus papilloma, we feel that surgical mortality in our cases was not strictly related to the degree of preoperative hydrocephalus. As pointed out above, more gentle and careful surgery ensured "per se" an encouraging drop in the mortality rate in our more recent cases, in spite of the less favourable location of tumours.

In the light of our experience the value of radiation therapy in these neoplasms also remains questionable. The general opinion is that this should be confined to cases showing features of malignancy (1, 6, 9, 22, 23, 27, 40). Accordingly, we recommended radiation treatment in only one case of the second series, in which a CT scan confirmed the absence of recurrence seven years after operation. One histologically similar case in the first series, which was not irradiated, recurred early after operation. However, the operation was thought to have been radical in the former, but subtotal in the latter. This limited experience does not allow any conclusion to be drawn.

The usefulness of preoperative irradiation in decreasing the vascularity and the size of papilloma has been stressed (2, 5, 38), also in recent literature (7). This would seem to facilitate operation. However, it was reported that it could cause adhesions of the tumour to the surrounding structures, sometimes making total removal impossible (7). It is our impression that, providid ligation of the vascular pedicle is correctly performed, considerable surgical difficulty due to the tumours vascularity is never encountered. In view of this, and of the well-known risks of long-term irradiation sequelae in benign lesions (4, 35), we would not recommend this treatment in any case.

In conclusion, we feel that the treatment of choroid plexus papilloma should be as aggressive as possible, and that encouraging long-term results can be expected when a radical operation is performed. As indicated by our experience in more recent cases, this is almost always possible, provided that modern technical advances are properly employed.

Summary

This paper reports the results of 27 choroid plexus papillomas surgically treated, out of 28 cases observed in the authors insitute during the period 1952–1978. These were divided into two groups: a) those operated on before 1969; b) those operated on in 1969 and the following years, when microsurgical facilities were routinely employed. As a rule, neither ventricular shunting nor radiation therapy

were used preoperatively. A total removal was attempted in all cases, and performed in all but three. The second group (1969–1978) showed better results as regards the number of totally removed tumours (92% against 87%), surgical deaths (16% against 31%), and long-term good results (67% against 44%). Considering both groups together, long-term good results were achieved in 15 patients (55%), followed up from 2 to 27 years. Radical operation is the treatment of choice for choroid plexus papilloma. As indicated by the author's experience, this produces satisfactory long-term results, whilst surgical mortality can be held within acceptable limits, provided that modern techniques are used properly. The value of radiation therapy, which in any event should be restricted to malignant cases, remains questionable.

Key words:
Choroid plexus papilloma – Surgical treatment – Surgical results.

Zusammenfassung

In der Arbeit wird über das operative Behandlungsergebnis von 27 Plexuspapillomen aus einer Serie von 28 Fällen berichtet, die zwischen 1952 und 1978 im Neurochirurgischen Institut der Universität Rom behandelt wurden. Eine Gliederung erfolgte in 2 Gruppen: a) vor 1969 und b) nach 1969 mit routinemäßiger Durchführung der Mikrochirurgie.
Ventrikuläre Shunt-Operationen und Röntgentherapie wurden praeoperativ nicht vorgenommen. Eine Totalentfernung des Tumors wurde stets angestrebt und mit Ausnahme von 3 Fällen durchgeführt.
In der mikrochirurgisch operierten Gruppe (1969–1978) war das Ergebnis deutlich besser. 92% wurden total extirpiert gegenüber 87% in der Gruppe a). Es verstarben postoperativ 16% gegenüber 31%. Die Spätergebnisse waren mit 67% besser als in der ersten Gruppe mit 44%. Insgesamt hatten 14 Kranke (55%) ein gutes Spätresultat mit Nachuntersuchungen nach 2 bis 27 Jahren.
Die radikale Entfernung ist die Methode der Wahl in der Behandlung der Plexuspapillome. Die eigenen Erfahrungen zeigen, daß sie zu zufriedenstellendem Spätergebnis bei akzeptabler Operationsmortalität führt. Voraussetzung ist die Beherrschung der modernen Operationstechnik. Die Bedeutung der Radio-Therapie bleibt fraglich. Sie sollte ausschließlich auf maligne Fälle beschränkt bleiben.

Schlüsselwörter:
Chorioid-Plexuspapillome – Operative Behandlung – Operative Ergebnisse

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